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Part I

Pelvic Tumors: The Fundamentals

1 Pelvis: General Considerations

Peter F. M. Choong

Resections of tumours of the bony pelvis are highly morbid and can challenge even the most expert of surgeons [1–5]. Decisions regarding the surgery and the ultimate outcomes of procedures are determined by the type and location of the tumour (bone or soft tissue) [4, 6–8], the operative approach [9], the reconstructive techniques utilised [1, 10–13] and the amount and type of tissue sacrificed in the surgery. The morbidity of pelvic reconstruction is high, and the intraoperative demand on expertise, resources and personnel is such that pelvic tumour surgery is best practised in a multidisciplinary team setting with members who are familiar and expert in the intra- and perioperative care of such patients [9].

1.1 Important Considerations When Planning Treatment of Pelvic Tumours

- Understanding the aetiology of the tumour (benign, malignant, primary, metastatic) will allow engagement of the relevant clinical experts, determination of oncologic surgical margins and planning of durable reconstructions.
- Comprehensive pathologic, local and systemic staging is mandatory and part of the treatment strategy of these complex tumours.
- Classification systems help to define the location of the tumour and the types of resections and reconstructions that may be required.
- A careful study of the anatomy of the pelvis in relation to the tumour and its planned resection allows anticipation and mitigation of intraoperative hazards.
- Careful planning of the surgical approach together with other specialist surgeons is critical for ensuring optimal patient positioning and draping, for achieving the best view of the operative field and vital structures, for anticipating the order of surgery when multiple specialties are involved and for facilitating the use of specialised equipment if required.
- Pelvic surgery often requires prolonged surgery that is frequently associated with episodes of haemodynamic and respiratory instability. This requires an expert anaesthetic team capable of managing rapid transfusion requirements and invasive monitoring. Managing the physiologic upset during the procedure is an important consideration and requires a close working relationship with the anaesthetic team.

1.2 Aetiology of Pelvic Tumours

1.2.1 Primary Tumours

Ten percent of primary tumours involve the pelvis, and of these, chondrosarcoma, Ewing's sarcoma and osteosarcoma are the commonest [14]. These may be treated with curative or palliative intent and, other than chondrosarcoma, will require adjuvant multimodal treatment. In either clinical situation (curative, palliative), local control of disease to minimise or negate tumour recurrence is the prime goal of surgery. If vital structures are not at risk, then wide surgical margins are indicated and may be defined as at least 2 cm of clear bone in the line of the bone and a cuff of normal tissue which is a named anatomic layer such as muscle or fascia that is parallel to the surface of the tumour [15]. Some authors have highlighted the importance of the quality of the surgical margin and that this may vary between tissues that comprise the margin [16]. In planned resections, which involve adjuvant treatment, surgery is often preceded by neoadjuvant chemotherapy or radiotherapy. Chemotherapy and radiotherapy aim to kill the tumour, reduce its size and

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incite a fibrotic reaction around the tumour to create a true capsule which effectively enhances the surgical margin. The extent of surgery including the resection of bone and soft tissue often leaves the operative bed associated with a substantial dead space. Dead space is a potential source of complications including haematoma, infection and wound dehiscence, which may demand pre-emptive or corrective surgery. Early involvement of plastic and reconstructive surgical expertise in the surgical planning will optimise surgical outcomes [17–19].

1.2.2 Secondary Tumours

Metastatic bone disease following carcinoma is a common occurrence [20, 21]. Common primary sites of carcinoma which metastasise to the pelvis include breast, lung, prostate, kidney and thyroid. Breast and prostate carcinoma metastases often present with mixed sclerotic-lytic disease. Lung carcinoma metastases often present with permeative and poorly circumscribed lesions. Thyroid and renal carcinoma metastasise with cannonball lesions that are often hypervascular and markedly lytic and may grow to considerable sizes. Metastases may also be associated with large soft tissue components, which present a similar complexity as large primary tumours by obscuring or impinging on vital structures.

The management of primary bone malignancies differs from metastatic bone disease because primary tumours are often solitary, are amenable to wide surgical margins and are resected with as large a cuff or margin of normal tissue as is possible. In contrast, the surgical management of metastatic disease, which is diffuse, progressive and multifocal, aims for conservative bone-preserving techniques that are durable and appropriate for what is often treatment with a palliative intent [20, 22]. For the latter, curettage or the removal of only macroscopically affected tissue is combined with reconstruction that is reinforced and not primarily dependent on bone union or ingrowth. Rarely, truly solitary metastases may be treated with wide resection like a primary tumour. More commonly, however, apparently solitary metastases are associated with micrometastatic lesions within the same bone, which eventually manifest their presence in the passage of time. Careful scrutiny of well-performed anatomic and functional imaging investigations should be undertaken before a decision is made to designate a tumour as solitary. Progression of metastatic disease is the norm, and to avoid failure of the device from rapid recurrence of tumour, reconstructions should be planned to achieve maximum durability within the anticipated lifespan of the patient [20, 22]. For example, internal fixation often reinforced by acrylic bone cement should span entire lengths of bones. Locking screws should be used and anchored in bone cement to enhance fixation and strength of the construct. If joint prostheses are to be used, then consideration should be given for cement fixation rather than cementless fixation, and for long-stemmed prostheses rather than standard-length prostheses for the femur, humerus and tibia for added protection.

1.3 Symptoms of Pelvic Tumours

Because of the large intra-abdominal and intra-pelvic volume, primary pelvic tumours may have an occult presentation, reaching large sizes before detection. Symptoms may vary between bone and soft tissue sarcomas with the former being associated with deep-seated and nocturnal pain, while the latter often presenting without pain. Abdominal fullness may be a feature of the latter's presentation. Irritation of the bowel and bladder or compression of the ureter may lead to obstructive symptoms, frequency or dissatisfied visceral evacuation. Often, symptoms are vague and misinterpreted for musculoskeletal injury or referred pain from the lumbar spine.

1.4 Imaging Modalities

Appropriate and adequate imaging is mandatory for surgery about the pelvis [23–25]. When dealing with tumours, both anatomic and functional imaging can be very useful for characterising the tumour and informing the planning of biopsy and surgical margins. Anatomic imaging includes plain radiography, computed tomography (CT) and magnetic resonance imaging (MRI). Functional imaging is used to examine the metabolic activity of tumours and include technetium bone scans, thallium scans and positron emission tomography. The results of functional scans can be superimposed (co-registered) onto CT scans to provide the exact location of metabolic activity in relation to the patient's anatomy.

1.4.1 Plain Radiography

Biplane and Judet views of the pelvis are simple and reliable tests that allow assessment of the bony architecture of the pelvis. These are particularly important not only for characterising the primary tumour but also for delineating bone destruction associated with metastatic disease. Digital imaging is now available in most centres, and standardised views with appropriate scaling allow more accurate templating and planning for acetabular or proximal femoral prostheses, as well as for selecting size-matched allografts.

1.4.2 Computed Tomograph

Fine-slice CT is an excellent modality for assessing the quality of bone. Not only is cortical bone very well delineated with CT, the high-resolution images available today make characterisation of subtle trabecular destruction much easier than in the past. CT scanning because of the contrast between tumour and fat easily demonstrates marrow replacement by tumour, and this may be particularly useful when investigating a potential tumour-related pathologic fracture. CT scanning can provide excellent multiplanar views from which three-dimensional images may be constructed. Computer algorithms (bone windows) that are able to suppress metal artefact allow this modality to interrogate prosthetic-bone interfaces. With more sophisticated computer software, whole prostheses within individual images may be suppressed to provide better visualisation of periprosthetic bone.

1.4.3 Magnetic Resonance Imaging

Magnetic resonance imaging is unsurpassed as a modality for imaging soft tissue. MRI should be a mandatory investigation for assessing all primary tumours of the pelvis because it combines excellent three-dimensional bone imaging with excellent delineation of organs, vessels and nerves to give a very accurate assessment of the relationship of pelvic tumour to the pelvic viscera. Moreover, MRI exploits the high water content of fat to accurately characterise the intraosseous extent of tumour.

1.4.4 Angiography

Angiography may be required to determine if vessels are compromised by a tumour and therefore need to be sacrificed, or if important feeder vessels exist and need to be embolised or ligated as part of tumour resection. Embolisation may need to be considered to avoid intraoperative haemorrhage when dealing with certain metastatic carcinomas arising from the kidney, thyroid or myeloma. Some primary tumours occur in highly vascular areas (sacrum), and embolisation may need to be performed to avoid troublesome intra-operative and post-operative bleeding. Preoperative embolisation using radiopaque coils, beads or gelfoam should be considered within 36 h before surgery to minimise the return of flow to the embolised lesion. Although traditional contrast angiography provides highly accurate images, non-invasive methods such as CT or MR angiograms may also be performed, although smaller vessels may not be as easily visualised.

1.4.5 Functional Imaging

This modality is particularly useful for metastatic screening or for determining the metabolic activity of tumours. Identifying multiple intraosseuos or soft tissue lesions has a profound impact on prognosis and will have important implications on the choice of treatment strategies. The extent of metabolic activity may also reflect the grade of the tumour. High-grade tumours are associated with high metabolic activity as compared to low-grade tumours. Of note, large malignant tumours are often associated with central tumour necrosis, and this should not be misinterpreted for low tumour grade. Biopsies are most representative when tissue of the highest grade and greatest viability is obtained. Functional imaging is excellent for identifying regions which may be targeted for biopsy. Post-treatment activity is also a measure of tumour response, and this may have important implications when planning surgical margins. Primary bone tumours with a good response to chemotherapy but close margins have a five times higher risk of local recurrence than tumours with a good chemotherapy response and good margins [26]. Tumours with a poor response and poor margins have a 50 times higher risk of local recurrence [26]. Knowing this information prior to surgery may help to discuss the pros and cons of amputation versus limb-sparing surgery.

1.5 Anatomic Considerations

The anatomy of the pelvis is complex. Imaging information from CT and MRI scans allows a good understanding of the relationship of the tumour to pelvic and intra-abdominal structures. Prominent tumours may distort anatomy, and structures most at risk include vessels, nerves and the ureters. Often small, these structures may be injured when dissecting around areas where the anatomy cannot be clearly defined. Prior to surgery, planning of surgical margins and the surgical approach should include a review of the pelvic anatomy, which should be orderly, beginning from the posterior pelvic ring and passing forward to the pubic symphysis.

1.6 Anaesthetic Considerations

Anaesthesia for pelvic surgery is challenging because of the extensive tissue trauma, the prolonged operative time, exposed bleeding bone and, in the case of malignancy, the impact of neoadjuvant therapies [27]. Post-operatively, patients face a number of major physiologic insults including ongoing blood and fluid loss, the effects of massive blood transfusion, ileus and pain. Typically, post-operative pain is severe, and multimodal anaesthetic techniques are required to provide safe and effective analgesia. While much has been written about pelvic resection and reconstruction, little is reported about the anaesthetic techniques employed in the surgery for these patients.

In well-planned surgery, preoperative anaesthetic assessment begins sometime prior to the scheduled surgery date. Ideally, the patient is seen by the anaesthetist, but patients from somewhere a great distance from the hospital may be contacted by telephone consultation.

While preoperative assessment includes a routine review of all systems, of particular importance is knowledge of location and pathology of the tumour and type and impact of preoperative adjuvant therapy. An understanding of the type, location and size of tumour helps the anaesthetist assess the possible extent of surgery required. For example, large tumours requiring extensive resection with close proximity to vascular structures will be more likely to have larger blood loss, greater tissue trauma and need for intensive post-operative support than those which are smaller and in a more favourable surgical position. Tumours that are situated posteriorly in the pelvis more often involve complicated and prolonged dissection around the lumbo-sacral plexus. The internal iliac vessels are more likely to be troublesome when tumours are situated posteriorly in the pelvis. Equally, anterior pelvic tumours that require dissection near the bladder neck are challenging because of the great tendency for the perivesical venous plexus to bleed heavily or continuously. Osteotomies expose bleeding bone and can provide a sustained source of haemorrhage. In anticipation of major blood loss during surgery, the hospital blood transfusion service should be notified of the date of surgery and the amount of blood and blood components that may be required. In most hemipelvectomy cases, ten units of packed red cells, five units of fresh frozen plasma and five units of platelets should be available at the commencement of surgery.

Preoperative chemotherapy or radiotherapy may have a deleterious effect on bone marrow function. Complete blood examinations are therefore required to ascertain if anaemia, profound leucopenia or thrombocytopenia exists. Consultation is made with the patient's oncologist to determine whether the haematological disturbances will correct themselves prior to surgery or whether further specific treatment is required. The hospital intensive care unit (ICU) is also notified at this stage of the date and type of surgery, as well as the possible duration of stay in ICU, so that resources can be allocated in advance.

1.7 Positioning and Pressure Care

As with any prolonged surgery, careful attention needs to be given to prevention of peripheral nerve compression. The patient should be positioned in the lateral position with the side supports holding the upper body at the sternum and mid-thoracic region well clear of the flank and abdomen. This allows the patient's body to move through an arc of 90°, from −45° to +45° when the operating table is rolled laterally from left to right side. The benefit of this position (floppy lateral) to the surgeon is that the rolling manoeuvre allows both anterior and posterior parts of the pelvis to be accessed. The potential hazards for the patient are that the points of potential nerve compression change each time the patient is moved as well as a risk of breathing circuit disconnection or dislodgement. Checking by the anaesthetist of areas at risk of compression neuropathy needs to be performed each time the patient is rolled from one side to the other.

1.8 Post-operative Analgesia

The method used for post-operative analgesia will be dependent on the technique chosen for anaesthesia. In our institution, a combination general and spinal anaesthetic and a post-operative epidural catheter with a continuous infusion of anaesthetic/opioid is preferred. The infusion is usually commenced in the intensive care unit or recovery room once the patient is haemodynamically stable. The infusion may be extended for up to 6 days. It is important that endotracheal intubation rather than laryngeal mask intubation is used because massive transfusion may cause subglottic and supraglottic oedema which is more safely managed using an endotracheal tube rather than a laryngeal mask. In the post-operative period, further bleeding can be expected. As an estimate, approximately one-third the volume of intraoperative fluid replacement is required in the first 24 h post-operatively.

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2 Anatomy and Physiology of the Pelvis

Robert Waldrop and Franklin H. Sim

2.1 Introduction

The origin of the word pelvis is Latin and refers to a basin which is appropriate as the pelvis not only acts as a link for the trunk to the mobile lower extremities but as rigid container for many important visceral structures. This chapter will aim to review the important anatomic structures, their relationships to one another, and the implications for these structures on surgery in and around the pelvis. If one is to perform surgical resection of the pelvis, it is imperative that he or she be familiar with the anatomy of the bony pelvis, the associated ligamentous structures, the surrounding musculature, the vasculature of the pelvis, the genitourinary structures, the terminal portions of the gastrointestinal tract, the neural structures in the pelvis, and the relationship of these structures to the fascia planes around the pelvis. When one begins to think of anatomy, it can be discussed from the order in which structures are encountered in a dissection or in relation of structures to one another. This chapter will try to incorporate elements of both approaches.

2.2 Osteology

The bony pelvis consists of the rigid ring formed by the junction of the sacrum, representing the caudal immobile portion of the spine, with the paired innominate bones.

2.2.1 Sacrum

The sacrum consists of five fused vertebrae that form a wedge-shaped bone that articulates with the paired innominate bones as well as the coccyx. While the coccyx does have attachments for some of the pelvic floor musculature, it will not be considered separate from the sacrum for the purpose of this text. The sacrum has ventral (sometimes referred to as the pelvic surface), dorsal, and lateral surfaces. The ventral surface of the pelvis has the observable sacral foramina with much of the rest of the surface being covered by periosteum, fascia, muscle attachments, and neurovascular structures. Emerging from the foramina are the ventral rami of the sacral nerve roots. These pass in a medial to lateral and cranial to caudal direction away from the midline on the anterior surface of the overlying piriformis muscles. The foramina also serve as a landmark because medial to the foramina and separate from the sacral nerve roots will lie the sympathetic trunks and median sacral vessels. The dorsal surface of the sacrum has the observable dorsal foramina which are continuous with the ventral foramina and create a perforation in the sacrum between the dorsum of the sacrum and the pelvic cavity. Like the ventral surface, the dorsum is covered by periosteum and muscle. The majority of the dorsal surface is covered by attachment of multifidus and erector spinae muscles. There is however much less in the way of neurovascular structures. The dorsal ramus of the individual sacral nerve roots exit the dorsal foramina to innervate the overlying myofascial structures and skin. The paired dorsal foramina flank the fused sacral lamina. Distal to the S4 foramina in the midline, one will encounter the sacral hiatus.

The lumbar sacral spine junction at the L5/S1 disc space and the associated sacral promontory serve as an important landmark in the posterior pelvis. This disc space can be used to localize the lumbar five transverse processes and thus both the traversing lumbar five nerve roots and iliolumbar ligaments.

2.2.2 Innominate Bones

The paired innominate bones begin as three separate structures joined by a cartilaginous center of ossification in youth that coalesce to form a single large osseous structure that

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gives much of the shape and support of the pelvis. The ilium forms the upper portion of the acetabulum and the bone more proximal and anterior. The ischium forms the posterior inferior portion of the acetabulum and the bone that extends in an inferior posterior direction. The pubis contributes the anterior inferior portion of the acetabulum and the bone which extends anteriorly to join the contralateral pubis at the pubic symphysis. The innominate bones serve as attachment for many muscles that overlie the pelvis, which provide both stability to upright posture and strength for locomotion. Particular areas of importance for the surgeon embarking on pelvic resection include the sciatic notch, pubic symphysis, acetabulum, and sacroiliac joint, as well as the relation of neurovascular and genitourinary structures.

2.2.2.1 Sacroiliac Joint

The sacroiliac joint is an irregularly shaped structure that allows for transmission of substantial forces from the axial skeleton to the lower extremities. Relatively little motion occurs at this joint as one ages. Over the course of life, the joint tends to become stiffer offering less motion. The bilateral sacroiliac joints are stabilized not only by the complex bony anatomy but also by the anterior sacroiliac, posterior sacroiliac, interosseous, iliolumbar, sacrospinous, and sacrotuberous ligaments. This ligamentous complex provides extremely strong fixation of the innominate bone to the sacrum, and close attention must be paid to these structures if one is to separate the innominate bone from the sacrum.

2.2.2.2 Sacrospinous Ligament

The sacrospinous ligament lies anterior to the sacrotuberous ligament attaching the anterior lateral aspect of the sacrum to the ischial spine. Often, the anterior portion of the sacrospinous ligament blends with the coccygeus muscle. This ligament is often a thin structure, but it is readily palpable in the posterior pelvis whether it is being approached anteriorly or posteriorly.

2.2.2.3 Sacrotuberous Ligament

The sacrotuberous ligament is a more robust but less distinct structure that lies more posterior than the sacrospinous ligament. The sacrotuberous ligament has a much broader attachment. It has attachment to posterior superior iliac spine, lateral sacral crest, lateral margin of the sacrum, and posterior sacroiliac ligament. Its fibers then run in a distal and lateral fashion to converge on the medial aspect of the ischial tuberosity. The bony anatomy of the ischium combines with the sacrospinous and sacrotuberous ligaments to create the greater and lesser sciatic notches which will be described in more detail in the discussion of the nervous and vascular structures.

2.2.2.4 Pubic Symphysis

The pubic symphysis represents the midline of the pelvis anteriorly where the two pubic bones converge at a cartilaginous joint surrounded by a thin capsule. The joint is reinforced by anterior and posterior pubic ligaments with the anterior being the more robust ligament.

2.3 Musculature

When studying muscle anatomy, one often groups individual muscles into functional groups. When one considers the musculature of the pelvis as it related to oncologic surgery, the divisions can be thought of in a slightly different way. It can be divided into two distinct groups: musculature related to and covering the inner surfaces of the pelvis and the musculature related to and covering the outer surface of the pelvis. While there is not a compartment in the sense it is referred to in the extremity, these muscles do aid in separation of neoplasm from pelvic viscera. The inner table of the ilium is nearly completely covered by the iliacus with contributions from the psoas major and minor. These muscles function as powerful hip flexors, but when considering oncologic resection, they also serve as a soft tissue boundary overlying the inner table of the ilium preventing or at least slowing the direct invasion of pelvic viscera by neoplasm arising from the bony pelvis. The lower portion of the inner pelvis is covered by the obturator internus medially and the coccygeus more distally. Deep to the obturator internus is the obturator fascia which also helps to create a barrier over the lateral sidewall of the inner pelvis. The piriformis takes its origin from the anterior surface of the sacrum around the ventral foramina, but it does not create a muscular barrier between the pelvic viscera and the bony pelvis.

The outer table of the ilium is largely covered by the abductor musculature. The gluteus minimus is located the most deeply, covering the distal portions of the outer table with the gluteus medius overlying and covering the more proximal outer table. The gluteus maximus is located superficial to the gluteus medius and completes the full covering of the outer table as its origin spans the iliac crest onto the posterior lateral surface of the sacrum. Often, there is an attachment of the gluteus medius to the undersurface of the large gluteus maximus. Overlying the gluteus maximus is the gluteal fascia which is quite robust anteriorly but tends to taper to a thin wispy layer posteriorly near the lateral border of the sacrum. With careful dissection, each of these layers can be taken down cleanly and maintained on their respective vascular pedicles for use in soft tissue coverage of the surgical wound following resection if not involved by neoplasm.

Abductor dissection from the outer table of the pelvis is largely determined by the location and extent of the tumor.

Henry and more recently Enneking have described the dissection of the gluteal fascia and the gluteus maximus to give excellent exposure of the underlying muscle and sciatic notch with the ability to repair the gluteus maximus. If the remaining abductors are free of neoplasm and their vascular pedicle is able to be saved, they can be repaired to remaining pelvic bone via bone tunnels or suture anchors.

2.4 Vascular Anatomy

There is considerable individual variation in the vascular anatomy of the pelvis, but there are general themes that prevail in most situations. This text will discuss the vascular anatomy beginning with the major retroperitoneal vessels just proximal to the division into bilateral iliac vessels through the terminal divisions in the lower extremity.

The abdominal aorta approaches the pelvis in the retroperitoneum along the anterior lateral aspect of the lumbar vertebral on the left side. It then divides into the bilateral common iliac arteries near the fourth lumbar vertebral body. The common iliac arteries continue on a path more distal and lateral to divide into the external and internal iliac arteries. This division occurs near the level of sacroiliac joint. The external iliac artery will continue on as the femoral artery as it passes under the inguinal ligament into the anterior thigh and serve as the principal blood supply to the lower extremity in combination with profunda femoris which divides from it in the proximal thigh. The internal iliac artery serves as the primary vascular supply to the pelvic viscera, the gluteal region, and the perineum. The course and divisions of the internal iliac artery are far more variable than the course of the common iliac or external iliac vessels. In addition to the named vessels that originate from the common iliac artery, there are numerous small unnamed vessels that supply viscera, the psoas muscle, and peritoneum. On some occasions, the renal artery can arise from the common iliac artery as can the iliolumbar artery.

The internal iliac artery divides into an anterior and posterior trunk as it passes into the true pelvis. The posterior trunk gives rise to the superior gluteal artery, lateral sacral artery, and iliolumbar artery, while the anterior trunk supplies the pelvic viscera as well as giving off the internal pudendal artery, obturator artery, and inferior gluteal artery. The first branch from the posterior trunk is usually the iliolumbar artery that ascends proximally to give branches to both the psoas and the iliacus before giving off branches that enter the spinal canal to supply the cauda equina. The lateral sacral artery can be present as a pair of separate arteries or as a single artery that will quickly divide into a superior and an inferior lateral sacral artery. These vessels supply the sacral vertebra and travel through the ventral foramina to supply the sacral canal before traversing the dorsal foramina to contribute to the vascular supply of the dorsal muscle and overlying skin. The superior gluteal artery is the largest branch of the posterior trunk. It can be thought of as the continuation of the posterior trunk. It exits the pelvis proximal to the piriformis. The obturator artery courses in an anterior and distal direction as it branches from the anterior trunk. It will then course between the deep surface of the obturator internus muscle and the bony pelvis until it exits the pelvis to supply the adductor compartment of the thigh. The internal pudendal artery and the larger inferior gluteal artery are the terminal branches of the anterior trunk. The internal pudendal artery lies anterior to the piriformis and the sacral plexus in the pelvis before exiting the pelvis between the piriformis and the coccygeus muscles. It then wraps around the ischial spine or sacrospinous ligament before entering the ischiorectal fossa. The inferior gluteal artery runs anterior to the piriformis but posterior to the internal pudendal artery in the pelvis before exiting the pelvis via the lesser sciatic foramen to be the major vascular supply of the abductors. The venous drainage of the pelvis is quite variable. The major veins tend to follow their named arterial counterparts, while the many smaller veins draining the sidewalls and viscera can have substantial variation. The internal iliac veins travel proximally, medial and posterior, to the internal iliac arteries and are formed by the convergence of a number of unpredictable vessels that serve to drain the regions supplied by the branches of the internal iliac artery.

2.5 Genitourinary System

In both sexes, the bladder and urethra are intimately related to the anterior pelvis. While a complete discussion of the anatomy of the genitourinary structures is beyond the scope of this text, it is important to have a firm understanding of the relationship between the anterior bony pelvis and the genitourinary structures. By its nature as a distensible reservoir, the bladder must have the ability to expand the volume it can retain. When the bladder is empty, it resides entirely within the true pelvis, but as it fills with urine, it expands and overflows the true pelvis into the false pelvis. The bladder is said to have four sides or surfaces. These are the superior, right and left inferolateral, and posterior. The bladder is relatively tethered in place at several points by structures such as the median umbilical ligament at the bladder apex, bilateral medial umbilical ligaments, and the attachment of the bladder neck to the fascia of the pelvic diaphragm as well by the puboprostatic ligament in males and the pubovesical ligament in females. For the surgeon attempting pelvic resection, the most important relationships are those of the bladder and urethra to the retropubic space and the course of the ureters which can readily be injured even when great care is taken in the dissection. The bladder is separated from the pubis and the rectus attachments by the retropubic space of Retzius. This space is akin to a bursa, being filled with fat that is well adhered to the bladder and a number of veins that can bleed profusely if one is unable to ligate them. It is important to ensure that the puboprostatic ligament in males or the pubovesical ligament in females has been taken down as it is possible to tear the bladder if this structure has not been ligated prior to specimen extraction. Care should be taken to ensure the bladder and the urethra are completely free from the specimen being removed as ligamentous attachments of the bladder can be variable. The paired ureters are small thick-walled muscular tubes that typically have a diameter of only a few millimeters and can easily be injured during pelvic dissection. The ureters originate at the renal pelvis and traverse distal to the pelvis in a retroperitoneal location near the medial surface of the psoas. The ureter then enters the true pelvis crossing anterior to either the distal portion of the common iliac artery or the proximal portion of the external iliac artery. Once in the pelvis, the ureter runs along the lateral sidewall anterior to the sciatic notch before turning medial around the level of the ischial spine to join the base of the bladder.

2.6 Large Bowel

The sigmoid colon and rectum are in close proximity when operating on the bony pelvis. While their exact anatomic position can vary from one individual to the next, there are general themes which are fairly consistent. The sigmoid colon serves as the segment of bowel connecting the descending colon to the rectum. It lies on the left side of the pelvis anterior to the external iliac vessels usually with a complete sheath of peritoneum covering it. In some individuals, it can be adhesed down to the parietal peritoneum that overlies the iliacus muscle. The sigmoid colon is described as continuing to the level third sacral vertebra where it is continuous with the rectum. The rectum then lies anterior to the distal sacrum until it passes through the puborectalis at the junction of the rectum and anus. The rectum itself is surrounded by a layer of fat, the mesorectum, which contains the blood supply as well as the venous and lymphatic drainage. The mesorectum is then surrounded by another distinct tissue layer, the mesorectal fascia, which also goes by the term fascia propria of the rectum in some texts. The mesorectal fascia is then separated from the presacral or Waldeyer's fascia by a retrorectal space.

2.7 Lumbar/Sacral Nerves

A working knowledge of the lumbar and sacral nerve roots and their general course is crucial to operating in and about the pelvis. The ventral rami of L1–L4 with possible contribution from T12 join to form the lumbar plexus within the psoas muscle ventral to the transverse processes of the respective vertebra and traveling in a dorsal to ventral course. The femoral nerve will then emerge on the lateral surface of the psoas muscle and eventually cross over the psoas. The obturator nerve also arises from the ventral rami of the lumbar nerve roots, most commonly L2–L4. The obturator nerve emerges from the medial side of the psoas and posterior to the common iliac artery and then travels lateral to the pelvic sidewall deep to obturator internus. The sciatic nerve originates from the L4 to S3 nerve roots and functions to control the lower extremity distal to the knee. The sciatic nerve exits the pelvis via the greater sciatic foramen and can have a variable relationship to the piriformis. In general, if one needs to free or locate the sciatic nerve, it is best to do so by taking down some of the gluteal sling approximately 40% of the way down the length of the femur and then working proximal. Nerves that supply only cutaneous sensation can be dissected and ligated with impunity, while nerves that provide motor function such as the sciatic, femoral, or obturator should be maintained unless tumor margin prohibits.

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3 Imaging Modalities and Differential Diagnosis

Matthew T. Houdek and Benjamin M. Howe

3.1 Introduction

The pelvis is a common site for primary and metastatic tumors. Although common, the diagnosis can be difficult. Patients with pelvic tumors often have a variable and nonspecific clinical presentation, with symptoms ranging from pain, fever, fatigue, bowel/bladder changes, weight loss, and lower extremity swelling. Due to the heterogeneity in the presentation, the diagnosis may be delayed until the tumors are either large enough to cause significant symptoms or pathologic fracture, or they are incidentally found on imaging for nonspecific symptoms such as back or hip pain. In the workup of a pelvic mass, plain radiographs, computed tomography (CT), magnetic resonance imaging (MRI), and positron emission tomography/computed tomography (PET/CT) are used to detect and characterize these lesions. The purpose of this chapter is to review the use of these imaging modalities and define the characteristics of some of the common lesions of the pelvis.

3.2 Imaging Modalities

3.2.1 Plain Radiographs

Plain radiographs are commonly the initial study in the evaluation of pelvic tumors. Large soft tissues masses may displace the bowel gas, obscure normal fat planes, or distort interfaces between muscle and the overlying fat allowing for detection on plain radiographs. Even very large soft tissue tumors can be challenging to detect on plain radiographs given the overlying bowel gas and soft tissues.

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The overlying soft tissues and bowel gas and complex osseous anatomy of the pelvis limit the detection of pelvic bone tumors on plain radiographs. Initial radiographs in unsuspected pelvic bone tumors are typically anterior-posterior projection, and the lack of orthogonal imaging limits the confident detection of pelvic bone tumors on the initial radiographs (Fig. 3.1). Likewise, the thick cancellous bone of the ilium allows bone tumors to be overlooked until large and destructive. This is in contrast to lesions of the pubis and the ischium, where the thin cancellous bone and lack of overlying structures make the lesions more conspicuous.

After detection of an osseous lesion on plain radiographs, an attempt should be made to categorize a lesion as having either nonaggressive or aggressive features. This distinction is more challenging in the pelvis compared to tumors of the extremities given the technical limitations of pelvic radiographs discussed above. The three categories of bone destruction are purely lytic, purely sclerotic, and mixed lytic and sclerotic.

The patterns of bone destruction and reaction of the host bone aid in the determination of aggressive and nonaggressive of lytic bone lesions. The margin formed between the lytic lesions and the adjacent host bone infers the aggressiveness of the tumor. In general, a narrow transition zone between the tumor and the host bone indicates a slow pattern of growth. A narrow zone of transition can be further subdivided into sclerotic and non-sclerotic. Aggressive tumors typically present with more indistinct margins (also referred to as a wide zone of transition) between the tumor and the adjacent host bone.

Periosteal reaction indicates the aggressiveness of both lytic and sclerotic bone tumors that involve or are adjacent to the cortex. The periosteal reaction type is an indicator of the host bone response and growth rate of bone lesions. Solid and unilamellar suggest an indolent growth pattern or reactive formation associated with adjacent inflammatory changes. Aggressive patterns of periosteal reaction include multi-lamellated, spiculated (also referred to as hair on end

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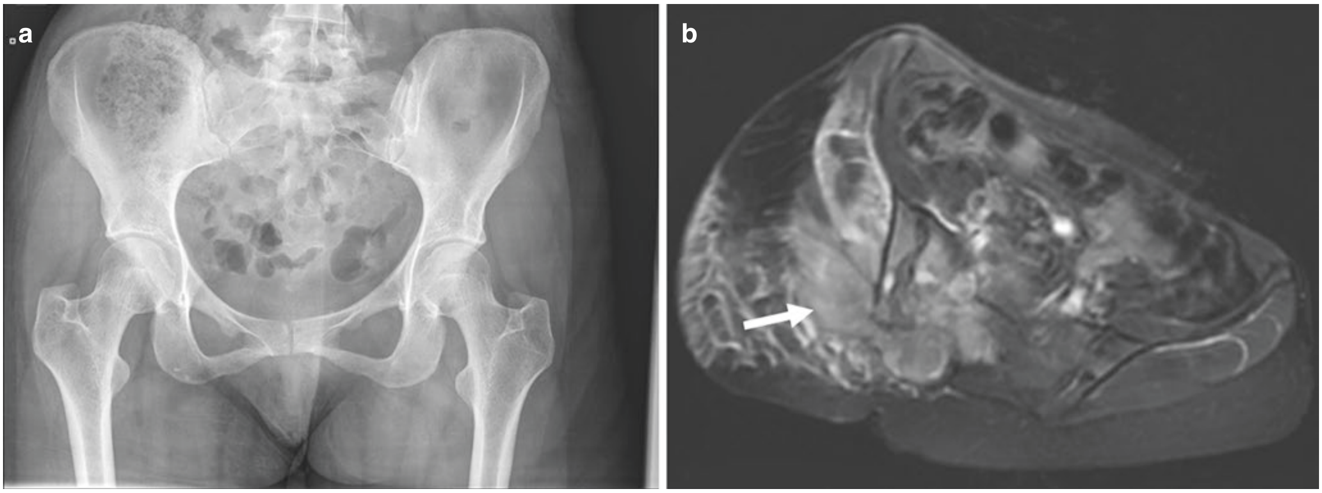

Fig. 3.1 Frontal radiograph of the pelvis (**a**) highlights the limitations of plain radiographs in the diagnosis of pelvic tumors. A subsequent axial T2-weighted fat-saturated image of the pelvis (**b**) performed 2 weeks later demonstrates a massive left sacropelvic destructive mass with marked surrounding edema and a large soft tissue mass (**b**—arrow). Biopsy demonstrated Ewing sarcoma

or sunburst), or interrupted which may present with elevation of the adjacent periosteum (Codman's triangle).

Matrix mineralization refers to calcification that forms typical patterns on the acellular matrix. The presence of either chondroid or osteoid matrix is an indicator of the type of tumor present, but matrix formation can be found in benign or malignant lesions. Chondroid matrix is defined as having a stippled, comma, or popcorn-shaped calcification (Fig. 3.2), while osteoid matrix is fluffy and ill-defined calcification.

3.2.2 Computed Tomography

CT serves as extension of radiographic evaluation of pelvic bone tumors given the limitations of plain radiographs in the detection and characterization of pelvic bone lesions. CT may better define periosteal reaction, lesions margination, and matrix formation (Fig. 3.3). CT is superior to MRI for characterizing periosteal reaction, cortical destruction, and tumor matrix formation [1]. Likewise, CT allows for the creation of three-dimensional (3D) models which can assist in the preoperative planning and assisting of surgical resection through navigated resections [2, 3]. CT is the modality of choice for image-guided percutaneous biopsy of pelvic tumors as it allows for planning a safe approach for the biopsy and documentation of the path of the needle that allows for resection of the tract in malignant tumors. Computed tomography (CT) scans are helpful in the staging of patients with pelvic sarcomas (chest, abdomen, and pelvis) and are also useful in providing cross-sectional imaging for patients with MRI contraindications.

3.2.3 Magnetic Resonance Imaging

Magnetic resonance imaging (MRI) is the modality of choice to diagnose and characterize tumors of the pelvis and has proven superior to other imaging modalities for assessing articular extension [4, 5]. Ideally performed prior to any biopsy in order to reduce the biopsy-related changes, multiple planes (axial, sagittal, and coronal) and sequences types are used to characterize the tumors and for planning resection and reconstruction. All MRI protocols for pelvic tumors should include T1-weighted non-fat-saturated and fluid-sensitive sequences (T2-weighted fat-saturated or inversion recovery). Straight axial and sagittal images are standard and should be performed in the preoperative evaluation of all pelvic tumors. Coronal images may be performed in relation to the axis of the body, which are particularly helpful in determining the extent of periacetabular tumors. Coronal oblique images orientated to the sacrum are helpful in the evaluation of lumbosacral plexus involvement. Intravenous gadolinium contrast helps distinguish cystic/necrotic versus solid components of tumors (Fig. 3.4) which may be helpful in subsequent planning of percutaneous image-guided biopsy. Fat saturation of the post-gadolinium images increases the conspicuity of the margins of the mass and allows for better visualization of the blood vessels for surgical planning. Despite the superb soft tissue contrast and ability of MRI to confidently identify the composition of multiple tissue types, many bone and soft tissue tumors cannot be confidently diagnosed on imaging alone. These lesions are referred to as indeterminate and biopsy is required for diagnosis. All MRI exams should be interpreted with plain radiographs when possible. Tumor matrix calcifications are better defined on CT and plain radiographs. Chondroid matrix has high T2 signals with a lobular

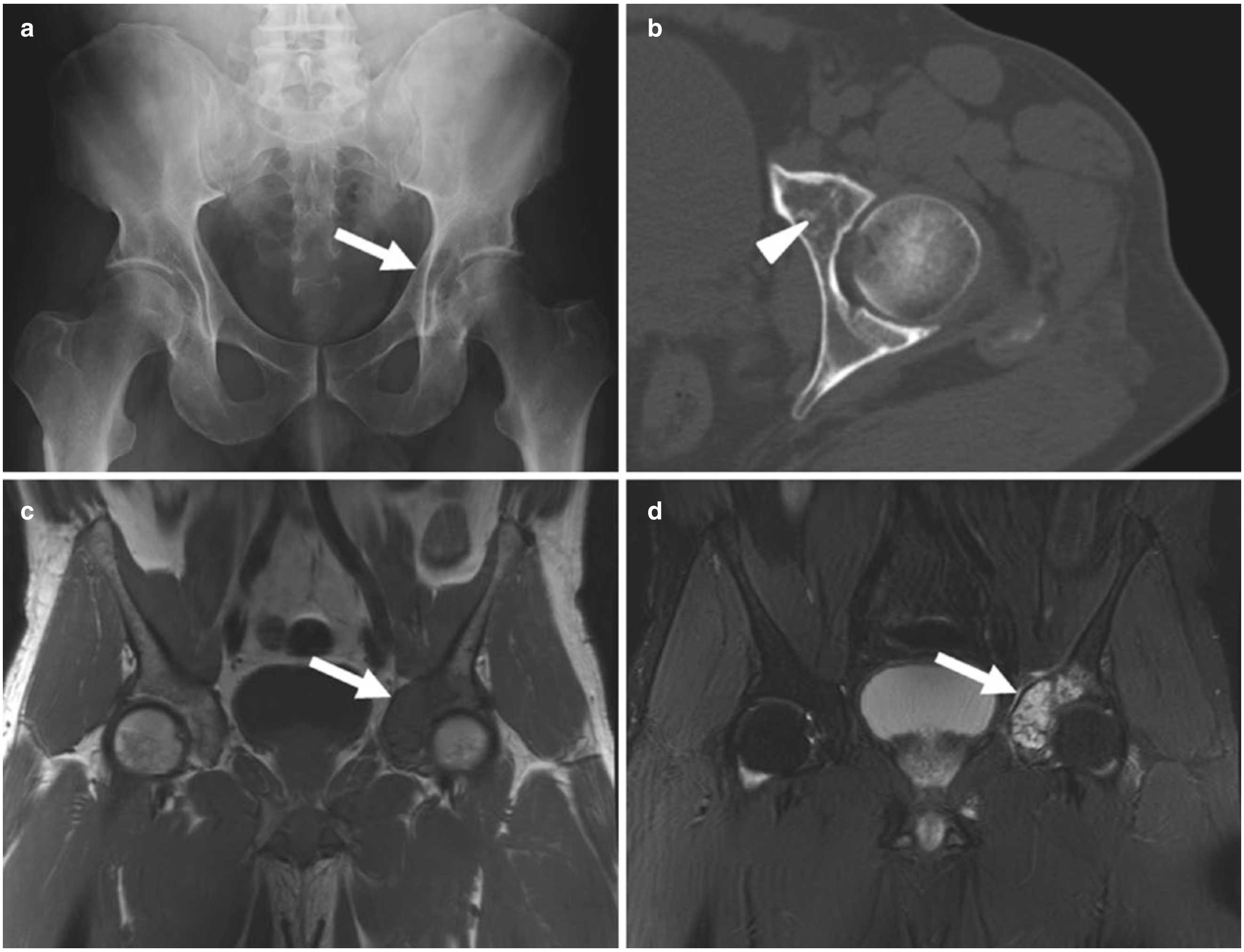

Fig. 3.2 A frontal radiograph in a 52-year-old man with left hip pain demonstrates a periacetabular lytic lesion (**a**—arrow) without cortical destruction or matrix mineralization identified. A subsequent CT demonstrates internal chondroid matrix (**b**—arrowhead). The T1 (**c**) and T2-weighted (**d**) coronal MRI images demonstrate the marrow-replacing mass with T2-weighted hyperintensity and lobulated margins consistent with a cartilage tumor. Pathology confirmed a grade II chondrosarcoma

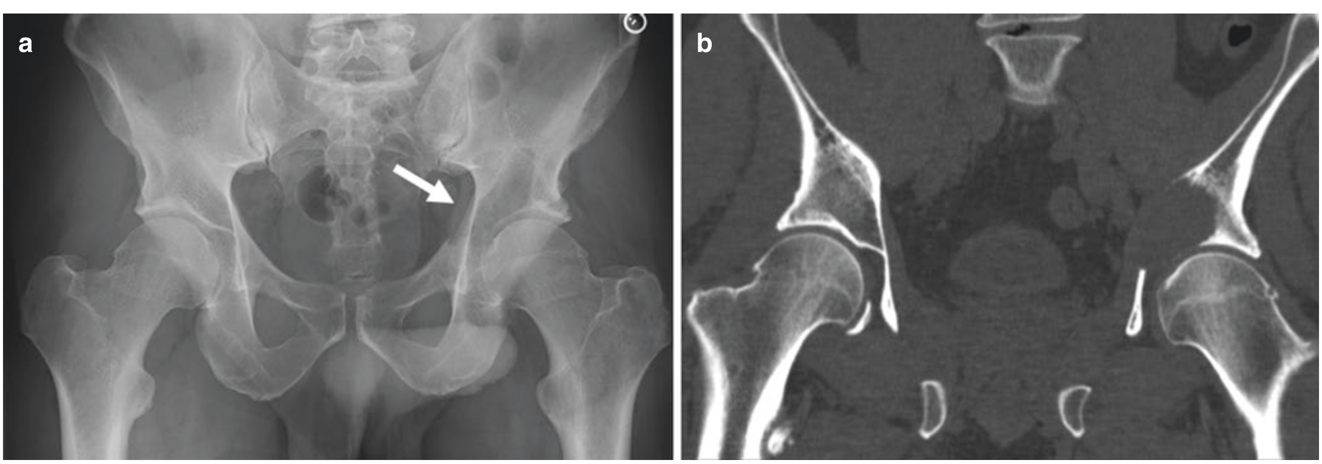

Fig. 3.3 A frontal radiograph (**a**) of a 52-year-old man demonstrates a purely lytic left periacetabular lesion. Although portions of the lesion appear to have a narrow zone of transition, cortical destruction is noted medial (**a**—arrow). A coronal CT image (**b**) of the pelvis better demonstrates the finding in this plasmacytoma

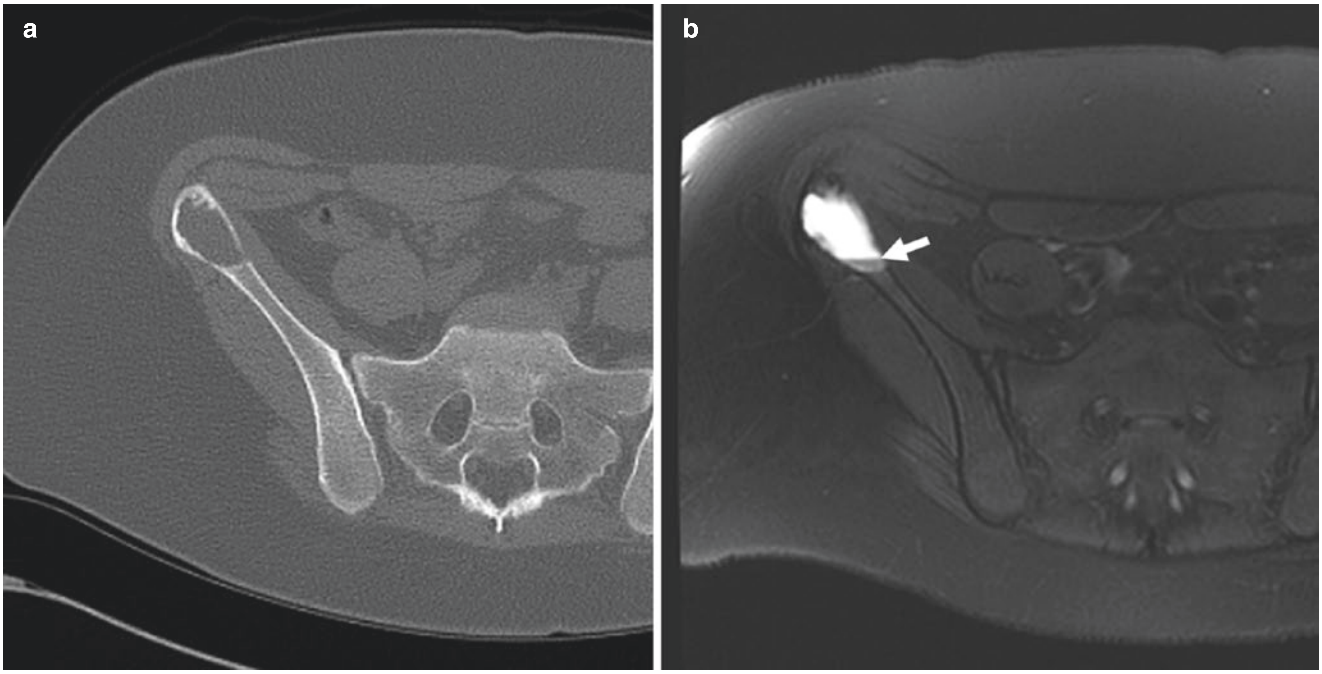

Fig. 3.4 Axial CT of the pelvis (**a**) demonstrates purely lytic lesion in the right iliac wing. The lesion has a narrow zone of transition, no cortical destruction, no periosteal reaction, and no soft tissue mass. Subsequent axial T2-weighted fat-saturated images of the pelvis show the lesion to be very hyperintense, similar to simple fluid. A linear fluid-fluid level is noted confirming the cystic nature (**b**—arrow). This lesion has benign imaging features. Curettage confirmed a simple bone cyst

growth pattern with low-signal septations (Fig. 3.2). Osteoid matrix has intermediate to low signal intensity on all sequences. The margin characterizations of lytic bone lesions used in plain radiography and CT should not be applied to MRI images. The bone tumor margins on MRI are formed by the interface of the tumor and the bone marrow which is predominantly fat in older individuals (Fig. 3.5). Aggressive intramedullary tumors may form a sharp boundary with the adjacent fatty marrow, but this does not necessarily indicate that the tumor is "well-circumscribed," a term that should be reserved for radiographs. Unlike plain radiographs, MRI is an excellent means to define the extent of a sarcoma but can overestimate the size of the tumor due to increased signal changes from edema. The spread of the tumor in the pelvis can provide clues as to whether a tumor is benign or malignant, with spread across the sacroiliac (SI) joint or articular involvement and breakthrough in the hip suggestive of a malignant process. Likewise, MRI can evaluate the extent of neurovascular involvement, specifically the T1-weighted sequences where fat planes are readily demonstrated.

In addition to being the imaging modality of choice to diagnose and characterize tumors of the pelvis, MRI is also the preferred modality to identify a preserved fat plane between the tumor and vascular structures. This is best seen in the non-fat-saturated T1 images, but it is also helpful to have post gadolinium-enhanced sequences to compare to. Although a CT angiography has better spatial resolution and is a better modality in determining patency of small arteries and intraluminal extension, it lacks the soft tissue contrast of MRI, and therefore it is difficult to determine if the vessels are abutted by the tumor. Due to this feature, MRI alone is typically adequate to determine vascular involvement.

Pre- and post-adjuvant treatment MRIs are essential for surgical planning. Often, the peritumoral edema on pretreatment studies is gone on the posttreatment studies. Typically, we plan to resect a margin beyond the pretreatment peritumoral edema in order to obtain a negative margin. This also highlights the importance of obtaining the same imaging sequences, in the same planes, for preoperative planning. Likewise, MRI imaging is also commonly used in the follow-up setting in order to evaluate for disease recurrence.

In addition to the common sequences obtained with imaging of the pelvis, specific sequences can be added in order to determine certain tumor characteristics. Since tumor tissue enhances with early dynamic images, dynamic-enhanced MRI may allow for the differentiation of tumor extension from reactive edema [6, 7]. Similarly, since active tumors enhance, and nonviable tumor doesn't readily enhance,

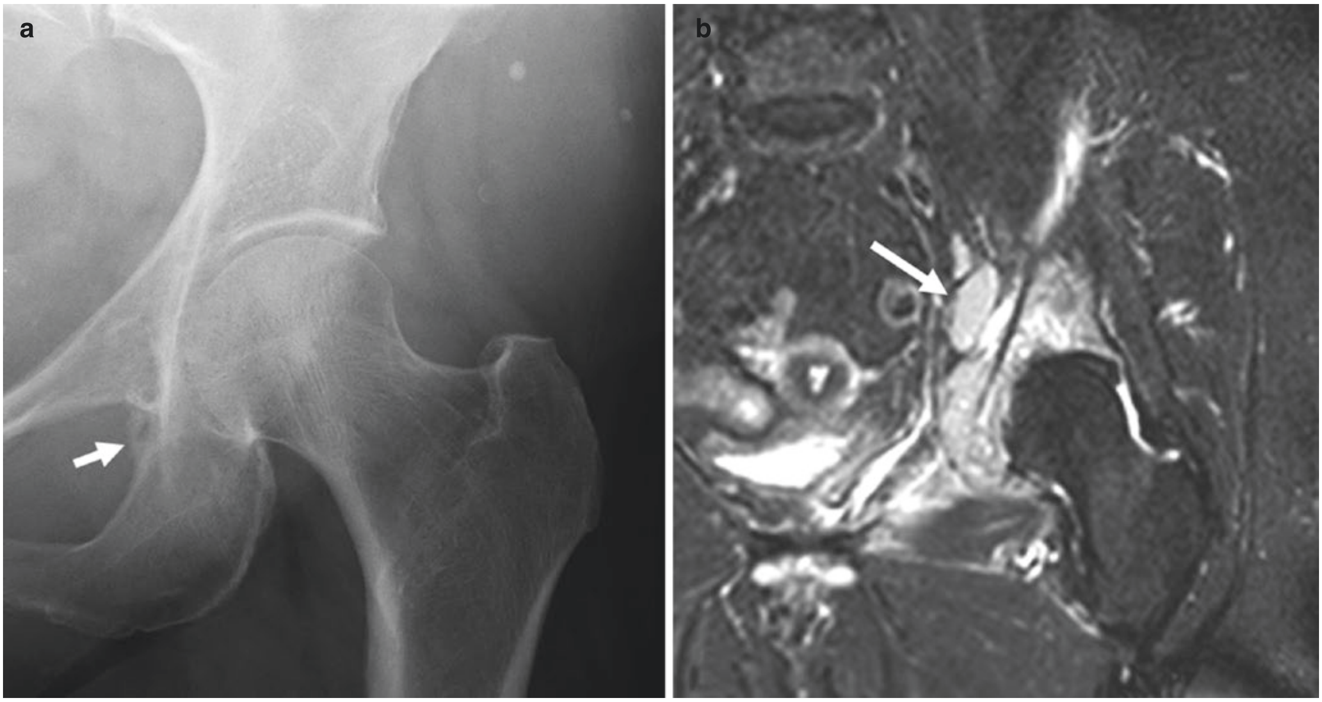

Fig. 3.5 AP radiograph of the left hip demonstrates subtle periosteal reaction along the medial margin of the ischium (**a**—arrow). The subsequent coronal STIR MR image demonstrates a large marrow-replacing mass in the left acetabulum with soft tissue extension and an enlarged left pelvic node (**b**—arrow). The differential diagnosis in the older patient is metastasis, myeloma, lymphoma, and primary sarcoma. The relative lack of findings with extensive involvement on MRI can be seen with osseous lymphoma, which was confirmed on a subsequent biopsy

dynamic MRI can assess the response to treatment by determining the extent of tumor necrosis [8].

3.2.4 Positron Emission Tomography/Computed Tomography

Positron emission tomography/computed tomography (PET/CT) has recently become increasingly used in the diagnosis and evaluation of pelvic tumors. PET/CT enhances the use of CT imaging and allows for the differentiation of benign from malignant lesions, detection of metastatic disease, and tumor surveillance [9]. PET/CT is based on the uptake of [^{18}F]-2-deoxy-2-fluoro-D-glucose (FDG) in the tumor which is associated with cellular glycolysis. The degree of FDG avidity can be compared to the metabolic activity of the surrounding tissue, with increased uptake typically found in malignant tumors [10–13]. In a study by Aoki and colleagues [13], the authors noted an increased uptake of two times greater uptake for benign lesions and four times greater uptake for malignant lesions compared to background tissue for bone lesions. The strength of PET/CT is in the ability to stage malignant tumors.

3.3 Imaging Features of Common Pelvic Tumors

3.3.1 Malignant Lesions

3.3.1.1 Chondrosarcoma

Chondrosarcoma is the most common primary malignancy of the bones of the pelvis and is characterized by a malignant proliferation of chondroid matrix. Chondrosarcoma can either be a primary bone tumor or a secondary tumor arising from an enchondroma or osteochondroma. In the pelvis, the tumors are typically low grade, with high-grade tumors having a worse prognosis [14].

Primary cartilage tumors in the pelvis should be approached with caution as the imaging features applied to differentiate enchondromas from low-grade chondrosarcomas in the extremities should not be directly applied to the pelvis. Aside from hereditary multiple enchondromatoses, pelvic enchondromas are rare. The incidence of chondrosarcoma and enchondromas is opposite from the extremities where benign enchondromas are quite common [15]. Typically, low-grade tumors have well-defined margins and may have cortical expansion. Chondroid matrix may or may not be present in pelvic chondrosarcoma. The "rings and arcs" pattern of chondroid matrix mineralization is better delineating on CT than plain radiographs for pelvic chondrosarcomas. On CT, the matrix is often described as stippled, and there can be cortical destruction, periosteal reaction, pathologic fracture, and an associated soft tissue mass with chondrosarcomas. On MRI, the mass demonstrates lobules of high T2 signal consistent with chondroid matrix. Dedifferentiated chondrosarcoma has components of both low-grade and high-grade tumors. A large lytic destructive tumor with a soft tissue mass adjacent to a cartilage tumor with matrix mineralization suggests the diagnosis of dedifferentiated chondrosarcoma [16].

Secondary chondrosarcoma may arise in the cartilage cap of osteochondromas (Fig. 3.6). A small cap, less than 2 cm thick, is typically benign. A cartilage cap thicker than 2 cm with irregularity of the osseous stalk, irregular calcifications, and a poorly defined cap is suspicious for malignant transformation into a chondrosarcoma [17].

3.3.1.2 Osteogenic Sarcoma

Although not as common in the pelvis as the extremities, conventional osteogenic sarcoma is the second most common primary malignancy of bone [18]. In the pelvis, osteosarcoma is most commonly located in the ilium and acetabulum (Fig. 3.7). On plain radiographs, osteosarcomas have aggressive features with "cloud-like" osteoid matrix formation and mixed areas of osteolysis and sclerosis. Osteosarcomas with greater areas of osteolysis are more aggressive tumors and associated with cortical destruction and a soft tissue mass. The diagnosis of osteosarcoma can often be made on plain radiographs alone. MRI is typically used to evaluate the

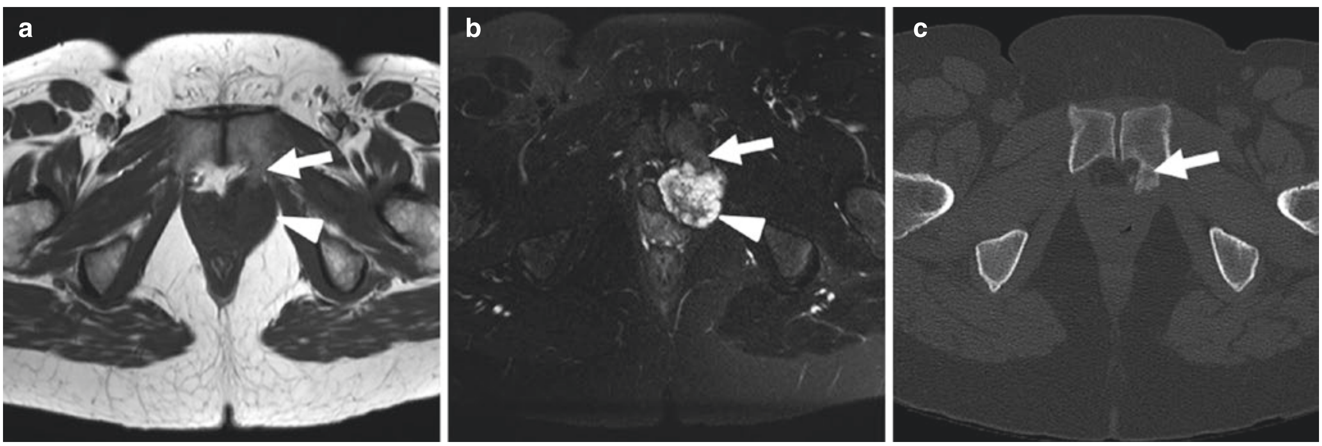

Fig. 3.6 Axial T1- (**a**) and T2-weighted fat-saturated (**b**) MR images demonstrate a lobulated T2 hyperintense mass compatible with a cartilage tumor (**a** and **b**—arrowheads). An osseous stalk from the adjacent pubic bone can be seen on the MRI (**a** and **b**—arrows) and is better visualized on the subsequent CT (**c**—arrow)

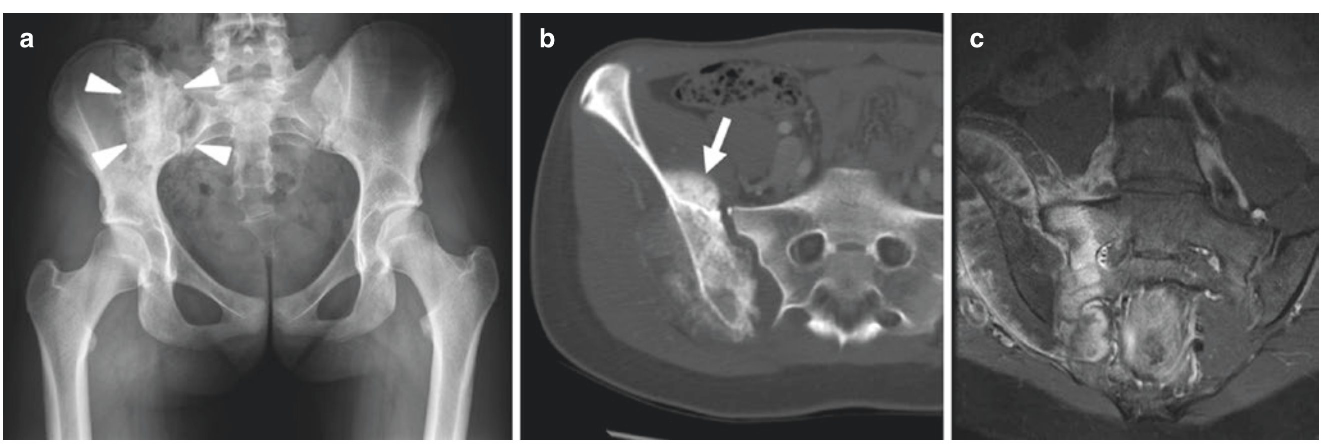

Fig. 3.7 A frontal radiograph (**a**) in a 16-year-old with low back pain demonstrates a large sclerotic lesion projecting over the sacroiliac joint (**a**—arrowheads). An axial CT image shows a large associated soft tissue mass with cloudy-like areas of matrix mineralization (**b**—arrow) diagnostic of osteoid matrix in this osteosarcoma. A coronal oblique T2-weighted fat-saturated MR image of the sacrum (**c**) shows the extent of the soft tissue mass and involvement of the sacrum

extent of the soft tissue mass and involvement of neurovascular structures. On MRI, the mass has heterogeneous signal intensity of low and high signal on T1 and T2 imaging and is the most common sarcoma with fluid-fluid levels [19]. Even with aggressive treatments, the reported 5-year overall survival of osteosarcoma of the pelvis is 38% [20].

3.3.1.3 Ewing Sarcoma

The most common axial site for Ewing sarcoma is the pelvis, and the imaging features of Ewing sarcoma of the pelvis are similar to that of the extremities [21]. On plain radiographs and CT, there is a moth-eaten and permeative destructive lesion with ill-defined cortical margins. In contrast to extremity Ewing, in the pelvis, there is often more reactive sclerosis with a concentric expansion of the cortical bone, leading to the classic "onion-skin" appearance. On MRI, the tumor is hypo- or isointense on T1, with increased signal intensity on T2 (Fig. 3.1). On MR, an associated soft tissue mass is commonly seen, most frequently in the gluteal and iliac compartments [22]. Likewise, Ewing sarcoma can cross the SI joint and is associated with a 75% 5-year survival with appropriate treatment [23].

3.3.1.4 Chordoma

Chordomas are the most common malignant lesions of the sacrum [24]. Since they are typically slow-growing tumors, they are frequently quite large at the time of diagnosis. On radiographs, chordomas appear as a solitary, midline lytic lesion centered typically at the lower sacral vertebrae (3rd through 5th) [25]. Although not readily apparent on radiographs, there is always a soft tissue mass, and in half of the patients, it is associated with focal calcifications and epidural extension on MRI and CT [26]. On MRI (Fig. 3.8), the tumor is slightly hypointense to isointense on T1 and, due to a high concentration of mucin, hyperintense on T2-weighted signal [25]. Having an adequate surgical margin has been found to be the most important factor on local disease recurrence and overall survival [27].

3.3.1.5 Liposarcoma

Liposarcomas are common tumors of the retroperitoneum [28, 29]. In the extremities, the term well-differentiated liposarcoma has been replaced by atypical lipomatous tumors given they do not metastasize unless they are dedifferentiated. In the pelvis, this may lead to confusion in the evaluation of greater sciatic notch tumors (Fig. 3.9). The MR features differentiating lipomas from atypical lipomatous tumors in the extremities have been well described. Typically, atypical lipomatous tumors of the extremities will have at least 75% adipose tissue; however, tumors with as little as 25% adipose tissue have been reported [30]. The use of contrast is helpful in differentiating between a benign lipoma and a well-differentiated liposarcoma. In a lipoma, the septations are thin (<2 mm thick) and have a faint enhancement; however, in a liposarcoma, they are thick and enhance on fat-suppressed T1 images [31]. In contrast, dedifferentiated liposarcomas have a non-lipomatous soft tissue mass, often with areas of necrosis and hemorrhage [32]. A biopsy should be considered for any solid, enhancing non-lipomatous mass associated with a lipomatous tumor to exclude tumor dedifferentiation. Notable benign mimickers of pelvic liposarcoma are extramedullary hematopoiesis and extra-adrenal myelolipoma. Both of these benign masses can occur in the presacral space and contain both lipomatous and soft tissue elements. Biopsy is typically necessary to confirm the diagnosis and to exclude liposarcoma.

Since they are fatty tumors, sometimes, they may be apparent on plain radiographs; however, the utility of plain

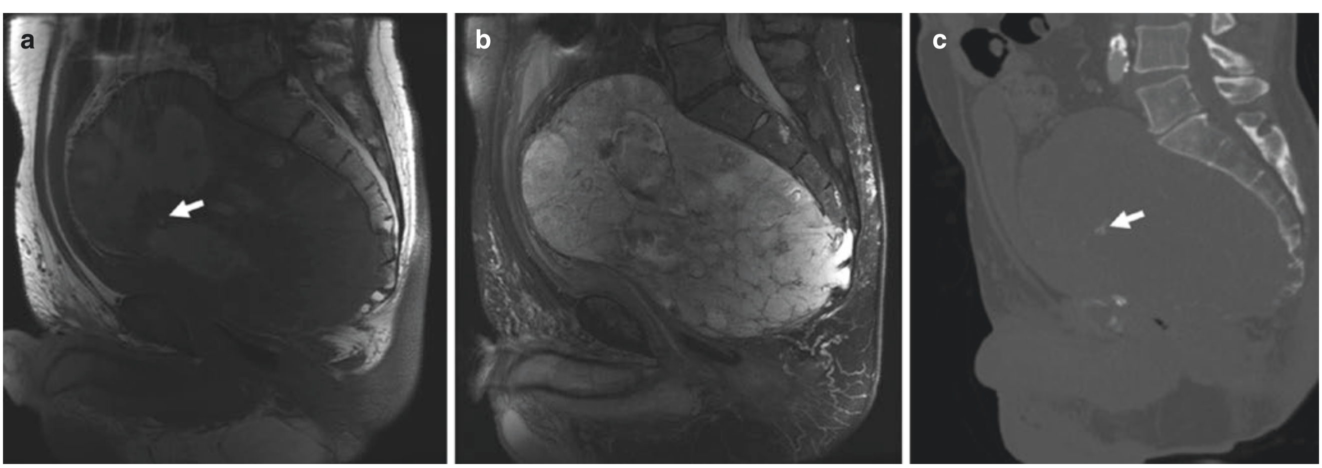

Fig. 3.8 Sagittal T1 (**a**) MR image demonstrating slightly hypointense mass with areas of increased signal intensity consistent with hemorrhage arising from the coccyx and S5 vertebrae. Sagittal T2-weighted (**b**) MR image demonstrates a lobulated T2 hyperintense mass with areas of septation and also focal calcifications (arrows) which are readily apparent on CT (**c**). Biopsy was consistent with a chordoma

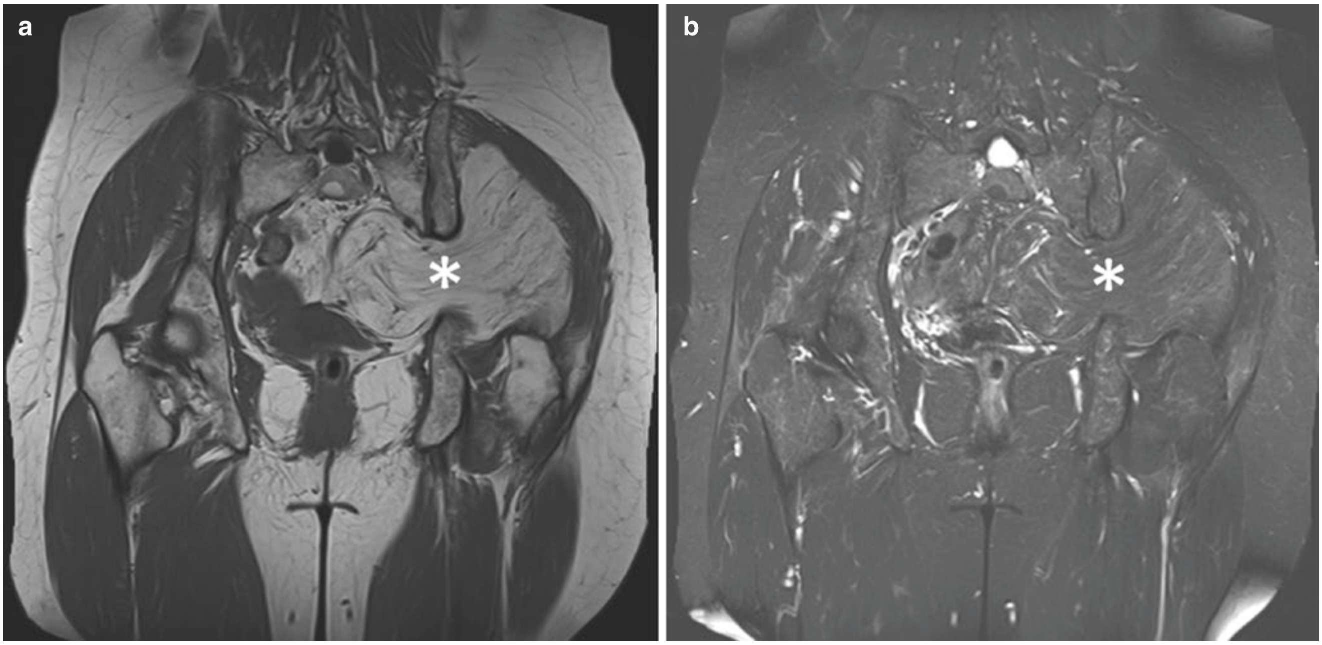

Fig. 3.9 Coronal T1- (**a**) and T2-weighted (**b**) images of the pelvis demonstrate a large "dumbbell-shaped" mass in the left greater sciatic notch (**a** and **b**—asterisks). The mass has both intrapelvic and extrapelvic involvement. The mass is comprised almost entirely of mature fat, following the signal characteristics of the subcutaneous fat compatible with a lipomatous tumor. The size, location, and internal septations raise concern for well-differentiated liposarcoma versus a large benign lipoma

radiographs for diagnosing these tumors is limited. On MRI, a well-differentiated liposarcoma images as predominantly fatty mass (hyperintense on T1, hypointense on T2), however, will have decreased signal intensity T1 in the areas of the thickened tumor septation and nodular areas [30].

3.3.1.6 Metastatic Disease

Metastatic disease is the most common malignant tumor of the pelvis and sacrum and, along with metastasis and myeloma, should be considered in the differential diagnosis of aggressive pelvic bone lesions in adults. Osseous metastatic disease may be lytic, sclerotic, or mixed lytic and sclerotic. Since solitary metastatic lesion can be treated and potentially cured with an en bloc resection, staging studies are recommended to exclude any additional sites of metastatic disease. Lytic lesions can be difficult to identify on plain radiographs; however, they are readily apparent on MRI due to confluent replacement of the fatty marrow on T1-weighted sequences and increase signal on fluid-sensitive sequences.

3.3.2 Benign Lesions

3.3.2.1 Giant Cell Tumor

Although classified as a benign lesion, giant cell tumors (GCT) are locally aggressive tumors with the ability to metastasize to the lungs [25]. In the extremities, they are metaphyseal and extend into the epiphysis. They most commonly involve the sacrum in the pelvis. On radiographs, GCT are lytic, often with a soft tissue mass (Fig. 3.10). GCTs typically lack periostitis, calcifications, and osteoid formation [33]. MRI is used to assess the extent of soft tissue, intra-articular, and interosseous involvement. GCT is hypointense on T1 and has a heterogeneous T2 signal; however, the solid component of GCT is dark on T2, due to hemosiderin [33]. Although GCT has a characteristic appearance in the long bones, pelvic GCTs have less specific imaging features, and biopsy is typically required to confirm the diagnosis.

3.3.2.2 Osteochondroma

Osteochondromas are the most common benign bone tumors, and the imaging features are characteristic. They are either a broad-based, sessile growth or a pedunculated lesion growing away from the growth plate with continuity of the cortex and medullary canal. Typically, these lesions are found incidentally; however, patients can also present with a painless, nontender mass and can be symptomatic if they compress on adjacent structures. A thin cartilage cap, less than 2 cm thick, however, larger lesions with irregular calcifications and a poorly defined cap are suspicious for malignant transformation into a chondrosarcoma [17].

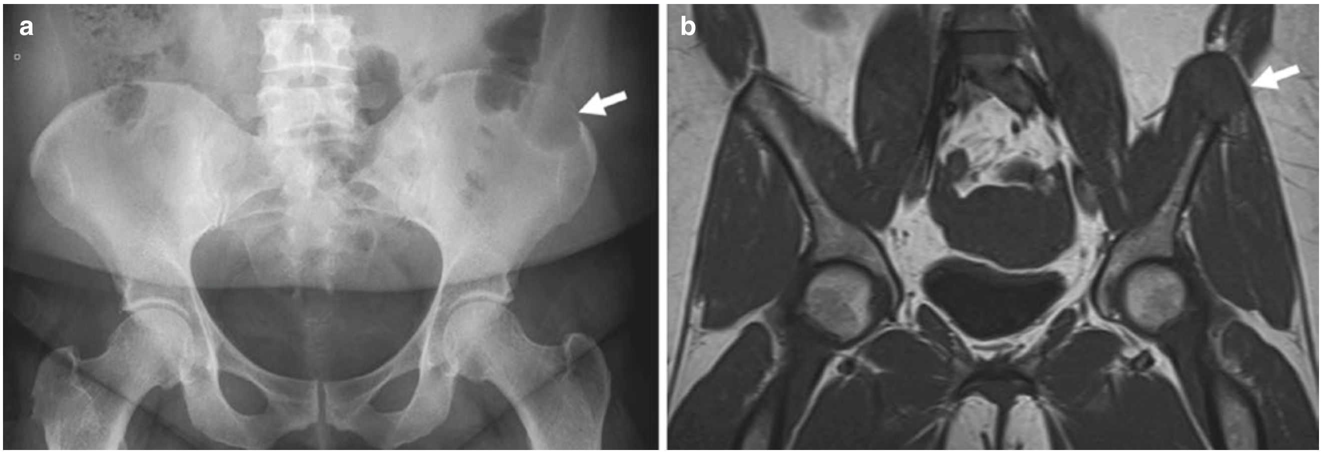

Fig. 3.10 Frontal radiograph of the pelvic demonstrates a lytic lesion in the left iliac wing with a narrow zone of transition (**a**—arrow). A subsequent coronal T1 MRI image demonstrates the expansile lytic lesion (**b**—arrow) diagnosed as a giant cell tumor on pathology

3.3.2.3 Aneurysmal Bone Cyst

An aneurysmal bone cyst (ABC) is an expansile osteolytic lesion with blood-filled spaces separated by connective tissue septa which contain bone, osteoid, and osteoclast giant cells [34]. The pelvis is a common location for these lesions in flat bones, accounting for up to 50% of cases [34]. Although the origin of an ABC is not entirely understood, they have characteristic imaging features. On plain radiographics, they are eccentric, lytic lesions with a well-circumscribed border. On CT and MRI, a cystic space with fluid-fluid levels and contrast enhancement of the septa is apparent. On MRI, there will be a high-to-low signal intensity of the fluid-fluid levels on T1- and T2-weigheted sequences. Well-circumscribed lesions on CT or radiographs that are comprised almost entirely with fluid-fluid levels on MRI are typically benign and are usually primary ABCs. Secondary ABCs can arise in a variety of other bone tumors with GCT and osteoblastomas most commonly associated with secondary ABC formation [19]. Telangiectatic osteosarcomas are often predominantly comprised of fluid-fluid levels as seen with primary ABCs; however, the differentiation of these two entities can commonly be made on plain radiographs. Primary ABCs typically have a narrow zone of transition and expand rather than destroy the overlying cortex. Telangiectatic osteosarcoma should have an aggressive radiographic appearance with a wide zone of transition, cortical destruction, and a soft tissue mass.

3.4 Summary

This chapter discusses the imaging modalities commonly used to diagnose a pelvic mass and illustrates some of the common malignant and benign tumors found in the pelvis. The differential diagnosis of a pelvic mass is broad; however, with the proper use of plain radiographs and cross-sectional imaging (CT and MRI), the diagnosis can be narrowed. However, a biopsy is typically needed to ultimately make the final diagnosis.

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4 Tumors of the Pelvis: Pathologic Aspect

Matthew T. Houdek and Carrie Y. Inwards

4.1 Introduction

Tumors of the pelvis are histologically similar to tumors arising in the extremities; however, less than 5% of primary sarcomas are found in the pelvis. Since patients with pelvic tumors often present with nonspecific symptoms, the diagnosis can be delayed, allowing these tumors to get quite large. Due to their size and relationship to other visceral structures, the surgical margins which are able to be achieved at the time of surgery are considered marginal, and disease recurrence is a problem. In order to formulate a treatment plan, and to have an educated discussion with patients on treatment outcome, an accurate diagnosis is needed. Imaging studies can narrow the differential diagnosis; however, a tissue diagnosis is imperative for treatment planning. The purpose of this chapter is to provide a general pathology review of some of the common tumors of the pelvis.

4.2 Soft Tissue Sarcomas

Primary soft tissue sarcomas of the pelvis are rare, with a majority originating from genitourinary system [1, 2]. These tumors are often large at the time of presentation and considered high grade with extra-compartmental extension, making it difficult to obtain a wide surgical margin, often making them a lethal disease [1, 3]. Liposarcoma and leiomyosarcoma represent the most common sarcomas of the abdomen and pelvis.

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4.2.1 Liposarcoma

Adipocytic tumors are frequently encountered by surgeons and pathologists, the majority of which are benign. Lipomas are the most common mesenchymal neoplasm in adults, but it rarely occurs in the retroperitoneum [4]. There are four types of liposarcoma including well-differentiated, myxoid, pleomorphic, and dedifferentiated liposarcoma. These tumors are typically low and intermediate grade; however, negative predictive factors for recurrence include poorly differentiated tumors, grade and stage 2 or 3 tumors, those larger than 20 cm, and a positive margin [5]. Liposarcomas are the most common retroperitoneal tumor in adults, but they can also be found in the true pelvis [6]. These tumors often present as a large mass, and often patients with retroperitoneal liposarcomas are older than patients with extremity tumors.

Well-differentiated liposarcoma (Fig. 4.1), which is synonymously called atypical lipomatous tumor (ALT) when located outside the body cavities, is the most common type of liposarcoma in the retroperitoneum. These tumors do not metastasize but may progress into a non-lipomatous form referred to as dedifferentiated liposarcoma. Well-differentiated liposarcomas are subdivided into three main histologic subtypes including adipocytic (lipoma-like), sclerosing, and inflammatory [7]. The presence of more than one histologic pattern in a single tumor is common, particularly in the retroperitoneum. The diagnosis is based on identifying adipocytes with varying degrees of cytologic atypia, often found within fibrous septa coursing throughout the tumor, in the background of mature fat. Most well-differentiated/ALT tumors can be diagnosed by histologic criteria, but at times the diagnosis can be challenging because areas of atypia can be focal. When faced with equivocal or borderline atypia, pathologists often rely on fluorescence in situ hybridization (FISH) testing for *MDM2* gene amplification, a consistent finding in well-differentiated liposarcoma/ALT [8, 9]. Moreover, FISH has

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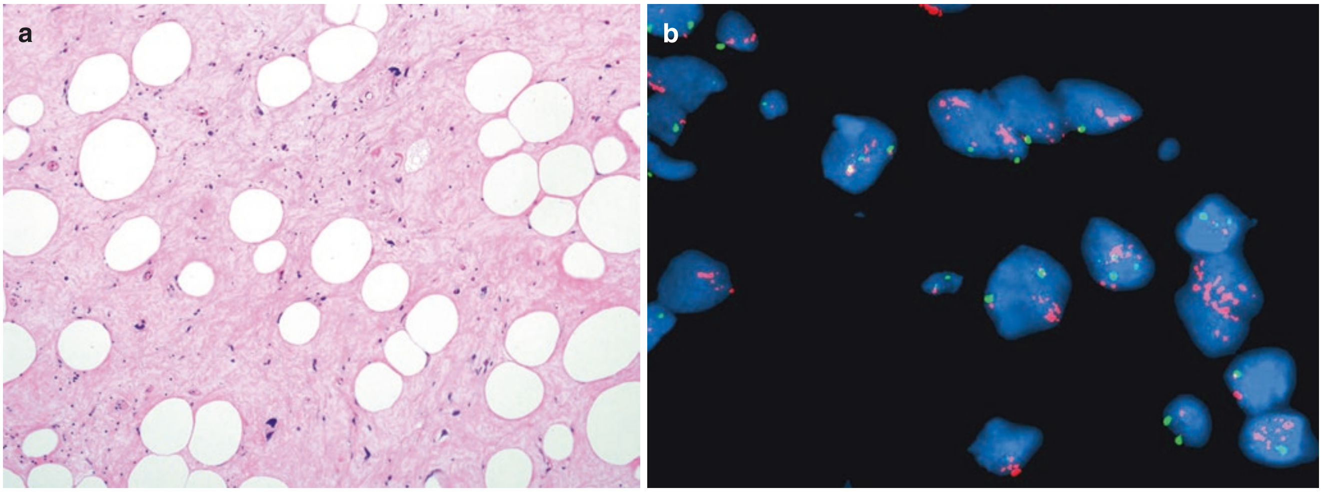

Fig. 4.1 Well-differentiated liposarcoma (sclerosing variant) demonstrating adipocytic cells embedded in a fibrillary collagenous background containing scattered atypical, hyperchromatic tumor cells (**a**). Fluorescence in situ hybridization (FISH) study illustrating *MDM2* amplification (**b**), a characteristic feature of atypical lipomatous tumor (ALT)/well-differentiated liposarcoma

been found to be a more reliable and cost-effective method when compared to immunohistochemical staining for *MDM2* and/or CDK4 [10]. In terms of treatment, patients with a sclerosing variant of well-differentiated liposarcomas, as well as those with positive surgical margins, are more likely to develop local recurrence [9].

Dedifferentiation occurs in approximately 10% of well-differentiated liposarcomas, although the risk is higher for those in the retroperitoneum where the majority of dedifferentiated liposarcomas occur. Histologically, most dedifferentiated liposarcomas are composed of a well-differentiated component and a high-grade non-lipogenic component most frequently resembling an undifferentiated pleomorphic sarcoma. These tumors may be misdiagnosed as the histologic subtype of the high-grade area due to inadequate sampling of a resected tumor or limited biopsy tissue. Dedifferentiated liposarcoma is characterized by a high incidence of local recurrence, and distant metastases arise in up to 18% of patients [11]. Occasionally, the areas of dedifferentiation are low grade, creating difficulties in the distinction from well-differentiated liposarcoma.

Myxoid liposarcoma occurs almost exclusively in the extremities in young adults. Genetically, it is characterized by the presence of *FUS-DDIT3* (>90%) or *EWSR1-DDIT3* (<10%) fusion genes. Myxoid liposarcoma is extremely rare in the retroperitoneum and usually represents metastatic tumor in this location [12]. Histologically, the tumor is composed of a myxoid stroma containing ovoid to round cells and prominent capillaries. Areas with myxoid features in well-differentiated liposarcoma may be confused with myxoid liposarcoma.

Pleomorphic liposarcoma is the rarest type of liposarcoma, accounting for 5% of all liposarcomas, and it carries a worse prognosis [13–15]. These tumors are high-grade pleomorphic sarcomas with multivacuolated lipoblasts with hyperchromatic scalloped nuclei and concave indentations created by clear cytoplasmic vacuoles [16, 17]. Compared to other liposarcomas, these are aggressive tumors with a reported 5-year survival of 63%, and as such an aggressive multidisciplinary care team is recommended [13].

4.2.2 Leiomyosarcoma

Leiomyosarcoma is the second most common soft tissue sarcoma of the retroperitoneum and typically presents as a large, nonfatty mass with the ability to extend into the surrounding vascular structures, specifically the inferior vena cava [18]. Histologically, leiomyosarcomas are typically composed of elongated spindled cells with blunt-ended nuclei arranged in intersecting fascicles. Epithelioid cytomorphology, myxoid change, and osteoclast-type giant cells are occasional findings. Poorly differentiated high-grade tumors may resemble other types of pleomorphic sarcomas. Immunohistochemical stains are often helpful in making the diagnosis. Smooth muscle actin and h-caldesmon are positive in >70% of leiomyosarcomas [19].

Previous studies have focused on the tumor depth and size, grade, mitotic rate, and presence of vascular invasion as to determine the prognosis of survival [19, 20]. In addition to these factors, it has also been shown that the location of the tumor plays an important factor in disease recurrence and can behave differently based on the anatomical site. Compared to patients with tumors of the extremity, those with tumors of the pelvis have worse overall survival [21].

4.3 Primary Bone Sarcomas

Treatment of malignant bone tumors of the pelvis is difficult due to the local anatomy, poor compartmentalization of the pelvis, and ability to achieve a wide surgical margin [22, 23]. In the pelvis, chondrosarcoma, Ewing sarcoma, and osteosarcoma are frequently encountered with the ability to achieve a negative surgical margin, the best predictor to prevent local recurrence and improve survival [22, 24].

4.3.1 Chondrosarcoma

Chondrosarcoma is the most common primary malignancy of the bones of the pelvis. The preliminary diagnosis of these tumors is often based on the clinical and radiographic features of the tumor; however, the pathologic findings confirm the diagnosis. Similar to the extremities, chondrosarcomas can be a primary bone tumor or a secondary tumor arising in patients with Ollier disease or an osteochondroma.

Histologically, chondrosarcomas show an infiltrative growth pattern characterized by tumor encasing preexisting trabecular bone (Fig. 4.2), the single most important finding that distinguishes it from enchondroma. Chondrosarcomas contain increased cellularity, nuclear enlargement/hyperchromasia, and myxoid matrix which become more prominent with increasing histologic grade (1 to 3). Dedifferentiated chondrosarcoma is a biphasic tumor, with areas of low-grade chondrosarcoma adjacent to a high-grade sarcoma, usually undifferentiated pleomorphic sarcoma. The diagnosis may be missed if inadequate tissue sampling fails to provide tissue from both components. Radiologic correlation can be helpful in avoiding this problem. In terms of treatment outcome, the grade of the tumor has been found to be one of the most important features for disease-specific and overall survival, with patients with higher-grade tumors (3 or dedifferentiated) having increased mortality compared to patients with lower-grade tumors (1 or 2) [25, 26].

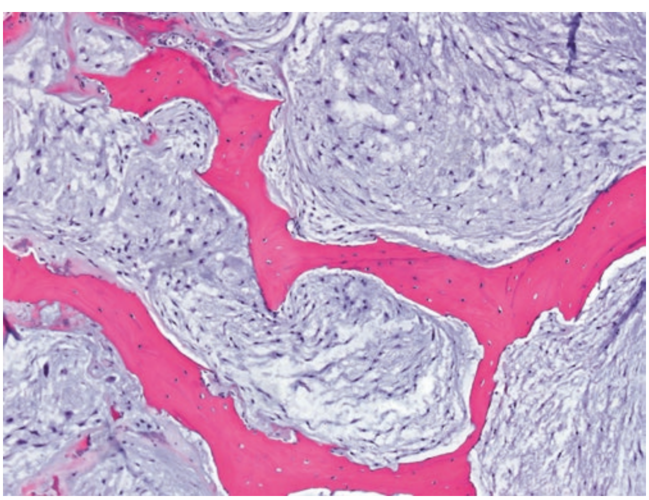

Fig. 4.2 Representative section of a chondrosarcoma showing a malignant hyaline cartilage tumor with increased cellularity and myxoid change in the matrix. The tumor is encasing the preexisting cancellous bone

The diagnosis of enchondroma involving a pelvic bone should be made with great caution as the vast majority of hyaline cartilage tumors in this location are at least low-grade malignant. On core needle biopsy tissue, it may be impossible to differentiate between enchondroma and low-grade chondrosarcoma at any anatomical site. In this situation, pathology can confirm the presence of a hyaline cartilage tumor which is then correlated with the radiologic features. The histologic differential diagnosis primarily includes chondroblastic osteosarcoma. In general, the cartilaginous component of chondroblastic osteosarcoma is higher grade than the majority of chondrosarcomas involving the pelvis. Moreover, the presence of malignant osteoid production rules out chondrosarcoma. At times, the distinction may be challenging, particularly with a limited amount of biopsy tissue. In these cases, mutation analysis looking for isocitrate dehydrogenase genes *IDH1* and *IDH2* may be helpful because these genes are mutated in 38–70% of primary chondrosarcomas and 86% of secondary chondrosarcomas [27].

The concordance of the preoperative biopsy and the final pathology for chondrosarcoma has historically been poor, especially in the pelvis [28]. This can potentially lead to treatment errors and is related to the heterogenicity of chondrosarcomas and the small amount of tissue obtained from a needle biopsy. This is especially apparent in the pelvis, where the tumors are often very large. Due to the heterogeneity, it is recommended to use the preoperative MRI to select specific areas with higher-grade features (reduced signal intensity of T2 MRI) to obtain the biopsy; however, even with obtaining tissue from this region, the diagnosis of a high-grade tumor can be missed [28].

4.3.2 Ewing Sarcoma

The most common axial site for Ewing sarcoma is the pelvis, and it is typically observed in pediatric and adolescent patients [29]. Grossly, Ewing sarcoma can have a liquid appearance resembling pus. Histologically, Ewing sarcoma demonstrates sheets or irregular islands of small blue cells containing oval nuclei with finely distributed chromatin surrounded by scant eosinophilic or clear cytoplasm. A small subset of Ewing sarcomas, previously classified as primitive neuroectodermal tumor (PNET), contain Homer Wright rosette formation suggestive of neural differentiation. By immunohistochemistry, the vast majority (90%) express CD99 and FLI-1. Approximately 20% show keratin positivity, a feature that may result in a mistaken diagnosis of metastatic small cell carcinoma. The histologic features of Ewing sarcoma can mimic hematopoietic neoplasms, particularly lymphoma. However, a thorough panel of immunohistochemical stains should be able to sort through this differential diagnosis. Similar to Ewing sarcoma, *BCOR-CCNB3* and *CIC-DUX4* tumors contain small blue cells and can show patchy CD99 positivity, and the distinction is made by molecular testing [30, 31].

The majority of Ewing sarcomas have a translocation between *EWSR1* and member of the ETS gene family. Most (85%) have t(11:22)(q24;q12), and 5–10% harbor t(21;22)(q2q12). This leads to the expression of the fusion oncogenes *EWS-ETS*, and detection of this gene fusion is used in the diagnosis (*EWS-FLI1* and *EWS-ERG*) [32]. Multi-agent chemotherapy and radiotherapy have improved the survival of patients with extremity Ewing. In the pelvis 5-year survival remains poor [33]; however, it can be improved when combined with appropriate surgical resection with negative margins [34].

4.3.3 Osteosarcoma

Conventional osteosarcoma is the second most common primary malignancy of bone in adults [35]. Secondary osteosarcoma involving the pelvic bones is usually seen in adult patients who have received radiation therapy to the area for a malignancy or in patients with Paget disease. Similar to Ewing sarcoma, unlike extremity osteosarcoma where the use of adjuvant treatments has improved survival, even with aggressive treatments, the reported 5-year overall survival of osteosarcoma of the pelvis is 38% [36]. The management is challenging due to the difficulty in achieving a complete surgical excision with adequate margins.

By definition, conventional osteosarcoma is a high-grade malignancy. Histologically, it is composed of malignant osteoid produced by atypical pleomorphic cells with a high nuclear-to-cytoplasmic ratio. The neoplastic bone usually has a disorganized lacelike appearance, but it can also be deposited in sheets or thick trabeculae. There are a variety of different subtypes, but the most common include chondroblastic, osteoblastic, and fibroblastic osteosarcoma. The radiologic features of osteosarcoma involving the pelvic bones are usually those of a malignant tumor, so the diagnosis is often made on tissue obtained from an image-guided needle biopsy procedure. The tumor cells typically display high-grade malignant features throughout the neoplasm, a feature that aids in making the diagnosis on a limited amount of tissue. However, the amount and distribution of malignant osteoid are variable and may hinder a definitive diagnosis. Immunohistochemical staining for SATB2, a marker for osteogenic differentiation, can be helpful in problematic cases [37]. However, caution is required as SATB2 is not specific for osteosarcoma and can be positive in benign and malignant mesenchymal tumors. Conventional osteosarcoma has complex karyotypes with an abundance of numerical and structural alterations. No specific translocation has been identified that would be helpful as a diagnostic test. Molecular testing for *IDH1* or *IDH2* mutations may be helpful in separating chondrosarcoma from chondroblastic osteosarcoma.

In the pelvis, the most common histologic subtypes are chondroblastic and osteoblastic osteosarcoma, with a high percentage of patients having metastatic disease at the time of presentation [36, 38–40]. In a study at a single institution with similar treatment protocols, the 5-year survival for extremity osteosarcoma was 75%; however, in the pelvis, it was 33% [38]. The authors of the study theorized that the poor chemosensitivity (20% good responders) and the high local recurrence accounted for the poor prognosis of pelvic osteosarcoma [38].

4.3.4 Chordoma

Chordomas are malignant tumors of notochordal origin and the most common malignant neoplasm of the sacrum [41]. Since they are typically slow-growing tumors, they are frequently very large at the time of diagnosis. Grossly, chordomas have a lobulated and gelatinous appearance. Histologically, at low magnification, the tumor has a lobular and infiltrative growth pattern. The tumor cells are arranged in nests and cords within a myxoid stroma (Fig. 4.3). Cytologically, they contain dark round nuclei with a moderate degree of atypia surrounded by eosinophilic cytoplasm. Intracytoplasmic vacuoles are a common feature, and cells containing them are known as physaliphorous cells. The amount of nuclear pleomorphism and necrosis can vary from tumor to tumor; however, this has not been associated with clinical outcome [42]. By immunohistochemistry, the vast majority of chordomas are positive for brachyury, the most sensitive and specific marker for diagnostic purposes. They are also positive for keratin markers, a feature that can be helpful in separating chordoma from other sarcomas, such as chondrosarcoma, but not of use when ruling out carcinoma. Clinically, the main prognostic factor in determining survival was the ability to achieve a wide margin [42].

4.3.4.1 Benign Lesions

Osteochondroma

Osteochondromas are the most common benign bone tumor. They arise on the surface of bones as either a pedunculated or broad-based mass which is in continuity with the cortex and medullary canal of the underlying bone. Histologically, osteochondromas contain a peripheral cartilaginous cap

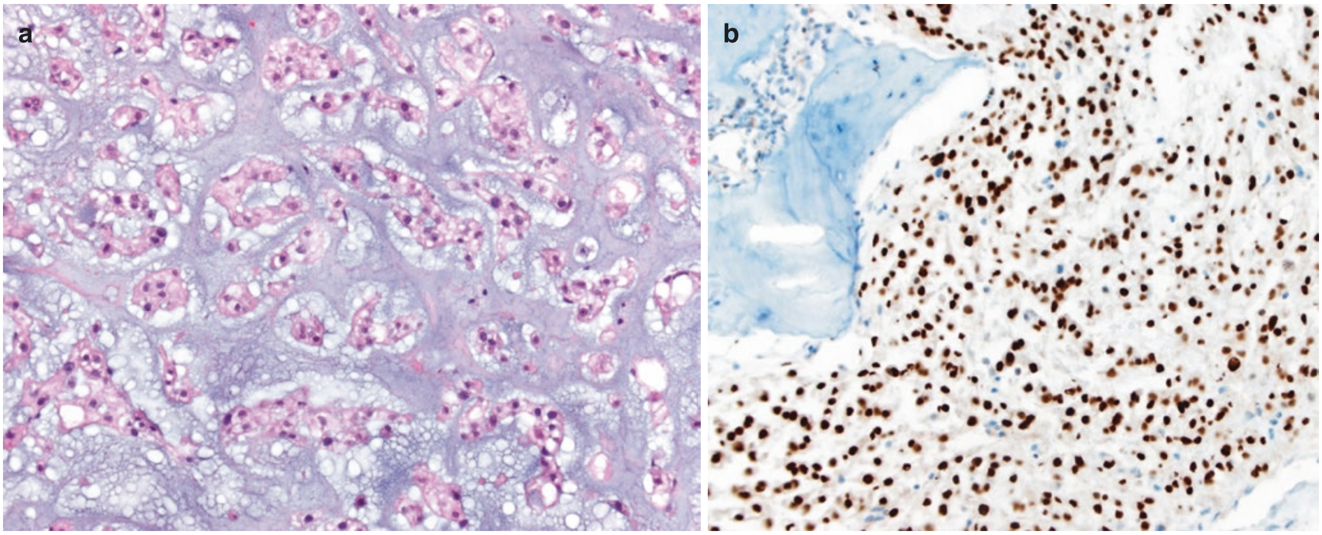

Fig. 4.3 Representative section of a chordoma showing nests and cords of cells with intracytoplasmic vacuoles (physaliphorous cells) embedded in a myxoid matrix (**a**). Chordoma cells also show diffuse nuclear positivity for brachyury immunohistochemical staining (**b**)

which undergoes enchondral ossification to form an underlying bony stalk of cancellous bone surrounded by fatty marrow. The cartilage cap typically contains chondrocytes with enlarged and hyperchromatic nuclei which can be mistaken for malignant chondrocytes if taken out of context with the organized architectural growth pattern. Since they contain an epiphysis, they can continue to grow until skeletal maturity; however, for lesions which continue to grow following maturity, malignant transformation needs to be ruled out.

These lesions are often found incidentally; however, in the pelvis, it can lead to nerve compression symptoms. Typically, the tumors are covered by a cartilage cap measuring <2 cm thick, and although malignant transformation is rare, it is more common in patients with multiple osteochondromas. Similar to tumors with continued growth, larger lesions with poorly defined cartilage cap and irregular calcifications are suggestive for malignant transformation into a chondrosarcoma [43]. Histologically, secondary chondrosarcoma arising in osteochondroma demonstrates nodular masses of neoplastic cartilage separating from the main mass and permeating through the surrounding soft tissue [44]. In general, it is extremely difficult, if not impossible, to make the diagnosis on limited biopsy tissue due to the increased amount of cytologic atypia seen in osteochondromas. Radiologic correlation usually provides compelling evidence for malignant transformation. Although these tumors can be treated expectantly, if there are signs of nerve compression or malignant transformation, they should be removed.

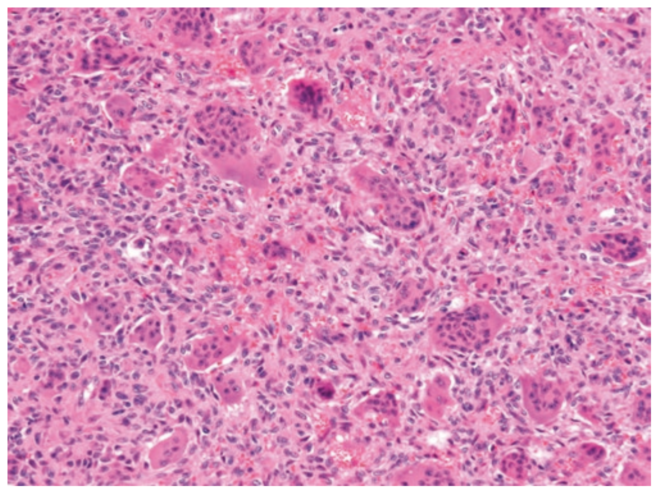

Fig. 4.4 Representative section of a giant cell tumor of bone showing neoplastic mononuclear cells without cytologic atypia admixed with numerous osteoclast-like giant cells

Giant Cell Tumor

Although classified as a benign neoplasm, giant cell tumor (GCT) is a locally aggressive tumor with the ability to metastasize to the lungs [45]. In the pelvis, these tumors most commonly involve the sacrum; however, compared to GCT in the extremities, the incidence of tumors in the pelvis is rare [46]. Histologically, these tumors are composed of neoplastic mononuclear cells without cytologic atypia admixed with numerous osteoclast-like giant cells (Fig. 4.4). The tumor cells contain round- to oval-shaped nuclei resembling those within the giant cells. Histologic heterogeneity including areas with spindled cells, necrosis, vascular invasion, cystic change, and reactive bone formation can create diagnostic challenges. Careful attention paid to the bland cytologic features of the mononuclear cells will help avoid a mistaken diagnosis of malignancy. Recently, recurrent somatic driver mutations in H3 histone family member 3A (H3F3A) (located at 1q42.12) have been identified in 92% of giant cell tumors of bone [47]. This finding leads to the development of immunohistochemistry for histone H3G34W, a highly sensitive marker for giant cell tumors which can be useful in challenging cases and core needle biopsy tissue [48, 49].

Treatment for GCT of the pelvis is controversial, ranging from intralesional curettage to a wide local excision with reconstruction [46, 50, 51]. One of the most difficult areas to treat these tumors is the periacetabular region of the pelvis, where patients undergoing an intralesional procedure have been shown to have an improved clinical outcome compared to a prosthesis [51]; however, this is outcome is worse compared to an intralesional procedure at another site in the pelvis [50]. The use of treatment adjuvants such as denosumab, which inhibits osteoclasts, has been used for the treatment of GCT in areas that are thought to be "unresectable" or for tumors which have metastasized [52]. Histologically, denosumab-treated tumors show a loss of the multinucleated osteoclast-like giant cells and replacement by variable amounts of fibrosis and woven bone. Therefore, it is important for pathologists to be aware of neoadjuvant therapy so as to avoid misdiagnosis of a benign or malignant bone-forming neoplasm.

Aneurysmal Bone Cyst

An aneurysmal bone cyst (ABC) is a benign, expansile osteolytic tumor containing blood-filled cystic spaces. In the pelvis, these tumors can be locally aggressive, with the most common location being the flat bones, particularly the sacrum and periacetabular region [53, 54]. Histologically, the tumors contain blood-filled spaces surrounded by a cyst wall composed of bland, slender spindled cells, scattered multinucleated osteoclast-type giant cells, and variable amounts of reactive woven bone which sometimes has a dark blue color. Solid areas of ABC containing the same histologic features of a cyst wall may be focal or the predominant feature. FISH for *USP6* gene rearrangement may be a helpful diagnostic tool since they are found in approximately 70% of cystic ABC [13]. The low-magnification appearance simulates telangiectatic osteosarcoma; however, ABC lacks the high degree of cytologic atypia characteristic of telangiectatic osteosarcoma. Solid areas of ABC resemble GCT. Radiologic correlation, immunohistochemistry for histone H3G34W, and/or FISH testing for *USP6* gene rearrangement can be helpful in making the distinction.

Treatment of these tumors is challenging and is individualized based on the tumor location, extent, and local bony destruction. We recommend treatment based on the size of the tumor, with tumors ≤5 cm with minimal bony destruction and do not threaten the integrity of the sacroiliac joint or acetabulum, treated with an intralesional procedure including exteriorization of the tumor, with or without an additional bone grafting procedure. For tumors >5 cm, large areas of bony destruction, or those which disrupt the sacroiliac joint or acetabulum, we recommend a more aggressive approach with resection and subsequent reconstruction of the defect [54]. Likewise, due to the propensity to bleed, we recommend for selective embolization for tumors involving the sacrum and acetabulum [54].

4.4 Summary

This chapter focuses on the pathologic features of commonly encountered pelvic tumors. The differential diagnosis of a pelvic tumor is broad, but through the use of appropriate imaging studies, combined with histologic examination of tissue with or without ancillary studies, a diagnosis can be ultimately made in a majority of patients.

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5 Staging, Preoperative, and Surgical Planning

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5.1 Introduction

Orthopedic oncologic literature has demonstrated that CAS is a useful and novel tool. PVP and IVN can potentially improve resection of tumors located in the pelvis (1–5) due to its geometrical complexity.

Formerly, surgeons planned their pelvic tumor surgery with a bidimensional printed slice image from computerized tomography (CT) and magnetic resonance image (MRI). Surgeons study the films and integrate CT and MRI, taking the oncologic margin into account. For years, this method has been the only alternative presenting insufficient accuracy and tumor recurrence due to wrong cutting margins. However, poor preoperative information led to wider resections as to achieve oncologic security.

Recently, technological developments allow physicians to manipulate digital medical images. In this way, integration of CT and MRI in a virtual simulation scenario is possible (6, 7). This virtual environment has the ability to include 3D bone tumor reconstructions. Specialists use a virtual simulation scenario to plan the cuts before the intervention and then execute it under IVN. That is to say that physicians have greater knowledge of the tumor area, improving the accuracy of the intervention (8, 9). The aim of this chapter describes the basics of PVP and its execution in pelvis sarcomas under IVN.

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5.2 Preoperative Virtual Planning

PVP works in a virtual simulation scenario where it is possible to merge digital medical images. This platform is potentially reliable for physicians in order to decide about further surgical techniques for a better quality intervention.

Moreover, PVP is a tool that can be used to plan a case and address possible surgical techniques. PVP is a platform where many specialists of different areas can see the case's approach together and interact before the real intervention.

Image acquisition protocols (CT and MRI) with cuts of 1 mm (millimeters) or less thickness were used.

The requested CT and MRI are merged (image fusion), and the oncologic margins are defined within the virtual scenario. The oncologic margin is calculated with dots in the healthy bone tissue and tumor limits. Specialists, depending on the tumor histology, measure the distances and determine the safe margins. In tumors such as in low-grade chondrosarcoma cases without edema, the oncologic margin limit is easy to define between healthy and pathologic bone tissue (Fig. 5.1), whereas in other histological types such as Ewing tumors, the dots are more difficult to fix (Fig. 5.2).

Each oncologic margin distance is unique according to the histological type and the definition of tumor limits. As general criteria in low-degree chondrosarcomas with no edema, the plan is done with a 5 mm margin, in osteosarcomas 1 cm, and in tumors with undefined limits 2 cm or more.

After the process of defining the margins of the tumor, physicians start planning the type of osteotomy they will perform in the OR. Each case is given a name after the number of planes in which it is going to be performed: uniplanar if it is only one plane, biplanar if there are two planes, and multiplanar if there are more than two planes (Fig. 5.3).

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Fig. 5.1 This MRI slice shows that the limit between healthy and pathologic bone is clear since there is no edema

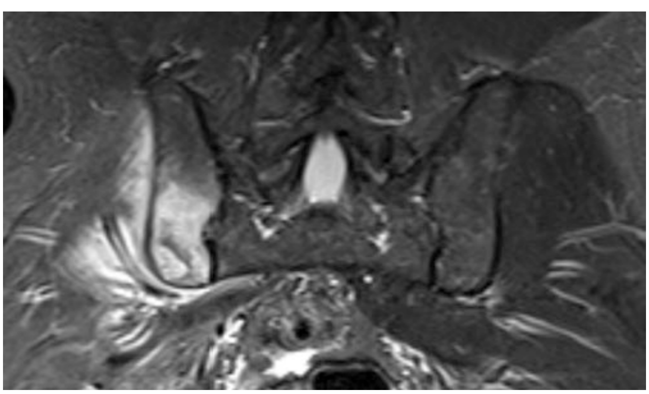

Fig. 5.2 This image shows a case where the tumoral limit is difficult to determine. This is frequent in Ewing tumors

The surgical approach is decided according to the proposed osteotomy; afterward, it is possible to perform a virtual resection. Specialists of different fields are able to actually see detailed features of the patient's pelvis and its tumor after virtual resection previous to the real intervention (Fig. 5.4).

Finally, after virtually resecting the tumor with its planned margins, physicians are able to foresee different reconstructive alternatives. Having access to the virtual library of banked allografts, bones allow to practice numerous reconstructions using structural bones (Fig. 5.5).

5.2.1 PVP Summary

1. Study the tumor in MRI.
2. Delineate margins according to histology.
3. Determine the conformation of planar osteotomy: uni-, bi-, or multi-planar.
4. Perform a virtual resection.

5.3 Intraoperative Virtual Navigation

Intraoperative virtual navigation (IVN) is the tool that executes PVP osteotomies.

This type of navigation is the optical navigation based on images, a system of infrared cameras (emission lights) found in specific instruments. The instrument is a tracker fixed on the patient and a pointer held by the surgeon. In this case, it has been used as active instrumental (powered by lithium batteries), although there is also passive instrumental (without batteries). The navigator has two monitors, one for technicians and other one for surgeons; both screens display the same (Fig. 5.6).

The first important aspect to mention is the place we are going to designate to the physical navigator in the operating room. It is important to place the device in the operating room after the sterile fields have been placed so as not to disturb the surgeon and avoid contamination.

The device is placed opposite to the surgeon so he can see the camera 2 m from surgical approach.

It must be determined in PVP the fixing site of the registrar. The fixing is the insertion of two pins between 3 and 4 cm diameter in the bone. It is of great importance to dispose such pins far from the tumor area so as not to disturb the surgeon during the intervention as well as disposing the pins where the camera can visualize them. Once the tracker is fixed, specialists proceed with registration process. Through this process, the virtual bone from planning and the real bone of the patient are compared and matched. Registration process can be divided in two parts, a primary and a secondary.

During primary registration, it is numerically determined those recognized bones from 3D reconstruction which can

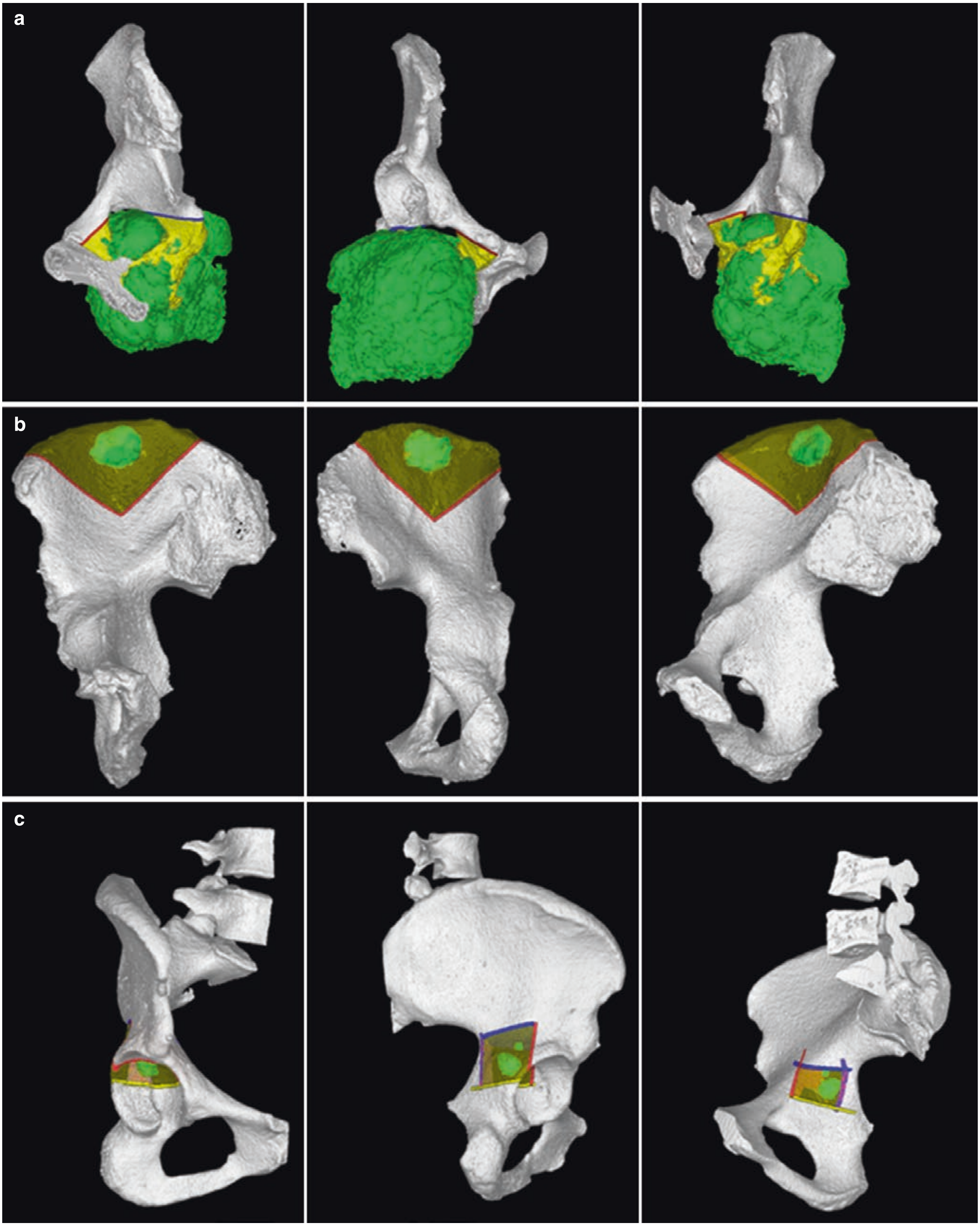

Fig. 5.3 Planar configurations: each configuration receives a different nomenclature. (**a**) Uniplanar, (**b**) biplanar, and (**c**) multi-planar

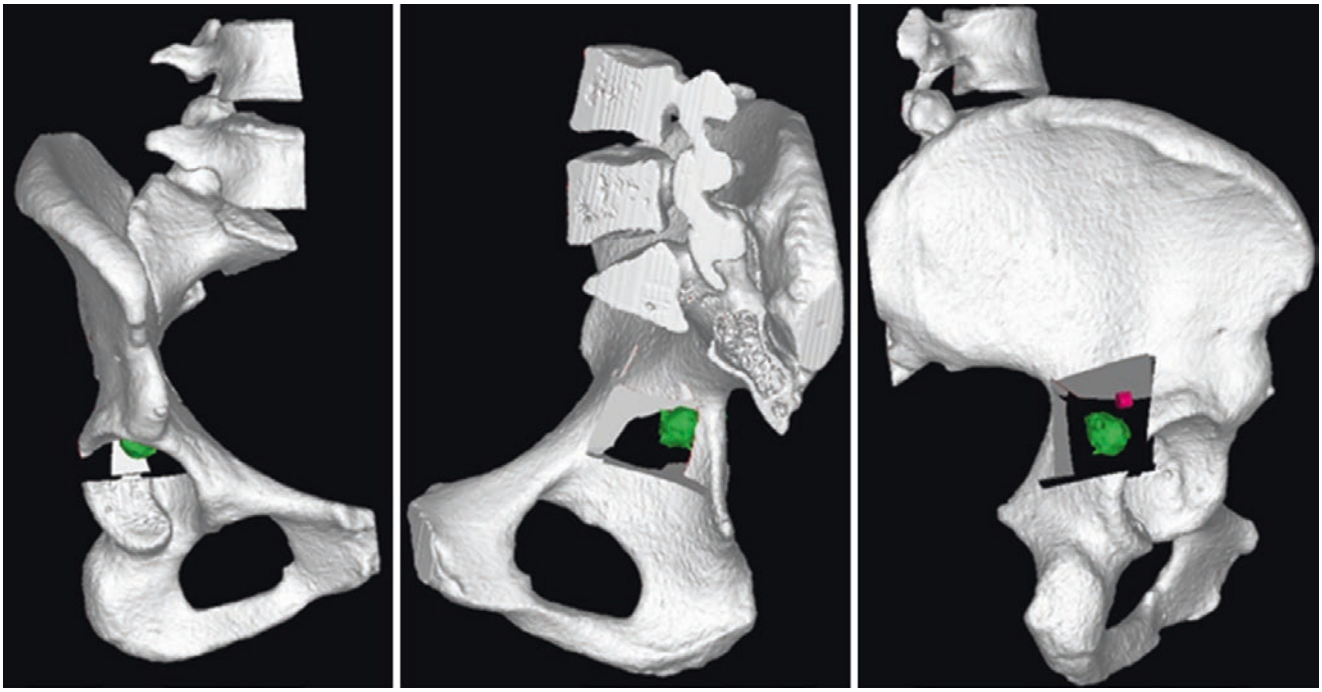

Fig. 5.4 A virtual resection is shown. The surgical approach is decided after a resection plan is built and evaluated in the virtual scenario

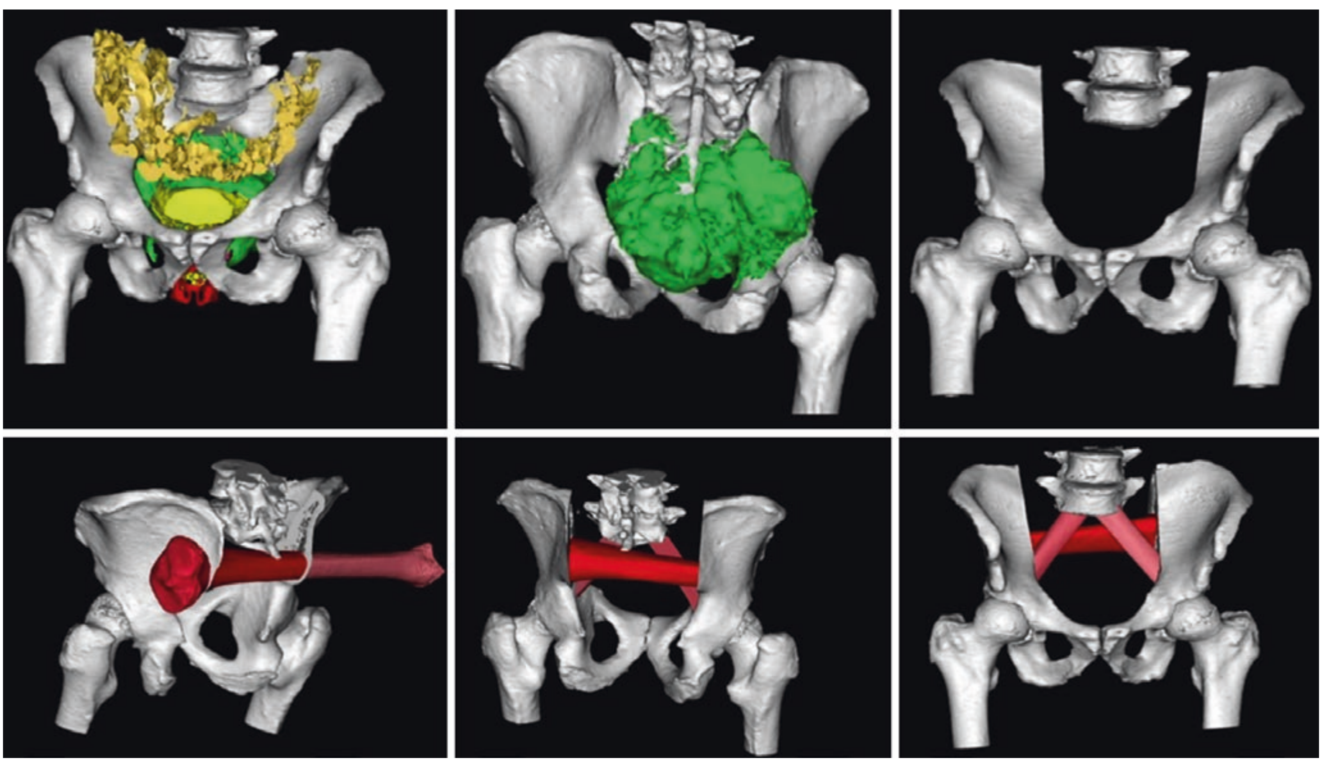

Fig. 5.5 Nonregular structural bone reconstructions using allografts bone models digitally stored in the virtual bone bank

Fig. 5.6 A navigation pointer in the front. In the back, the navigator screen showing the location of the tumor and the tip and angle of the pointer

be visualized after the approach. At least three landmarks are set. Then secondary registration consists of adding more landmarks to refine the area. This last step allows specialists to match completely 3D image with the real bone. In order to verify the accuracy of the process, specialists have to dispose the pointer over three different bone landmarks and corroborate visually that the pointer seen in 3D representation is found in air-bone interface (Fig. 5.7).

The following step is to mark the cut path in the bone with methylene blue that also resembles with PVP.

After marking the bone, the osteotomy is done with the saw blade. The saw blade thickness is foreseen in PVP, so it is left a 2 mm cut thickness.

When osteotomy is initiated, IVN is an asset in order to guide the procedure. The pointer is placed in the created slot, visualizing the exact position on the navigator screen and trajectory by three bidimensional windows.

Finally, an ultimate control can be done with the pointer after tumor resection. It is pointed to the remnant defect in order to verify visually the exactness of the osteotomy.

5.3.1 IVN Summary

1. Place the navigator device.
2. Tracker fixation.
3. Primary registration and secondary registration.
4. Air-bone check over three spots.
5. PVP navigation.
6. Defect control.

5.4 Accuracy in PVP and IVN

An experimental design scans the surgically removed specimen in a CT in order to determine the accuracy of the above-described procedure. The following procedure is to reconstruct three-dimensionally the surgical specimen and superimpose it to PVP (Fig. 5.8). This allows physicians to perform a comparative study between osteotomies virtually planned and those executed.

Although the first impression of the comparative study is a visual appreciation, virtual reconstructions and planes can be transformed into a map of dots (Fig. 5.8). This enables physicians to measure dot to dot, obtaining as a result a numeric value. A comparative study informs physicians quantitatively the accuracy between planned and executed.

The values can be expressed by colored graphs (colorimetric). For instance, green color is given when planned and executed osteotomy match. Red color is given when the osteotomy dangerously approached the tumor, and blue color is given when osteotomy shifted away from the tumor. Colorimetric graphics depict cut pathways behavior tridimensionally (Fig. 5.8).

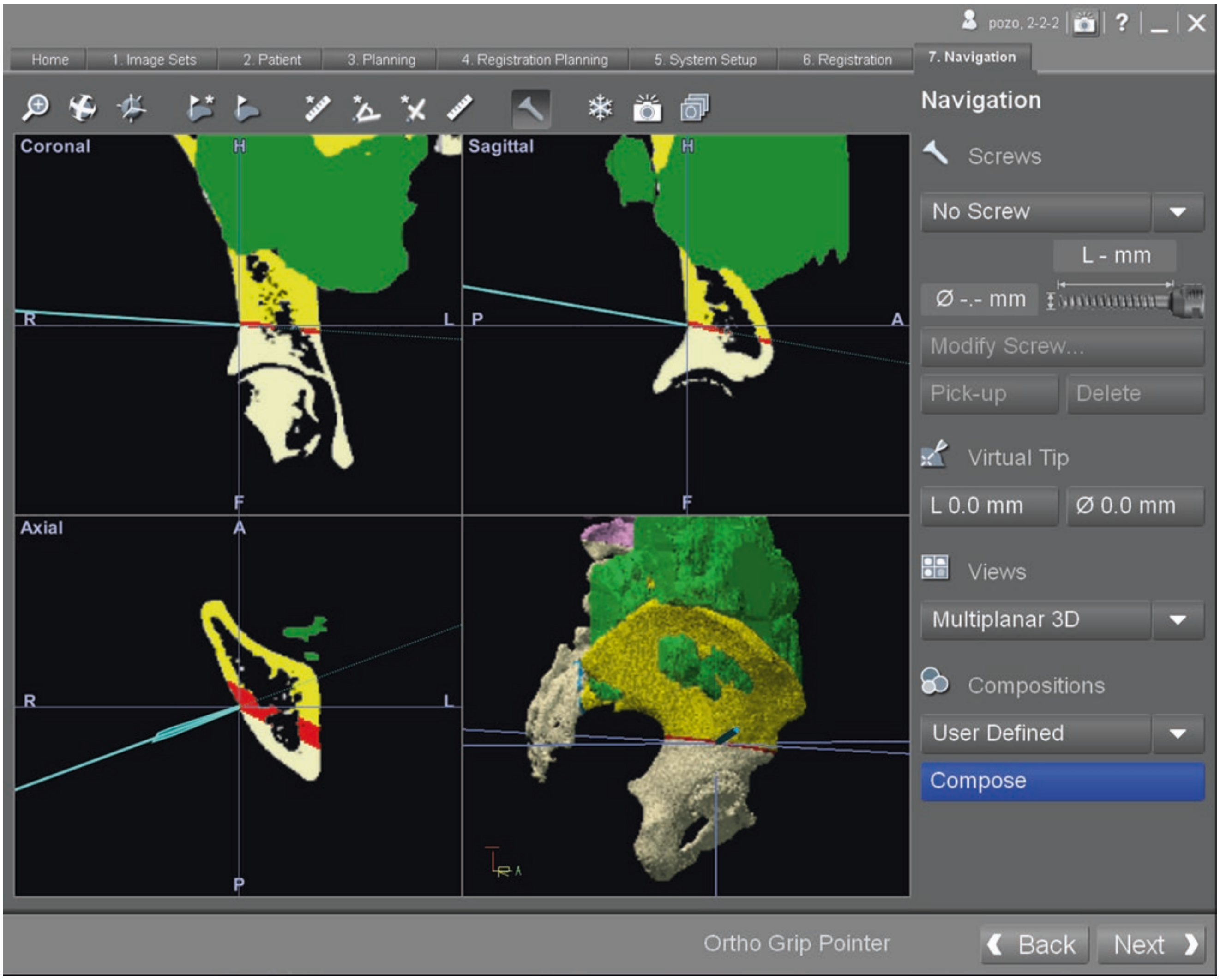

Fig. 5.7 The pointer is used to visually verify the registration process. This is done by checking the air-bone interface area in well-known locations

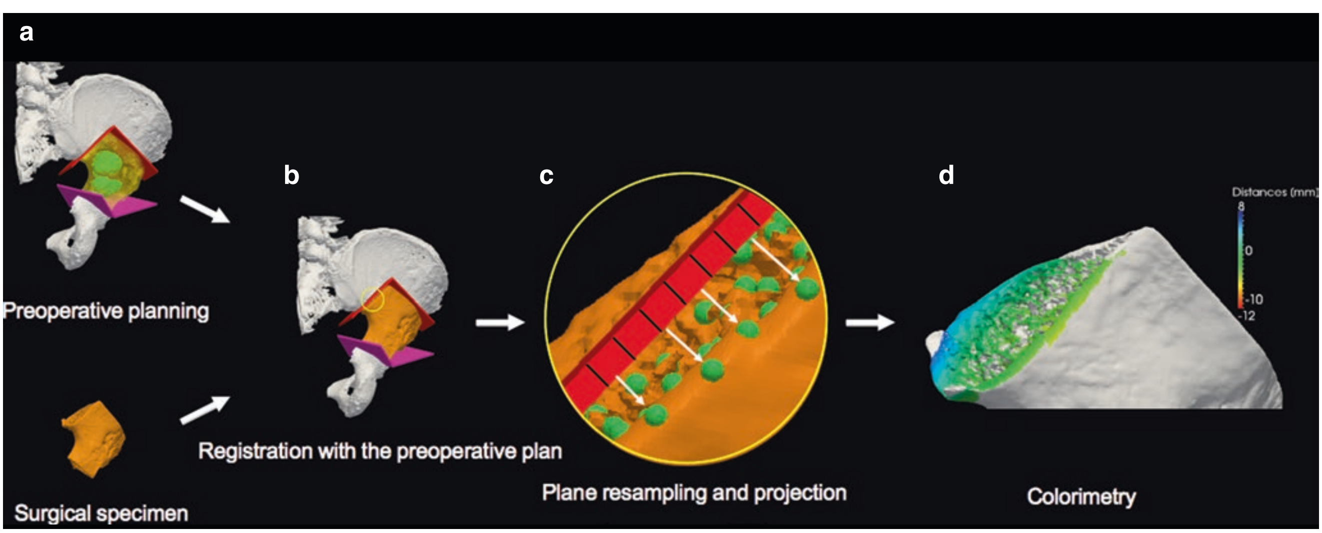

Fig. 5.8 The accuracy evaluation workflow. (**a**) The surgical specimen is CT scanned; (**b**) the preoperative plan is registered against the surgical specimen; (**c**) the distances from the osteotomy surface to the planar cut are measured; (**d**) the measured data is displayed so the surgeon can perform adjustments

5.5 Conclusion

PVP and IVN are tools used in the novel field of computerized assisted surgery area. They are important assets to perform pelvic sarcomas resections as they provide potentially optimal surface approach before surgery. Furthermore, these tools give specialists the necessary instruments to study a case in depth before the intervention and to prevent unexpected situations. Executing what was previously planned supported by accurate tridimensional images potentially improves safeness, quality, and, foremost important, predictability in osteotomies during oncologic surgery.

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Part II

Pelvic Tumors: Surgical Procedures

Overview on Pelvic Resections: Classification, Operative Considerations and Surgical Approaches

6

Peter F. M. Choong

Fifteen percent of all primary malignant bone tumours are located in the pelvis [1]. Their location and potential size have a major impact not only on patient survival but also the complexity of surgery. The three commonest sarcomas are chondrosarcoma, osteosarcoma and Ewing's sarcoma, and these may grow to substantial sizes before referral. Their requirement for adjuvant therapy differs, and this may impact the type of surgery performed as well as the complications post-operatively. Previously, transpelvic amputation (external hemipelvectomy) was the operation of choice [2]. However, with considerable improvements in survival from multimodality treatment, and advances in imaging that have improved the planning of surgical margins, pelvic resection with limb-sparing surgery (internal hemipelvectomy) is indicated in the majority of patients [2–4].

6.1 Classification of Tumours According to Location

Limb-sparing pelvic resections are associated with destabilisation of the pelvic ring, and depending on which part of the pelvis is resected, various adjacent structures (bone, muscle, joint and viscera) may also be affected. Classifying the location of the tumour and the associated resection allows a common language between surgeons, highlights the potential for involvement of adjacent structures related to certain locations and also allows discussion about the type of reconstruction required.

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The pelvis can be divided into four parts, namely, ilium, periacetabulum, pubis and sacrum. These anatomic locations have been classified as Type I, II, III and IV, respectively [5, 6]. Tumours located within any of these areas are designated according to the location, i.e., Type I, II, III or IV. If tumours cross over between locations, they are designated according to the combination of locations that they occupy, for example, Type I-IV for sacroiliac tumours and Type II-III for tumours that involve the periacetabulum and the pubis [5].

6.2 Classification of Internal Hemipelvectomy According to Surgical Margins

Primary tumours are usually resected with a wide bone margin. The anatomic extent of the margin will dictate the classification of the resection. For example, a Type I (iliac) tumour may be resected simply with a resection of the iliac wing. This is referred to as a Type I resection. However, if the iliac tumour is adjacent to the sacroiliac joint and the resection includes part of the sacral alar to ensure that it is oncologically sound, this is referred to as a Type I-IV resection. If a large periacetabular Ewing's tumour extends into the ilium and into the pubic rami, then a resection from sacroiliac joint to symphysis pubis is classified as a Type I-II-III resection. Inclusion of muscle as a margin with the resections may also be denoted as a subclassification of the resection [5].

6.3 Indications and Contraindications for Limb-Sparing Surgery

Limb-sparing surgery is indicated if oncologic margins can be achieved with a reasonable chance of success and if the surgery and reconstruction leaves the patient with a functional lower limb and adequate soft tissue cover of the reconstruction.

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Limb-sparing surgery is contraindicated in the setting of pre-existing sepsis, if the soft tissue component renders safe dissection around the major neurovascular structures impossible without violation of the tumour boundaries or if there is systemic metastases, which is progressive or has little prospect of control. Resection of either the sciatic nerve or the hip joint with internal hemipelvectomy is permissible but not both. In the presence of any of the contraindications, if appropriate, careful consideration should be given to external hemipelvectomy.

6.4 Availability of Resources

Major pelvic resections and management of subsequent complications cannot be optimally achieved without a trained and coordinated team. Additionally, intensive care facilities, an experienced anaesthetic team, ready access to a blood transfusion services and excellent ward facilities are mandatory.

6.5 Preoperative Preparation

6.5.1 Staging Investigations

Complete preoperative staging investigations are mandatory prior to major pelvic surgery. These include plain radiographs, whole body bone scans, computed tomography of the pelvis and lungs, magnetic resonance imaging of the pelvis and functional nuclear scans to assess metabolic activity of the tumour. These are required to determine the local and systemic extent of disease and the response of the tumour to neoadjuvant therapy and to plan surgical margins. The immediate preoperative local imaging on which surgical margins are based should include both anatomic and functional imaging. If there has been an unsatisfactory response to chemotherapy as reflected by functional scans or an increase in tumour size, surgical margins should be based on imaging obtained prior to the commencement of neoadjuvant treatment and should be far wider than if the tumour was a good responder [7]. This allows for the widest possible margin. In the event that neither a wide margin is possible nor a good response anticipated, consideration should be given to external hemipelvectomy [7].

Of note, magnetic resonance imaging is particularly important when tumour and major neurovascular structures such as the lumbosacral trunk, the internal iliac vessels, the sciatic nerve and the gluteal vessels in the greater sciatic notch are adjacent to each other. Magnetic resonance imaging is also excellent for assessing the intraosseous extension of tumour and therefore has a key role when planning surgical margins.

6.5.2 Blood Transfusion Requirements

Pelvic resections are often associated with considerable blood loss. Cross-matching should be performed early to ensure availability of blood as soon as it is required and also to identify any abnormal antibodies in the recipient. Recommendation for initial blood cross-matching includes 10 units of packed cells, 5 units of fresh frozen plasma and 5 units of platelets. Further blood requirements may be determined on intraoperative testing of patient haemoglobin. Preparation of relevant equipment to allow rapid transfusion of warmed blood should be undertaken [8]. Transfusion of high volumes of cold blood may induce a coagulopathy [8].

6.5.3 Preoperative Optimisation

Patients on chemotherapy or receiving radiotherapy may experience a reduction in health status. Even large inflammatory-type sarcomas may induce profound constitutional upset. Common effects of neoadjuvant chemotherapy include a fall in peripheral blood counts resulting in anaemia, neutropenia and thrombocytopenia. Radiotherapy may also cause marrow suppression with similar effects. GM-CSF is a commonly used drug to stimulate bone marrow activity. Renal and cardiac function may also suffer during chemotherapy, and these need to be closely monitored and optimised prior to surgery. Renal and cardiac function may be stressed under conditions of excessive blood loss or rapid high-volume blood transfusion. Close communication between surgical and medical oncology teams is necessary to plan the optimal timing of surgery, which should be past the nadir of marrow suppression. The inclusion of a perioperative physician in the team is critical, and they should communicate closely with the anaesthetic team to ensure the patient is fit for anaesthesia and surgery.

6.5.4 Informed Consent

It is critical that patients and their carers understand the full extent of surgery, the anticipated outcomes and the attendant risks of complications. Furthermore, they should also understand the prolonged course and duration of post-operative rehabilitation and recovery. It is not uncommon for recovery to take between 1 and 2 years to stabilise. Rarely do patients ever experience normal gait and function after major pelvic surgery [9–12]. Moreover, the cosmetic impact of the scar and also change in habitus can be a source of concern and anxiety for the patient. If patients understood the material risk of complications, they may elect external hemipelvectomy over limb-sparing surgery.

6.5.5 Urinary Catheterisation

Insertion of ureteric stents prior to the commencement of surgery is recommended when operating on large pelvic tumours. This allows easy identification of the ureters, which may otherwise be hidden under the overhang of a large soft tissue component. Moreover, the use of radiotherapy may cause induration of the pelvic mesentery, making the ureter indistinguishable from vessels, nerves and thickenings within the retroperitoneal tissue. These are placed endoscopically and secured to a bladder catheter which is inserted at the same time. Bladder catheters also aid identification of the bladder neck and urethra which can be damaged when operating on tumours at the front of the pelvis.

6.5.6 Prophylactic Antibiotic

Post-operative sepsis is a risk of prolonged pelvic surgery. The extensive dissection, reduced immune state of the patient and use of biologic or prosthetic implants increase the vulnerability of the patient to infection. Broad-spectrum combination antibiotic therapy should be given as a prophylaxis and continued until all drains are removed. Antibiotic coverage that is effective against gram-positive and gram-negative organisms is recommended and may include vancomycin and ceftriaxone which should be given preoperatively to allow circulation prior to the commencement of surgery. Vancomycin should be given slowly over 30 min to avoid a vascular reaction sometimes referred to as "red man" syndrome.

6.5.7 Anaesthetic Considerations

Pelvic surgery may result in wide cardiovascular variations [8]. These may occur from acute or persistent blood loss, embolus, cardiac arrhythmia or failure, intra-abdominal or intra-pelvic vascular compression or the effects of spinal anaesthesia. Invasive central venous and arterial monitoring allows for early and accurate diagnosis of haemodynamic upset and quick volume replacement. No pelvic surgery should be performed without this. In addition to vascular monitoring, some institutions now recommend periodic intraoperative transesophageal echocardiography to visualise the function of the heart. This is a skill acquired by anaesthetists and can be performed without interfering with ongoing surgery. In some institutions, regional anaesthesia is combined with general anaesthesia to allow a reduction in the depth of general anaesthesia and to facilitate better post-operative control of pain.

6.6 Surgical Technique: Exposing the Pelvis

6.6.1 Patient Position and Draping

The patient is held in a "floppy" lateral position with loosely applied supports against the thoracic spine and sternum. These positions of restraints allow clear access to the abdomen and buttock as far back as the sacrum, and the loose restraint of the patient permits the surgical team to roll the patient forward and backward to improve visualisation of the intra- and extra-pelvic anatomy. A strap on the bottom leg secures it in position to avoid slippage when rotating the body backward or forward. A thoracic arm rest keeps the upper arm free for the anaesthetist, and an axillary support lifts the chest off the bottom arm to avoid vascular obstruction or nerve compression. Careful positioning of padding should be undertaken to protect any pressure areas which may be compromised during a long operation. The abdomen up to mid-chest is draped free to allow access to the entire abdominal field down to the symphysis pubis, and the ipsilateral limb is draped free to allow manipulation during surgery.

6.6.2 Incision

The most extensile incision is a curved ilio-femoral incision. This incision maps out the iliac crest from posterior iliac spine to anterior iliac spine. This may be continued posteriorly to the midline of the lumbar spine and then dropped vertically to allow the creation of a flap which will expose the underlying sacrum. Distal to the anterior superior iliac spine, the incision passes a few inches in front of the greater trochanter before sweeping slightly distally, and posteriorly, a hand span distal to the centre of the greater trochanter. This allows the creation of a buttock flap which will expose the sciatic notch, the spine of the pelvis and the ischial tuberosity. For a more extensile approach to the anterior pelvis, an additional incision beginning at the anterior superior iliac spine then passes along the line of the inguinal ligament toward the pubic tubercle (Fig. 6.1). The addition of an inguinal incision creates a tri-radiate incision, and there is a risk that the points of the skin flaps may undergo skin necrosis. To avoid this, the inguinal flap created by the inguinal incision should include the underlying sartorius muscle which provides a vascular supply to the point of the flap. Another method for reducing the risk of wound edge necrosis is to extend the medial bulge of the ilio-femoral incision so that it passes over the junction of the lateral and middle thirds of the inguinal ligament. This

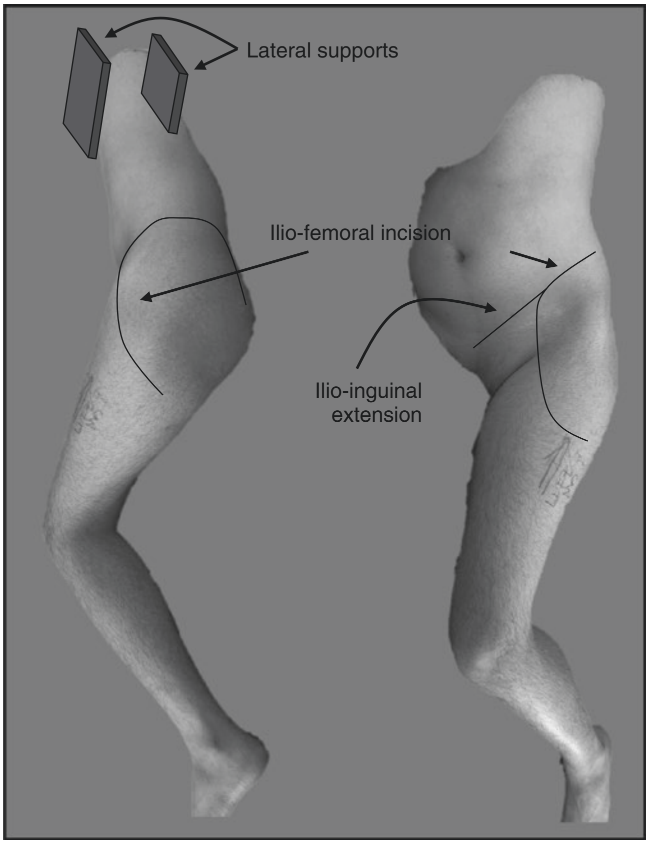

Fig. 6.1 Incision

will allow a reasonable view of the anterior pelvis without creating flaps with points that have an acute angle.

The skin incision is carried down to and through the deep fascia. This creates a posteriorly based gluteal flap that survives on the inferior gluteal vessels. Pelvic tumours that are covered by the gluteus medius and minimus muscles spare the gluteus maximus muscle.

If an anterior-based pelvic flap is required, the iliac incision stops three to four fingers before the anterior superior spine before passing posteriorly and laterally to the anterior superior iliac spine distally to the greater trochanter and then laterally down the line of the femur as far as is required (Fig. 6.1). The supply of the anterior flap comes from the superficial femoral artery and the obturator artery. The medial skin incision follows the line of the medial hamstring musculature and then is joined by a transverse anterior thigh limb that meets the lateral incision described previously.

6.6.3 Exposure of the Posterior Pelvis

Unless the tumour is extending posteriorly through the buttock and surgery in this area needs to be avoided, posterior exposure of the pelvis is always required to secure the sciatic notch. The purpose of securing the sciatic notch is to preserve the inferior gluteal vessels; protect the sciatic nerve; identify, protect and sweep the internal pudendal neurovascular structures off the spine of the pelvis; and ligate and divide the superior gluteal vessels as they pass upward from the notch closely applied to the bone.

The deep fascia is incised off the iliac crest from posterior to anterior superior iliac spines. The split between the sartorius muscle and the tensor fascia lata is identified and incised and the two muscles separated. The sartorius is kept on the medial side of the skin incision. The gluteus maximus muscle is strongly attached to the fascia lata and the outer border of the linea aspera below the greater trochanter. This strong attachment is divided to allow the muscle flap to be released and reflected posteriorly. This exposes the sciatic nerve along the posterior thigh up to the sciatic.

The sciatic nerve lies posterior to the inferior gluteal vessels at the notch. This space is confined and may be further compromised by the presence of adjacent tumour. If not careful, inadvertent injury of the vessels at the notch may result in retraction back into the notch making haemostasis a challenge.

Division of the sacrospinous and sacrotuberous ligaments expands the portal of the sciatic notch which improves exposure of important structures as well as allowing admission of several fingers into the true pelvis.

6.6.4 Division of the Sacrospinous Ligament

The sciatic notch is guarded anteriorly by the point of the spine of the pelvis. The sacrospinous ligament is a very strong and round condensation of tissue that passes from the sacrum forward to the tip of the spine of the pelvis. It may extend up to 1 cm or more along the spine from its tip. The internal pudendal neurovascular structures pass out of the notch near the tip of the spine before passing medially back into the pelvis once they have crossed the ligament (Fig. 6.2). To avoid injuring these structures, the sacrospinous ligament should be elevated off the base of the spine of the pelvis and dissected off the tip with a periosteal elevator. By sweeping the ligament off the tip of the spine, the internal pudendal structures are also safely swept free and allowed to drop free back into the pelvis and away from danger.

6.6.5 Division of the Sacrotuberous Ligament

The sacrotuberous ligament passes from the inferior sacrum forward and downward to the ischial tuberosity (Fig. 6.2). A periosteal elevator can be used to sweep the soft tissue off the ligament before it is isolated between two Hohmann retractors and divided under direct vision. The sacrotuberous liga-

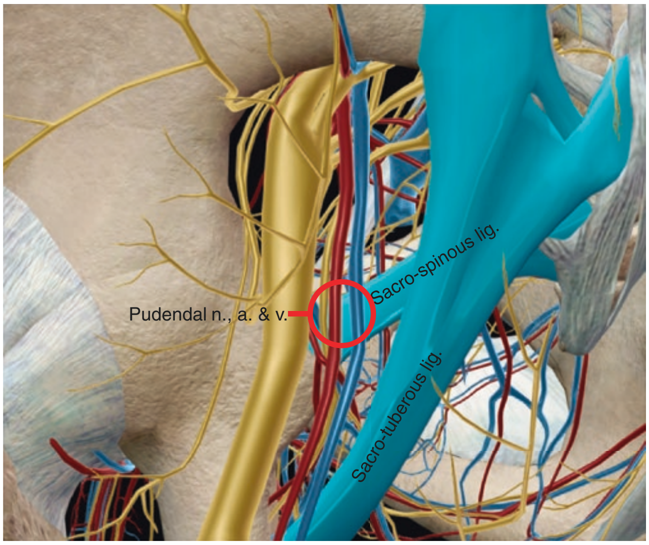

Fig. 6.2 Sciatic notch

ment can be divided at its attachment to the ischial tuberosity. When both the sacrospinous and sacrotuberous ligaments are released, there is greater room for dissection and protection of vessels and nerves that pass out of the pelvis. This part of the operation should be done early. Type II and II-III resections require division of these structures or the resected bone will not rotate or be released after the osteotomies.

6.6.6 Exposure of the Inner Wall of the Pelvis

The tri-laminar muscle layers of external oblique, transversalis and internal obliques can be incised over a finger using electrocautery. Beginning midway along the iliac crest is a good site to breach these layers and gain entry into the retroperitoneum before inserting two fingers into the rent in the tri-laminar layer and sweeping off the retroperitoneal fat and other structures prior to dividing the muscle layer safely over the finger which tents the overlying muscles. The inferior epigastric artery is an important structure, which can be encountered along the lateral border of the rectus abdominis muscle. This vessel should be formally ligated and divided or alternately protected if a rectus flap is required for closure of any defects.

Once in the retroperitoneal space, the fat within the space can be swept medially and held medially with a moist pack and abdominal retractors. The main structures which come into view are the psoas as it passes down medial to the iliacus muscle, the external iliac vessels which pass anterior and medial to the psoas and the internal iliac vessels which can be identified more proximally along the external iliacs as they pass downward and backward toward the notch. Typically, the external iliac vein is much larger than the artery, and there may be several sizeable branches exiting the sciatic notch. Once the internal and external iliac vessels have been identified, vessel loops should be applied around each major structure. The psoas major muscle can be quite prominent, and its size may obstruct a good view of the lesser pelvis. Segmental division of the psoas and its removal from the operative field will give an excellent view along the pelvic brim into the lesser pelvis. Doing so also allows the femoral nerve that passes between the psoas and the iliacus to be protected and the lumbosacral trunk to be visualised.

Good exposure of the posterior pelvis is required to safely ligate and divide the many vessels that pass across the lumbosacral trunk and the sciatic nerve. With release of the sacrospinous and sacrotuberous ligaments, the space of the sciatic notch is expanded to admit fingers together with blunt dissection from within that can help to clear tissue away from the sciatic nerve and vessels and these structures from the bony margins of the notch.

6.6.7 Superficial Femoral Vessels

The inguinal ligament is freed from its attachment to the anterior superior iliac spine and retracted medially to expose the femoral nerve, artery and vein, from lateral to medial. It is important to note that an investing fascia holds the vessels securely from the retroperitoneum as they pass between the inguinal ligament above and the underlying pectineus muscle over the superior pubic ramus. Careful dissection and application of vessel loops will help to handle these structures without inadvertent injury.

6.6.8 Femoral Nerve

The femoral nerve is easily found between the iliacus and psoas muscles under the iliacus fascia where it can be lifted free and a nerve loop applied to allow easier handling. It passes freely down to and under the inguinal ligament lateral to the superficial femoral artery.

6.6.9 Bladder

The bladder is easily palpated posterior to the pubic rami and can be swept to the opposite side and protected with a pack and retractor. The urethra passes from the bladder neck under the arch of the pubis and needs to be protected particularly if dissecting behind the symphysis pubis. A crest of bone on the inner surface of the pelvis in the midline anteriorly marks the symphysis pubis. By remaining close to the symphysis from inside and outside the pelvis and also using a right angle forceps to stay close to the symphysis at the pubic arch, the symphysis can be divided either by electrocautery under direct vision or by using a wire saw.

6.6.10 Pelvic Osteotomy

Pelvic osteotomies are performed either through the lateral sacral alar, through the sciatic notch, across the inferior and superior pubic rami or through the symphysis (Fig. 6.3 - yellow dashed lines). Osteotomy of the sacral alar is the most challenging as the front of the sacrum needs to be cleared of nerve and vessels (Fig. 6.3) and the line of the osteotomy clearly identified. The ilio-lumbar vessels must be sought (Fig. 6.3 - solid black arrow) and ligated as they can cause considerable bleeding if inadvertently lacerated (Fig. 6.3). Branches of the internal iliac vessels (Fig. 6.3 - white solid arrows) need to be identified and tagged with a loop to ensure that these are protected from injury during the dissection of the inner surface of the sciatic notch and also when clearing other branches around the lumbo-sacral plexus. The posterior sacral alar requires the clearing of all the soft tissue from this. The sacroiliac joint is identified by a definite ridge along the inner wall of the posterior pelvis, and the osteotomy must pass medial to this. It is easier to begin the osteotomy from the external surface of the sacrum by creating a groove with a burr or bone rongeurs and continuing the removal of bone until the anterior cortex of the sacrum is identified. This can be appreciated by percussing the depths of the groove with a periosteal elevator. The completion of the osteotomy then can be easily performed using a broad osteotome to crack the inner table from within.

Osteotomy through the notch, pubic rami and symphysis may be undertaken under direct vision using a wire saw. The approach to the osteotomy of the ischium is best done after disarticulating the hip and clearing soft tissue medial to it as this gives the best view and appreciation of the ischial tuberosity and the obturator foramen through which the wire saw or retractors can be passed.

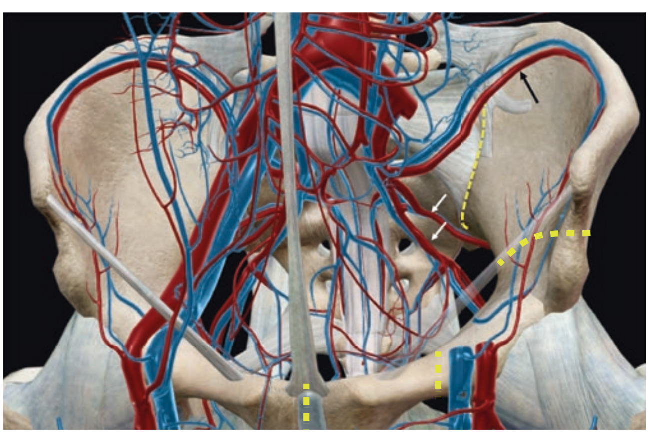

Fig. 6.3 Pelvic osteotomies

6.7 Post-operative Management

The post-operative management must be clearly planned with the aim of minimising complications, promoting healing of soft tissue and protecting union of any graft sites.

6.7.1 Bed Rest

Most patients require 1–2 weeks of bed rest to overcome the pain of surgery and the discomfort of movement. Adequate and appropriate analgesia should be charted to facilitate recovery. During this time, close attention should be paid to the skin flaps to identify early any skin edge necrosis. If present, this should be treated early with operative debridement and resuture undertaken.

6.7.2 Blood Loss

With the bone cuts and dissection and reperfusion of soft tissue, ongoing bleeding of up to 30% of the intraoperative volume of blood loss may occur over the next 12–24 h after surgery. As patients may already be marrow suppressed, ensuring adequate haemoglobin levels is important. Coagulopathies are also common, and close monitoring of coagulation profiles is advisable in the first 2 h, and this should be combined with appropriate and adequate replacement of platelet and plasma products. This is particularly important if the patient has received significant transfusion volumes intraoperatively. It is also advisable to apply a soft hip spica constructed from Velband and crepe bandaging to allow ongoing compression of the operative site as well as to act as a tamponade against an expanding abdominal volume. Large-bore drain tubes should be used in all dead spaces and checked regularly.

6.7.3 Thromboprophylaxis

Deep vein thrombosis is common and should be avoided. Options for this include the application of lower limb compression devices and the use of low-molecular-weight heparins. Early ankle mobilisation should be encouraged. Occasionally, inferior vena cava filtering devices may be indicated in the presence of a pre-existing lower limb or pelvic thrombosis. Chemical thromboprophylaxis should be commenced after confirmation that a coagulopathy does not exist.

6.7.4 Antibiotic Therapy

Antibiotic therapy should be continued till at least the drain tubes are removed. Institutional practices may warrant ongoing use after this.

6.7.5 Mobilisation

Post-operative mobilisation should occur under supervision and may require brace support following any pelvic reconstructions. The extent to which weight bearing may be allowed will depend on the reconstruction. The use of platform frames and four-point walking frames can be very helpful. From time to time, solid spicas may be required, and these should be carefully applied to allow sitting and standing as well as to compensate for any acute abdominal swelling.

6.8 Early Surgical Complications

6.8.1 Haemorrhage

Acute heavy intraoperative bleeding is common with pelvic resections. Careful preoperative assessment of the vascularity of the tumour with appropriate imaging, involvement of a vascular surgical team for consultation prior to surgery and also during the time of surgery and preoperative embolisation are ways of reducing this risk. Osteotomies should be done last, after completion of as much of the soft tissue dissection as possible as cut bone will continue to bleed through the duration of the operation. The application of bone wax to the surface of cut bone or the use of argon gas spray cautery may also assist with reducing haemorrhage. Specific sites where bleeding may come include the sciatic notch, branches of the internal iliac vessels as they approach the sciatic notch, the venous plexus around the base of the bladder and the obturator vessels as they pass through the obturator foramen.

6.8.2 Infection

Infection is a high risk of pelvic surgery. Prolonged exposure, implantation of devices or biologic material or injury to viscera may lead to devastating infection. If infection is proven, all attempts must be made to bring this under control including washout and debridement to allow early reinstitution of chemotherapy if this is required.

6.8.3 Nerve Injury

Injury to the sciatic and femoral nerves is common with pelvic resections and the majority follow traction. The femoral nerve is injured when traction is applied while trying to clear the superior pubic ramus. The sciatic nerve may be injured where it is held to the bone anterior to the sacrum. Twisting of the resected bone segment without first ensuring the nerve is free is another potential cause for nerve injury. Retraction of the nerve while trying to achieve haemostasis is another potential cause for injury. Nerve injury should be avoided at all cost because of the instability it may cause to the lower limb and the hip joint.

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Resection of Periacetabular Lesions

7

Howard Y. Park and Francis J. Hornicek

7.1 Introduction

Peri-acetabular pelvic resections are demanding operations with complex indications, anatomy, and postoperative rehabilitation. Sir Gordon Gordon-Taylor of Britain in 1935 called hindquarter amputations "one of the most colossal mutilations practiced on the human frame." [1] It was attempted with and without success prior to the turn of the twentieth century with Girard of Berne documenting the first nonfatal pelvic resection for sarcoma in 1895 [2]. As the knowledge base of pelvic anatomy, oncology, and imaging technology grew, more attempts at hemipelvectomy were made, and various techniques were developed. The application of cross-sectional imaging as well as the rise of metallurgy and implant development in the 1970s expanded the indications of this operation.

Internal hemipelvectomy with limb salvage or reconstruction has become an essential operation in the armamentarium of surgical oncologists. The goals of surgery are often negative or noncontaminated surgical margins while trying to achieve maximal function. The pursuit of maximal function has often favored internal hemipelvectomy in concept as opposed to external hemipelvectomy also known as hindquarter amputation. Primary musculoskeletal tumors, metastatic lesions, trauma, and infection of the pelvis are among the common indications for this relatively uncommon procedure. The relative contraindications for the limb-sparing procedures around the pelvis have changed in the past decade. The sciatic nerve can be sacrificed while still keeping the lower extremity. Some general surgeons have not realized that involvement of the acetabulum with direction extension from a carcinoma is not a contraindication for removal.

The scope of this discussion is focused on Enneking and Dunham Classification Type 2 hemipelvectomy, otherwise known as peri-acetabular resection [3]. In comparison to Type 1 (resections about the ilium) and Type 3 (resections about the pubis), Type 2 peri-acetabular resections are relatively more challenging with regard to resection and reconstruction [3, 4]. However, careful patient selection, scrutiny of cross-sectional imaging, and evolving reconstruction techniques have collectively improved the complication profile and survival of this relatively morbid operation.

7.2 Indications

Hemipelvectomies occur with relative rarity with rough estimates approximating 1 per 1 million persons annually [5]. In their seminal paper, Enneking and Dunham described resection in patients with sarcoma of the innominate bone that failed to be treated with radiation or chemo, no evidence of metastasis, and with preoperative imaging indicating that the anatomical location of the lesion would permit resection [3]. In principle, the aforementioned indications for hemipelvectomy remain today, but advances in reconstruction and cross-sectional imaging have sophisticated and widened these indications.

Primary bone tumors of the pelvis account for approximately 15–20% of all bone tumors with osteosarcoma, chondrosarcoma, and Ewing's sarcoma constituting nearly 50–80% of all pelvic primary bone tumors [6, 7]. Osteosarcoma and Ewing Sarcoma are most often diagnosed in adolescent and young adult patients where chondrosarcoma is most often diagnosed in older patients between 40 and 75 years of age [6]. Other indications include fibrosarcoma, Langerhans cells histiocytosis, aneurysmal bone cyst, giant cell tumor, and fibrous dysplasia, although these entities require pelvic resection with much less frequency [7]. When they occur, giant cell tumor and aneurysmal bone

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cysts have a predilection for localizing about the acetabulum [8]. Metastatic lesions to the pelvis can originate from various sources, most commonly the breast, lung, prostate, kidney, and thyroid. However, many of these lesions can be effectively managed with radiation or chemotherapy, and only a minority of metastatic lesions indicate pelvic resection [9, 10]. Infection and trauma to the pelvis have been described as indications for hemipelvectomy, although they are less likely to specifically indicate a peri-acetabular resection.

7.3 Relevant Anatomy

The complex anatomy of the pelvis requires vigilance and experience from the surgeon in order to navigate the osseous pelvis, muscular attachments, intrapelvic contents, and viscera. The pelvis can be compartmentalized into the ilium, acetabulum, and pubic rami or obturator rings. The Enneking and Dunham classification of pelvic resections is based on these specific anatomical locations of resection: resections of the ilium are Type 1, resections of the peri-acetabular region are Type 2, resections of the pubic rami or obturator rings are Type 3, and resections of the sacrum are Type 4 [3]. Posteriorly within the pelvic ring, the sacroiliac joint is bounded by the sacrospinous and sacrotuberous ligaments, which are among the strongest ligaments in the body. The iliac wings extend from the sacroiliac joint, in which the inner table serves as an attachment for the iliacus muscle which eventually joins with the psoas muscle to form the iliopsoas tendon inserting into the lesser trochanter.

The sciatic, femoral, obturator, and lumbar plexus sensory nerves are vital structures to be identified and protected when operating about the pelvis. The sciatic nerve (with the inferior gluteal artery) is transmitted through the greater sciatic notch inferior to the piriformis muscle although 10% of patients have a sciatic nerve that transmits within the piriformis. The femoral nerve (with the femoral artery) lies superficial to the iliacus muscles and courses underneath the inguinal ligament to enter the anterior compartment of the thigh. The obturator nerve courses through the iliopsoas muscle over the sacral ala and is transmitted into the medial thigh through the obturator foramen.

The pelvic vasculature requires careful consideration during pelvic resections. The aortic bifurcation into two common iliac vessels occurs at approximately the L4 spinal level. The common iliac arteries then bifurcates into the internal and external iliac vessels at approximately the S1 spinal level, although these levels can vary especially in the cases of space-occupying tumors. Characterization of these vessels in cross-sectional imaging or preoperative angiography is crucial for preoperative planning as tumor can abut, displace, or encase these vascular structures. An anastomosis of the external iliac and obturator artery is known as the corona mortis which occurs in a third of patients [11]. When present, it is often found within 3–7 cm of the pubic symphysis and requires careful handling as damage to this vessel can lead to profound blood loss. The ureter courses through the retroperitoneum medial to the psoas major muscle, and it traverses medially at the level of the common iliac bifurcation, eventually inserting into the bladder.

7.4 Imaging Studies and Preoperative Planning

As aforementioned, pelvic anatomy is complex, and it is made more challenging when tumors displace structures and anatomic landmarks. Therefore, preoperative scrutiny of cross-sectional imaging is required for safe pelvic resection. Plain x-ray is limited in the setting of hemipelvectomy relative to cross-sectional imaging as it is unable to provide details that would aid in resection and reconstruction. Oftentimes, tumors can be obscured by osseous structures and underestimate the extent or size of lesions. However, they can be of utility when planning reconstructions with regard to leg lengths.

Computer-aided tomography (CT) and magnetic resonance imaging (MRI) with intravenous contrast should be completed on all patients who are under consideration for pelvic resection. CT scans have the ability to elucidate osseous details including the extent of bone involvement and extension to pelvic viscera. Contrast enhancement of tumor and vascular structures can provide crucial information for safe resection. MRI is useful in assessing the soft tissues and tumor size. Vessels, nerves, and muscles are best visualized with MRI, and it is essentially a required study in the preoperative assessment of pelvic lesions. In general, cross-sectional imaging should be scrutinized for the location and size of tumor as well as vascular and viscera involvement in order to aid resection.

Other adjunct imaging studies can be utilized including bone scan, vascular studies, and fluorine-18 2-fluoro-2-deoxy-D-glucose-positron emission tomography (FDG-PET). Three-phase bone scans can give valuable information regarding foci of metastases and blood flow to tumors. Vascular studies are critical when space-occupying lesions have distorted the vascular anatomy, and embolization of tumor vasculature can aid in reducing blood loss and defining tumor margins. FDG-PET studies are sensitive to metabolic demands of tissue, which can localize tumors and give information regarding the responsiveness to chemo or radiation therapy.

Biopsy of the lesion is often performed with a needle biopsy technique as pelvic tumors are often deep within the pelvis, precluding open biopsy. If needle biopsy is performed, minimizing the risk of contamination is critical, and the biopsy should follow a plane of future resection in consultation with an orthopedic oncologist.

7.5 Surgical Technique

Type 2 resections of the peri-acetabular region are among the most challenging orthopedic procedures, and it is associated with the highest complication rates compared to Type 1 or Type 3 resections. A utilitarian pelvic incision is utilized, which courses along the pelvic brim and anterior superior iliac spine and, in the case of Type 2 peri-acetabular resections, extends laterally down the thigh. Careful dissection consistent with an ilioinguinal approach is performed to expose the anterior and posterior aspects of the pelvis. Major nerves and vessels as aforementioned in this chapter must be identified and protected throughout the approach and resection. The ilium/supra-acetabular osteotomy is performed once the femoral and external iliac vessels are identified and protected. A myocutaneous flap is developed with the gluteus maximus muscle to access the retrogluteal contents which include the ilium, sciatic notch, and hip joint. The ischium is osteotomized above the level of the hamstring attachment. Portions of the pelvic floor and sacrospinous ligament are released to resect the peri-acetabulum.

7.6 Reconstruction Options and Outcomes

There exist multiple options for reconstruction of peri-acetabular resections including resection arthroplasty, total hip arthroplasty with massive alloprosthetic reconstruction, saddle prostheses, and various custom devices. The remaining pelvis after a Type 2 resection includes the ilium and obturator rings with discontinuity between those two structures. This void eliminates the pelvic hip articulation which can result in significant disability for the patient. Reconstruction options have attempted to address this disability in an effort to maximize function following peri-acetabular resections, although complication rates are high and approach 20–60% [12–20].

7.6.1 Resection Arthroplasty

Resection arthroplasty avoids complications associated with reconstruction. The Friedman-Eilber resection arthroplasty was described in 1979 which consisted of an internal hemipelvectomy and proximal femur resection followed by soft tissue closure. Several studies have reported that the instability due to this flail hip leads to poor ambulation and inferior patient acceptance [21–25]. However, other studies have shown acceptable function can result and has the distinct advantage of avoiding the complication profiles of prosthetic reconstruction [26, 27]. Modifications on the technique to achieve ilium-proximal femur fusion and hip transposition have been made in order to avoid flail limb with some success [28, 29]. Beadel et al. suggest that iliofemoral arthrodesis can be attempted if bone loss to the ilium is less than 5 cm and the femoral head is conserved [13]. Resection arthroplasty results in leg length discrepancies which can be overcome with shoe lifts [18, 26].

7.6.2 Pelvic Reconstruction with Total Hip Arthroplasty

Total hip arthroplasty with alloprosthetic pelvic reconstruction is one option for peri-acetabular reconstruction. Beadel et al. reported that if patients did not subsequently develop an infection, functional results were "reasonable." [13] However, the infection rate approached 50% of reconstructed patients in their series [13]. In their series of 147 patients who underwent pelvic resections for pelvic sarcoma, Puchner et al. found that endoprosthetic reconstruction and high-volume tumors were significant risk factors for experiencing a major complication and infection [30]. Several studies have reported results on extracorporeal irradiation (ECI) and reimplantation of peri-acetabular resections with subsequent total hip arthroplasty with varying results [3–33]. A recent study of 23 patients with Type 2 resections below the anterior superior iliac spine treated with ECI and total hip arthroplasty had very good functional results, no recurrences, one dislocation, and no infections at an average of 21 months follow-up [34]. The authors attributed these promising results to their specific patient selection which was based on the levels of resection to maximize bone union after reconstruction [34].

7.6.3 Saddle Prosthesis

Saddle prostheses implant into the femoral canal and attach to the remaining ilium. The femoral preparation and implantation are, in general terms, similar in concept to femoral stems in total hip arthroplasty or hemiarthroplasty. The proximal saddle portion attaches to the ilium and is modular to help achieve correct leg lengths. Retention of hip musculature including the iliopsoas and abductor muscles is important for function and stability of the prosthesis. The clinical results of this procedure are mixed at best. Long-term results for saddle prosthetic reconstruction report poor restoration of function and substantial morbidity [35]. In contrast, Aboulfaia reported good to excellent results in 12 of 17 patients treated with saddle prostheses for peri-acetabular resections [36]. Another study noted that saddle prosthesis could provide early weight-bearing and reduce leg length discrepancy with fair functional results [37], and other studies have reported satisfactory functional outcomes [38]. However, there is a high complication rate and questions regarding the longevity of this device as proximal migration leading to limb shortening, infection, and hardware failure are common which have led some institutions to abandon this reconstruction technique [39]. From a case series of 15 patients, Renard et al. posited that results were satisfactory if certain contraindications to saddles prostheses were taken into consideration which were osteoporosis, iliac involvement, and poor soft-tissue quality [40].

7.6.4 Custom Pelvic Implants

Custom-made implants based on preoperative CTs constructed by manufacturing companies have also been utilized to reconstruct peri-acetabular voids. Rudert et al. found that in their series of 38 patients, revisions were required in 52.6% of patients with a 21% deep infection rate with poor mobility and patients requiring walking aids [41]. When applying CT-based osteotomy guides and custom implants for peri-acetabular resections, acceptable functional and oncologic outcomes were reported although with 62.5% of patients experiencing at least one complication and 25% of patients experiencing an infection [20]. The MUTARS system (Implantcast, Buxtehude, Germany) has been utilized in Germany for the last two decades with acceptable functional and oncologic outcomes but at the risk of implant loosening and infection [42]. Promising results were reported from China where modular hemipelvic prostheses for peri-acetabular resections resulted in a 14% complication rate with only one dislocation [43]. Other reports from China have also reported promising results with modular hemipelvic prostheses [44–46] as well as a report utilizing bulk femoral head autograft for pelvic reconstructions [47].

7.6.5 Pedestal Cup Prosthesis

Recently, pedestal cup prostheses have been applied in Europe with similarly mixed results in comparison to other reconstruction options. Pedestal prostheses are implanted into the ilium with a distally protruding cup to articulate with a femoral component. Bus et al. reported complications in 15 of 19 patients with dislocations, loosening, and infection as the leading causes which led them to advise caution in utilizing this construct [48]. More recently, the LUMiC prosthesis (Implantcast, Buxtehude, Germany) has been utilized in Europe which is a pedestal cup prosthesis. This prosthesis is unique in that it is coated silver to prevent infection, although infection occurred in 28% of patients. Mechanical complications occurred in 30% of patients with dislocation being common; however, once a dual-mobility cup was utilized, the dislocation rate reduces substantially to 4% [49]. The mid- to long-term results of this implant have yet to be reported. Results from the Schoellner cup (Zimmer Biomet Inc., Warsaw, Indiana) revealed similar results in that 15% of patients suffered a dislocation and 17% of patients suffered a deep infection requiring revision [50]. A somewhat similar prosthesis labeled the Ice-Cream Cone Prosthesis also utilized an ilium implant that depends on minimal to no ilium bone loss. In their series of 10 patients with minimum of 2 years follow-up, functional results were fair with wound complications and infections prevalent [51].

7.7 Complications

Hemipelvectomy is associated with a high complication rate that ranges from 20% to 60% [12–20]. Careful scrutiny of advanced imaging can help mitigate intraoperative complications which include intraoperative hemorrhage and damage to viscera or pelvic contents including the ureter, bladder, and bowel. However, pelvic resection is fraught with complex anatomy rendering this a relatively morbid operation. Postoperatively, the most common complications are wound infection and flap necrosis, and deep infections occur frequently with reconstructions. Presumably from the large surgical beds which are often open for extended durations near bladder and bowel, the infection rates following this procedure are high especially in the setting of prosthetic reconstruction. Aljassir et al. reported a 37% infection rate in a review of 27 saddle prosthetic reconstructions, and Abudu et al. reported a 60% complication rate with infection leading all others at 26% [14, 52]. In an analysis of 270 pelvic resections, 166 which were peri-acetabular, the infection rates were 26% in patients who were reconstructed and 15% without reconstruction [53]. Those patients that developed infections often required extensive treatment including conversion to external hemipelvecotomy [53]. Hardware failure, dislocation, and loss of fixation are problematic of reconstructive prostheses which have led some surgeons to abandon reconstruction in favor of resection arthroplasty.

7.8 Conclusion

Peri-acetabular pelvic resections are a relatively rare procedure performed for primary bone tumors, metastatic lesions, and with less frequency, trauma, and infection. Advanced imaging and interdisciplinary care with radiologists, oncologist, and other surgeons are necessary to safely navigate this complex surgery especially in the cases of anomalous anatomy. Due to the resultant disability of peri-acetabular resections, many techniques to reconstruct the hip have been described. However, reconstructive options are associated with a high rate of complications including wound issues and infection leading some to abandon reconstruction in favor of a flail limb. As such, future innovations to enhance mobility and reduce infection rates are necessary to improve the safety and efficacy of this potentially curative procedure.

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8 Pelvic Floor and Pubis Resection

Huayi Qu and Wei Guo

Pubic and ischial tumors were not very common, and most of the pubic or ischial tumor may involve the acetabular region. The pubis and the ischium maintain the continuity of the pelvis and bear less body weight, so no reconstruction is required following the resection of the pubis or ischium because of tumor involvement. The urethra or the rectus is adjacent to the tumor in the pubis or ischium, and care should be taken to avoid injury to these organs. For the elder patients, hernia may happen after the tumor resection because of the weakness of the abdominal muscles. Reconstruction may be required for these patients after extensive tumor resection. Marlex or Lars may serve as the reconstruction materials, and there are also some literatures reporting the utilization of allograft. Plate may be fixed to the ipsilateral acetabular region and the collateral pubis and with cementation, which can serve as an obstruction to prevent the protrusion of the pelvic organs. The disruption of the continuity of the pelvic ring may lead to the detachment of the sacroiliac joint. Mild pain may happen after the weight-bearing, which may also lead to the loosening of the internal fixation, but the normal function of these patients is not affected.

8.1 The Resection of Pubic Tumor

The superior and inferior rami of the pubis and the pubis body can be resected by the perineal approach and the general pelvic approach. But they are not sufficient enough to expose the ischium.

8.1.1 The Procedure of the Resection of Pubis

The combination of the perineal approach and the general pelvic approach is required to expose the pubic tumor. Patients are laid in the lateral decubitus position or lithotomy position.

After the operation, area is disinfected, draping the surgical site with sterile drapes, and the drapes can be fixed to surrounding skin with stitches to avoid the contamination of the incision. The perineal approach, the pelvic approach, or the combination of them may be applied according to the size and location of the pubic tumor.

The incision starts from the anterior superior iliac spine and then runs along the inguinal groove to the pubic symphysis. At the pubic symphysis, the incision curves down to the middle part of the thigh and then runs distally at the surface of the adductor muscle for 5–8 cm.

The abdominal muscles are dissected 1 cm above the inguinal ligament, and rectus abdominis are detached from the pubis. The inferior abdominal vessels are identified and ligated. The visceral organs and the surrounding adipose tissue may be easily pushed away from the pubis, and the line of the pubic symphysis at the inner pelvic side can be palpated. The femoral artery, the femoral vein, and the iliopsoas muscle are mobilized and elevated from the pubis. The sperm cord in the male and the round ligament in the female are identified and protected.

The obturator vessels, which run through the obturator foramen, are ligated from the inner side of the pelvis to lessen the bleeding. All the muscles attached to the pubis are detached to expose the tumor. The inner and outer obturator muscles may be left to ensure an adequate margin.

The hip joint is placed at a flexion status, elevating the femoral artery, the femoral vein, and the iliopsoas muscle to expose the pubis. Jigsaws may be passed through the obturator foramen to perform the osteotomy at the superior and the inferior ramus of the pubis. Dissect the space between the urethra and the pubic symphysis, and place a piece of gauze beneath the pubic symphysis to protect the urethra. Dissect the cartilage of the symphysis and the inner and outer obturator muscles, and remove the tumor.

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Drainage tubes are placed on the inner side of the pelvis and the cavity following tumor removal before closing the incision.

The whole pubis can be resected by this surgical approach. Though some authors reconstruct the defect with fibular or allograft, the related complications without reconstruction are not very common.

8.2 The Resection of the Ischium Tumor

A posterior approach is usually applied to perform the resection of the ischium. If a tumor involved the pubis and ischium at the same time, a general pelvic approach must be combined to perform the resection. For such a procedure, the patient is placed in the lateral decubitus position. For patients with an ischial tumor, patients are placed in the prone position, with the body and the pelvis elevated and the lower limb hanging down naturally. The incision starts from the posterior inferior iliac spine and runs along the inferior border of the gluteus maximum to the greater trochanter. The gluteus maximum is separated along the muscle fiber for the full explosion of the ischium. The superior gluteal neurovascular bundle is identified and protected if possible. The gluteus medius and the gluteus minimus are distracted away from the surface of the ischium. The ischial nerve is mobilized from the tumor, and the whole ischial body can be exposed in the field.

9 Combined Approach for Iliosacral Tumor Resection

Tao Ji and Wei Guo

En bloc resection of iliosacral sarcomas is a surgical challenge. Eight percent to 25% of posterior pelvic tumors may have sacral extension [1–3]. Tumors extending slightly into the sacrum or tumor mass covering IS joint may be excised with an osteotomy through the lateral sacral mass, whereas resection of tumors involving sacral nerves and vertebrae or the lumber spine is more complex. The complex local anatomy and the surgeon's desire to protect neural function may increase the likelihood of inadvertent tumor penetration [4, 5].

Detailed surgical planning is extremely important for iliosacral tumor resection. A multidisciplinary approach should be considered due to visceral involvement. Despite the emphasis on individualized surgical planning, a standardization in surgical management can facilitate surgical planning and procedures. The conventional Enneking classification of pelvic tumors has been useful for the resection of tumors located in the innominate bone. Based on the tumor location and the planned osteotomy site, the authors' institution proposes a novel categorization system for pelvic tumors with sacral invasion, standardized surgical procedures, as well as reconstructive methods.

Differing from the classification designed for tumors localized in the sacrum, the system (Table 9.1 and Fig. 9.1) was devised based on the sagittal extent of sacral invasion rather than the transverse level of sacral invasion of the tumor. Type P-s (abbreviation for "pelvic tumor with sacral invasion") I resection refers to osteotomy through the ipsilateral sacral wing when the tumor only invades the adjacent sacroiliac joint. Type P-s II resection refers to osteotomy through the sacral midline when the tumor invades the ipsilateral sacral foramina. Type P-s III resection refers to osteotomy through the contralateral sacral wing when the tumor invades the contralateral sacral foramina. And type P-s IV resection refers to osteotomy through the contralateral iliac wing when the tumor invades the contralateral sacroiliac joint. Meanwhile, the extent of invasion to the innominate bone is categorized as type "a" when confined to the iliac wing or type "b" when there is invasion in the periacetabular region. The combined approach is usually planned for type P-s II, III, and IV resection (Fig. 9.2).

Depending on whether the ipsilateral sacral nerve foramen is involved, combined approaches or single lateral approach is used. The margin on sacral side is technique demanding and paramount in type IV resection. For the cases in which the sacral foramen or sacral canal is free of tumor invading, the tumor resection can be completed through one semimobile lateral decubitus position. Extended ilioinguinal incision is normally used, and the retroperitoneal space is first exposed. The plane between the iliacus and psoas muscle is developed. The iliacus muscle is transected through its substance. A large posterior gluteal myocutaneous flap is developed, and this allows exposure of the sciatic notch. A Gigli saw is then introduced through the sciatic notch, and ilium osteotomy is performed. Another Gigli saw is placed medial to the iliosacral joint. The lumbosacral trunk, iliac vessels, and ureter should be retracted medially and carefully protected. The osteotomy is then performed through ipsilateral sacral ala lateral to sacral foramen. Part of ilium and whole iliosacral joint is then resected, similar margin on sacral side as that of extended hemipelvectomy. In the circumstance of sacral foramen being involved, single-stage combined approaches should be used (Fig. 9.3). First, the prone position is adopted. A midline incision with one or two caudal branches extending along gluteal maximum is shaped into a curve or reversed Y for the posterior approach to lower lumbar spine and sacrum. The gluteus maximus myocutaneous is developed, and erector spinae are detached from the insertion on sacrum. The sacral nerve roots and dura within the sacral canal are exposed by a laminectomy. Ligation of the ipsilateral S1–S3 nerve roots is obligatory, while the contralateral nerve roots are preferably preserved. The ipsilateral facet joint and accessory articulation of L5–S1 are resected, along with partial intervertebral disc resection. The presacral

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Table 9.1 Definition of the categorization system and suggested surgical managements for each type of resection

	Categorization by sacral involvement (I–IV)			
	P-s I	P-s II	P-s III	P-s IV
Tumor location				
The sagittal extent of sacral involvement	Ipsilateral sacroiliac joint	Ipsilateral sacral foramina	Contralateral sacral foramina	Contralateral sacroiliac joint
Osteotomy site				
Osteotomy site in the sacrum	Ipsilateral sacral wing	Sacral midline	Contralateral sacral wing	Contralateral iliac wing
Osteotomy site in the innominate bone				
P-s a (acetabulum involved)	Ilium			
P-s b (no acetabulum involvement)	Ischium and pubis or pubic symphysis			
Suggested surgical managements				
Surgical position	Lateral	Prone + lateral	*P-s IIIa*: Prone + lateral (or supine) + prone *P-s IIIb*: Prone + lateral (or supine)	*P-s IVa*: Prone + lateral (or supine) + prone *P-s IVb*: Prone + lateral (or supine)
Surgical approach	Anterior[a]	Combined posterior and anterior	Combined anterior and posterior	Combined anterior and posterior
Opening of sacral/ lumbar canal	No	Yes	Yes	Yes
Transection and ligation of dural sac	No	Yes	Yes	Yes
Salvage of sacral nerve roots	All	1. All contralateral sacral nerve roots 2. Ipsilateral S1 nerve root (only when osteotomy is through low sacrum)	Contralateral S1 nerve root (only when osteotomy is through low sacrum)	None

[a]Posterior approach is optional for fixation of contralateral lumbopelvic conjunction in case of lumbopelvic instability

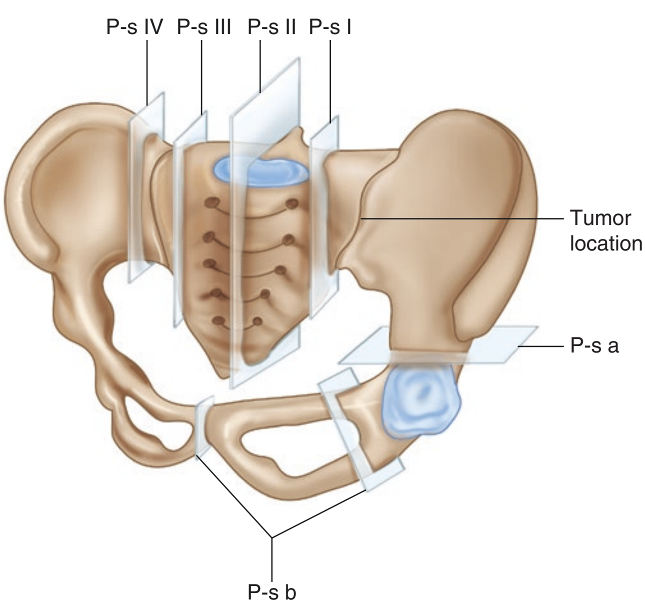

Fig. 9.1 The surgical classification for iliosacral tumor resection by the auhtors' institution. P-s stands for the osteotomy at sacrum and P-s indicates the osteotomy site at pelvis side

space is packed with moist gauze so that the vessels and rectum can be pushed away from the anterior surface of the sacrum. By retracting the severed nerve roots and ligated dural sac to the counter lateral side, sagittal vertebrectomy can be performed posteriorly. The wound is closed temporally with continuous suture. Then the patient is placed in lateral decubitus position. Similar procedure is performed as that of standard type IV resection. The ipsilateral half-discectomy of L5–S1 is performed, and sagittal sacral osteotomy through midline is carried out to complete osteotomy. Then the extended type IV resection is completed.

Farid [4] introduced a method to improve the accuracy of osteotomy for iliosacral tumor resection. High-speed burr was used to perform the osteotomy. At the authors' institution, an ultrasonic osteotome was usually used to perform precise osteotomies. In the prone position, a midline posterior incision and sacral laminectomy allow exposure of sacral roots. The ventral foramina are located directly anterior to the dorsal foramina. A sacral osteotomy begins by dividing the posterior sacral cortex using Kerrison rongeur. The dura and involved nerve roots were ligated. Division of involved roots allows dural retraction and facilitates the vertical osteotomy. This is followed by the cranial osteotomy. If tumor extension spares the upper part of the lateral mass of S1 and the base of the articular process, an oblique cranial osteotomy is performed to preserve lumbosacral stability. Ultrasonic osteotome is used to finalize the osteotomy down to the anterior cortex. Sacral depth substantially increases in

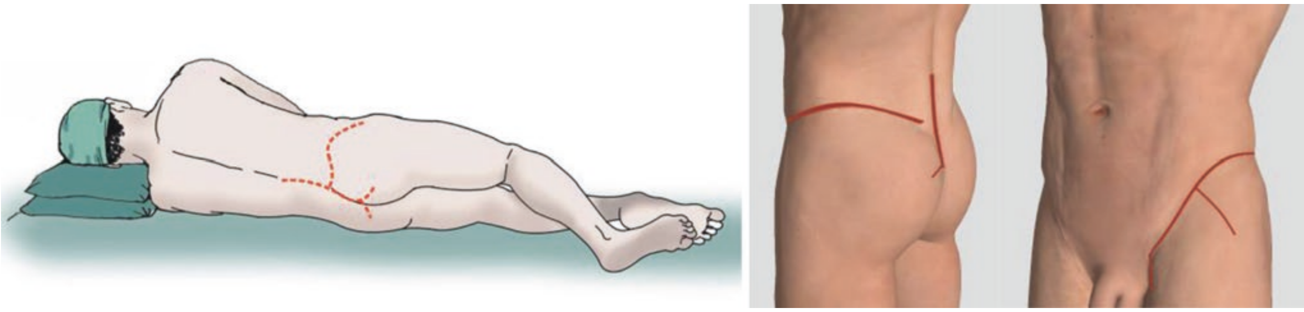

Fig. 9.2 The incisions for combined approach for iliosacral tumro resection

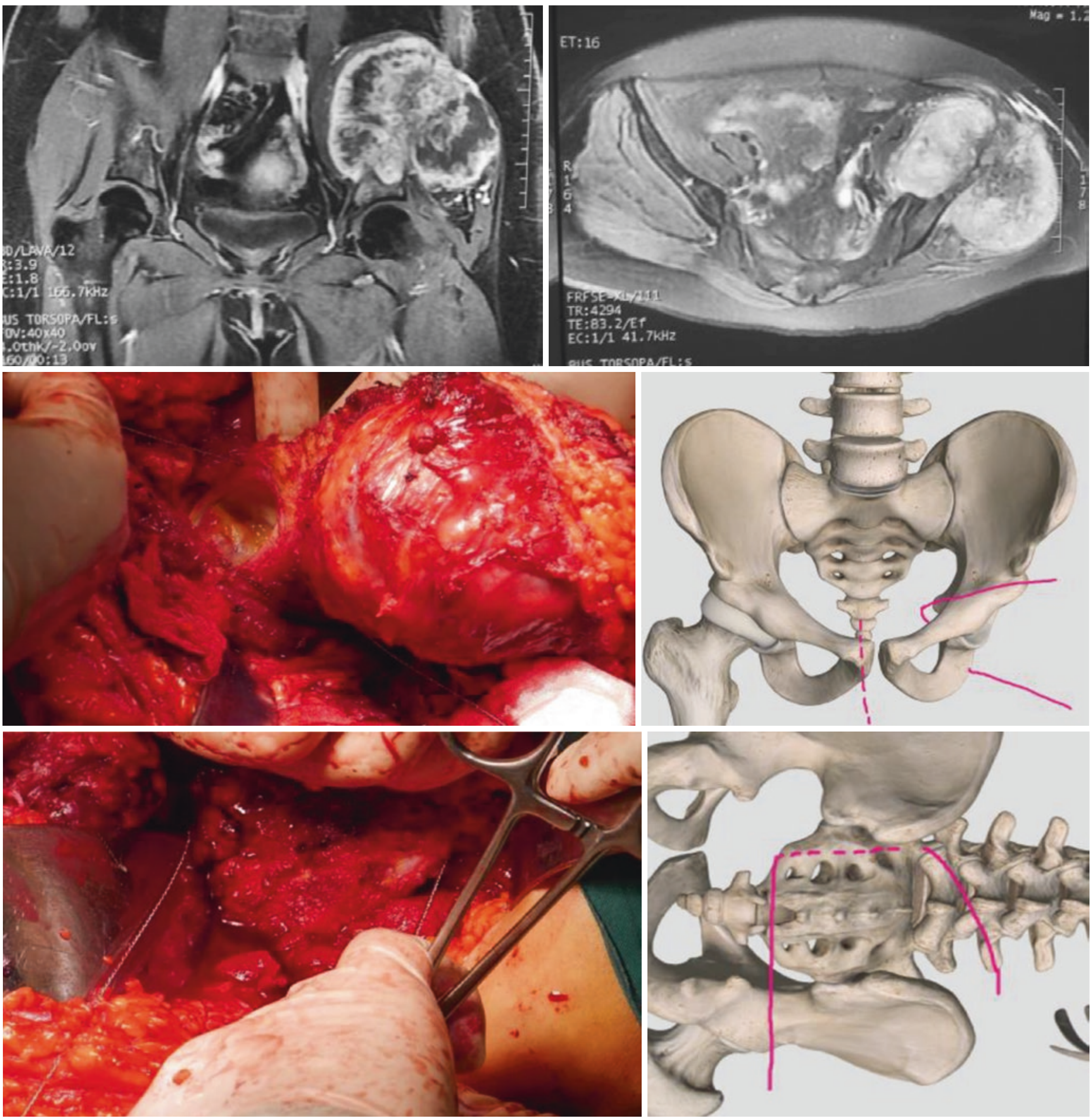

Fig. 9.3 Illustrative case. A 41-year-old female was diagnosed of osteosarcoma

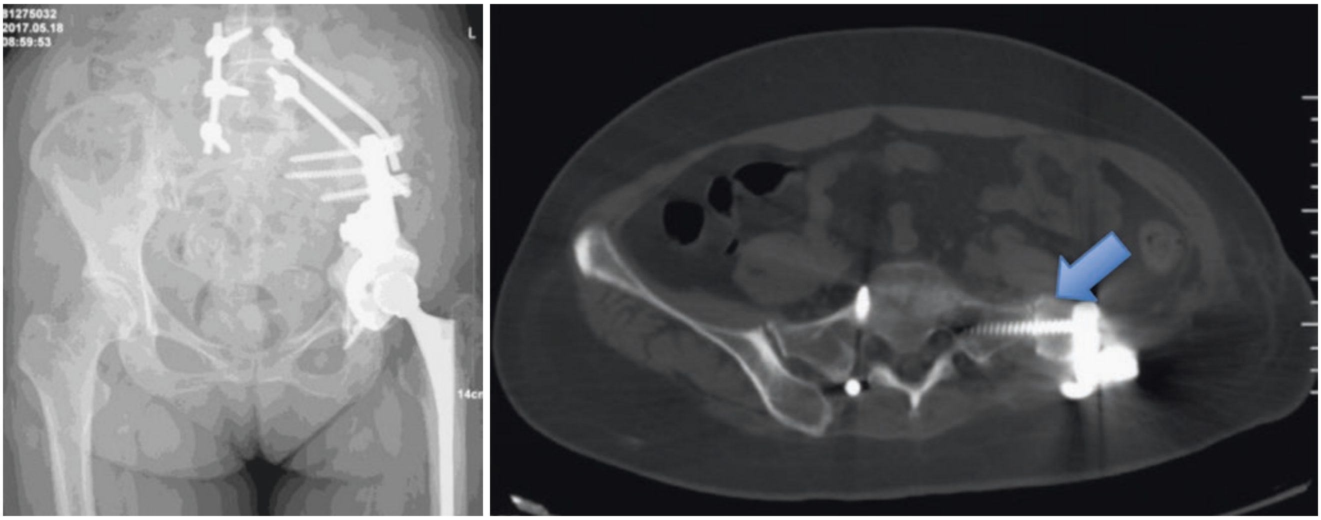

Fig. 9.3 (continued)

the midline in S1 particularly in the promontory. If the tumor extends into the body of S1, the vertical osteotomy should extend cranially to the tip of the sacral promontory. Then transverse division of the L5–S1 disc and the facet joint is performed. The superior end plate of S1 is divided vertically to join the vertical osteotomy at the promontory. This osteotomy destabilizes the lumbosacral articulation and may require instrumentation and fusion. Finally, the osteotomy surrounds the tumor caudally.

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10 Surgical Treatment for Metastatic Lesions in Pelvis

Daniel A. Driscoll, Francis J. Hornicek, Joseph H. Schwab, and Santiago A. Lozano Calderon

10.1 Introduction

Primary epithelial cancers can metastasize to distant organs of the body through hematogenous or lymphatic spread. Bone is the third most common metastatic site after the lung and liver. In fact, metastatic cancer is the most common malignancy of bone overall. Of the cancers that spread to bone, approximately 80% of bone metastases are caused by either prostate, breast, kidney, lung, or thyroid cancer [1]. The pelvis is the second most common osseous metastasis site after the spine and of all pelvic metastases; the most common sites are the ilium, ischium, and pubis, in descending order of frequency [2].

Pelvic metastases not uncommonly result in pain with patients presenting with limited function and even disability in many cases. Patients are frequently at risk for pathologic fractures, hypercalcemia, and anemia. As oncologic therapies such as immunotherapy, chemotherapy, hormonal treatment, and radiotherapy continue to advance, the life expectancy of patients with metastatic cancer continues to increase [3]. Along with this increased life expectancy, however, comes increased risk of bone metastasis, including metastasis to the pelvis.

In order to treat pelvic metastases, coordinated multidisciplinary care involving orthopedic surgeons, medical oncologists, radiation oncologists, and others is essential. Goals of care should include reduction of the patient's pain and restoration of function as rapidly as possible and for as long as possible. For some very specific and unusual cases, total resection of metastases in the pelvis may prolong a patient's life (Fig. 10.2 *Thyroid Metastasis Laura Walsh* case). However, in most instances, long-term control of metastatic disease is not usually realistic, and instead, the goal of treatment is palliation and attaining a weight-bearing as tolerated or stable status. Surgery is one tool among many in the treatment of pelvic metastases but can be especially effective in helping with pain control by stabilizing impending or frank pathologic fractures.

In this chapter, we describe the evaluation and management of pelvic metastatic disease. Our suggested protocol of treatment is based on location of the lesion, tumor histology, sensitivity of the tumor to radiation and/or chemotherapy, functional status of the patient and independency, and life expectancy. Most patients with pelvic metastases never even reach the surgeon for surgical intervention as most patients with metastatic disease are managed by their medical and radiation oncologists.

10.2 Location and Characteristic of Lesion

A main consideration in determining proper management of pelvic metastases is anatomic location of the metastasis within the pelvis. The Enneking classification divides the pelvis into four anatomic regions (Fig. 10.1; [4]). Zone 1 represents the ilium, zone 2 represents the periacetabulum, zone 3 represents the pubis, and zone 4 represents the sacrum. Zones 1 and 3 are non-weight-bearing, and metastases in these regions do not tend to compromise mechanical stability of the pelvic ring. Lesions in zone 2, however, do have risk for destruction of the hip joint and subsequent deterioration of function. In general, lesions involving more than one zone compromise stability of the pelvic ring. In addition, lesions that are osteolytic in nature are inherently at a higher risk for fracture than osteoblastic or mixed lesions.

10.2.1 Non-periacetabular Lesions

Many times, nonsurgical management may be initially adequate for treatment of metastases in Enneking zones 1 and 3 that do not extend into the acetabulum. Options include pain

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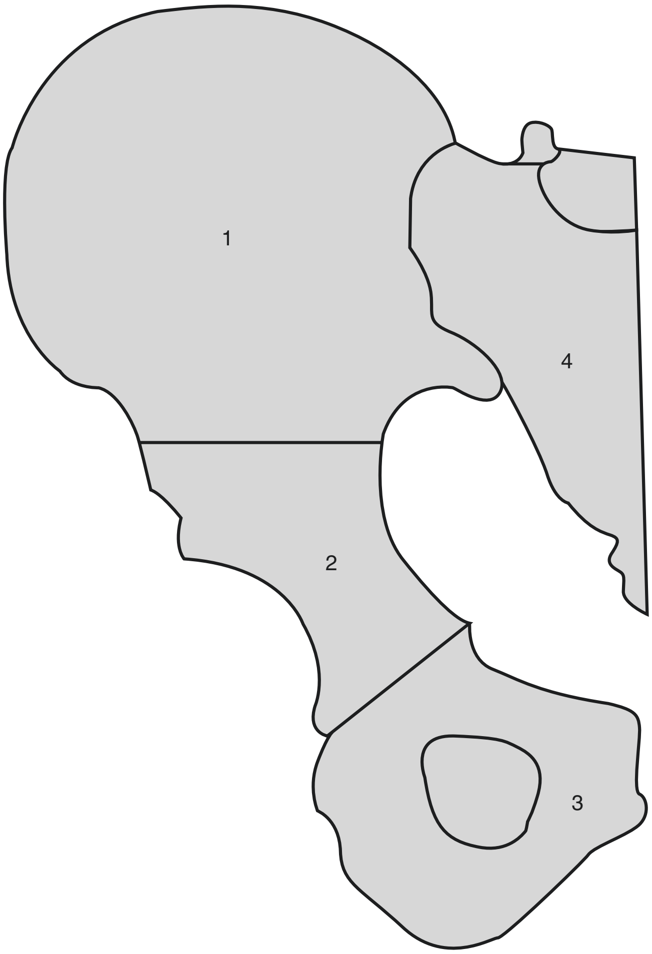

Fig. 10.1 Anatomic regions of the pelvis according to the Enneking classification. Type I: ilium, Type II: periacetabulum, Type III: pubis, Type IV: sacrum

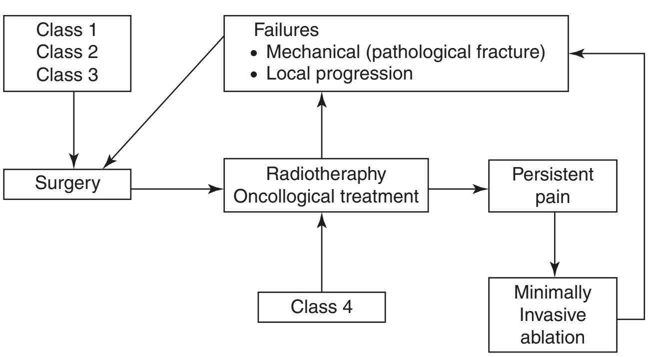

Fig. 10.2 Indications for surgical and conservative treatment according to the patient classes

control with analgesics, radiation therapy, hormone therapy, chemotherapy, bisphosphonates, and modification of weight-bearing [5]. Even in cases of severe bone destruction, patients with cancers that are responsive to medical or radiation therapy may not require surgery as long as the patient is compliant and able to maintain restrictions in terms of weight-bearing. Destruction of tumor through any of these methods may lead to, but not usually, eventual regeneration of bone. It is important to protect the area of bone healing with restricted activity or protected weight-bearing for comfort while potential healing may occur. A particular distinction needs to be made for a subgroup of patients that present with oligometastatic or isolated metastatic disease in the ileum or the pubis. As recommended by Müller and Capanna [6], lesions in zones 1 and 3 should be resected with wide margins in patients with solitary metastases. Lesions in these areas do not require further reconstruction as zones 1 and 3, as stated, are non-weight-bearing. Metastasectomy is further more important in certain histologic subtypes, such as renal cell carcinoma and thyroid adenocarcinoma. Metastasectomy of oligometastatic disease or isolated bone metastasis in these cases prolongs survival [7, 8]. Those isolated metastases occurring late in the disease have a better prognosis than those patients who present with an isolated metastasis.

10.2.2 Periacetabular Lesions

Periacetabular lesions (Enneking zone 2 lesions) can vary in the amount of destruction created in the pelvis. Many patients with zone 2 lesions also have extension to adjacent zones. Several classification systems exist in order to accurately describe periacetabular involvement of metastasis and guide management. The Harrington classification is arguably the most commonly used classification system. There are four groups in the Harrington classification with increasing severity. Group I represents intact subchondral bone of the acetabulum. Group II is defined as a destroyed acetabulum medial wall with an intact superior aspect (roof) and lateral wall. Group III is defined as destruction of the medial wall, superior aspect (roof), and lateral rim of the acetabulum. Group IV is defined as total collapse of the acetabulum.

An alternate to the Harrington classification is the metastatic acetabular classification (MAC) that enumerates four types of acetabular destruction: (1) acetabular dome, (2) medial wall, (3) anterior column, and (4) posterior column. Additional classification systems include the American Academy of Orthopaedic Surgeons (AAOS) Classification of Acetabular Bone Loss and the Column Classification Scheme, each of which is comprised of four grades. Regardless of which classification system is used, an understanding of how much acetabular damage a patient has is important for surgical planning.

Müller and Capanna [6] put forth a scoring system for pelvic metastases: the "Capanna classification." This scoring system is similar to a previous algorithm for long bone metastases reported by Capanna and Campanacci in 2001 [9]. Using this system, four groups of patients are created:

Table 10.1 Characteristics of the different patient classes comparing metastatic lesions in long bones and pelvis

Class	Long bones	Pelvis
1	Solitary metastatic lesion	
	Primary with good prognosis	
	(well-differentiated thyroid, prostate, breast sensitive to adjuvants, rectum, clear-cell renal, lymphoma, and myeloma)	
	Interval over 3 years since detection of the primary	
2	Pathological fracture at any site	Pathological fracture in periacetabular region
3	Impending fracture in a major weight bearing bone	Supra-acetabular osteolytic lesion
4	Multiple osteoblastic lesions at all sites	Multiple osteoblastic lesions at all sites
	Osteolytic or mixed lesions in nonstructural bone	Osteolytic or mixed lesions in iliac wing and anterior pelvis
	Osteolytic lesion with no impending fracture in major weight bearing bone	Small periacetabular osteolytic lesions

(1) solitary lesion with good prognosis, (2) pathologic fracture, (3) impending fracture due to supra-acetabular osteolytic lesion, and (4) other lesions. The authors suggest that generally, classes 1, 2, and 3 should have a surgical intervention while class 4 should engage in radiotherapy and oncological treatment and surgery should be used if other therapies fail (Table 10.1).

In general, each case needs to be evaluated on a case by case or individual basis. Having various options of surgical reconstruction of defects is important to provide immediate weight-bearing and improve pain management. The quality of life is important for these patients.

10.3 Surgical Indications and Contraindications

Surgical treatment for pelvic metastasis is indicated if (1) acute symptoms have not abated after a combination of protected weight-bearing, analgesics, and chemotherapy and (2) function has not returned within 1–3 months after radiation therapy. Additional indications that have been proposed include (1) a displaced fracture in a non-weight-bearing portion of the pelvis causing severe pain, (2) compromised structural support of the weight-bearing pelvis (periacetabular location) that is unlikely to respond to nonsurgical management, (3) metastatic disease causing impending fracture symptoms in the proximal femur, with concurrent structurally compromising metastatic acetabular disease, and (4) metastatic lesions with historically poor response to radiation or chemotherapy measures. Finally, it is requisite that a patient's expected prognosis be longer than the expected time for recovery after surgery so as to allow for potential net increase in quality of life (Fig. 10.2).

The five contraindications to surgical treatment for pelvic metastases are: (1) asymptomatic lesions; (2) expected prognosis less than 6 weeks; (3) contained acetabular defects in the non-weight-bearing pelvis amenable to nonsurgical treatment; (4) diffuse bony involvement of the hemipelvis not allowing for reliable, durable reconstruction; and (5) diffuse visceral metastases causing physiologic inability to handle the stress of a large operation.

10.4 Periacetabular Reconstruction Techniques

Multiple techniques for reconstruction of the periacetabulum have been described. The aim of these techniques is to reconstitute bone stability of the anterior and/or posterior columns of the acetabulum and reconstruction of the acetabular done. These include the traditional Harrington procedure; modified versions of the Harrington procedure; total hip arthroplasty with or without augments, or jumbo cups; saddle prosthesis; protrusio cage; and acetabuloplasty.

Harrington first described his eponymous technique for acetabular reconstruction in 1981 [10]. The procedure consists of intralesional curettage, filling of the acetabular defect with PMMA, and reinforcement of the structure with large threaded pins within the surrounding hemipelvis that allows for transfer of weight-bearing stress from deficient acetabular bone to healthy areas of the pelvis. Since that time, others have proposed multiple further modifications to the procedure.

For most cases, the amount and location of periacetabular bone destruction is an important determinant for type of surgical intervention. In cases in which subchondral bone of the acetabulum is intact, simple curettage and placement of PMMA are oftentimes sufficient (cementoplasty). Although use of bone graft is technically an option in these situations, techniques that require a prolonged healing time for osseous healing are not suited for patients with metastatic disease who require immediate return of function and relief of pain. Harrington class I defects can be treated with total hip arthroplasty and cementation of the acetabulum. The use of long-stem femoral components is helpful to mechanically protect the bone from potential metastasis in the proximal femur. Harrington II defects require use of THA with fixation of the medial wall of the acetabulum with a reinforcement ring. For Harrington III, reconstruction can be achieved via the use of a Harrington technique [6]. Others suggest that a column plate should be used instead of the Steinman pins of the Harrington procedure. In patients with Harrington IV defects (minimal bony integrity of the acetabulum), more resection of the acetabulum is required, and thus aggressive reconstruction is subsequently needed. This reconstruction can be achieved with use of either a megaprosthesis, a saddle prosthesis, or a massive allograft with THA. In addition, Lozano-Calderon et al. [11] describe a technique for Harrington type III lesions that involves curettage of the lesion with retrograde placement of Steinmann pins through the ischial tuberosity and posterior column into the sciatic buttress. The authors performed the technique on 11 patients and found that the 6 patients surviving at 12-month interval after surgery had improved mobility and good quality-of-life scores.

10.5 Nonsurgical Treatments

In addition to reconstructive techniques, nonsurgical techniques for treatment of pelvic metastasis have been described. These include ethanol treatment, cryoablation, radiofrequency ablation, embolization, acetabuloplasty, laser-induced thermotherapy, and electrotherapy. These treatments are especially useful in patients who are indicated for surgical management but are not candidates for surgery due to health status. These nonsurgical treatments tend to have relatively low rates of complication. However, in patients with lesions within 1 cm of important structures, such as major vessels or nerves, these treatments are contraindicated due to inability to visualize important structures easily and risk for damage. Moreover, many of these ablation-based procedures can be combined with acetabuloplasty with PMMA (described below).

10.5.1 Ethanol

Ethanol injection under the guidance of CT has been used as simple and economic treatment for metastasis. Gangi et al. [12] described a cohort of 25 patients with 16% of patients experiencing complete pain relief (defined as no longer needing analgesic drugs) and 74% of patients experiencing a reduction in need for analgesics by at least 25–50% 2 weeks after the procedure. Twenty-six percent of patients experienced a reduction in tumor size, while 18.5% had an increase in tumor size.

10.5.2 Radiofrequency Ablation

Radiofrequency ablation (RFA) is a treatment performed under CT or fluoroscopy guidance by interventional radiologists. It allows for necrosis of tumor cells in situ by introducing a high-frequency alternating current into the tissue. This treatment is most appropriate for patients with one to two lesions of either osteolytic or mixed osteolytic/osteoblastic disease. It can also be used in combination with acetabuloplasty. Dupuy [13] used RFA to significantly reduce pain in patients with persistent pain after radiation therapy. Goetz et al. [14] used RFA in 43 patients and noted pain relief at 24 weeks after treatment in 95% of patients.

10.5.3 Cryotherapy

Cryotherapy is performed with the goal of shrinking tumor by causing cell death via intracellular ice formation and creation of osmotic differences. Ideal candidates for this treatment are patients with one to two bony metastases causing pain. Cryotherapy is much less effective in causing necrosis of soft tissue, so this therapy is less appropriate for patients with tumors with significant soft tissue components. A prospective multicenter study was conducted which reported significant pain relief in 61 patients. Then, a retrospective comparison was performed on 58 patients treated with either RFA or cryoablation and found that those treated with cryoablation had significantly shorter hospital stays and significantly larger reductions in narcotic requirement. Additionally, Callstrom et al. [15] treated 14 patients with cryoablation and noted pain relief in all patients.

10.5.4 Percutaneous Cementoplasty/Acetabuloplasty

This treatment, adapted from vertebroplasty techniques, involves the percutaneous injection of PMMA in order to increase structural integrity of the acetabulum. It is best suited for treatment of osteolytic lesions, which leave a cavity to be filled. Moreover, an exothermic reaction occurs as the cement hardens, theoretically providing a local cytotoxic effect. Maccauro et al. [16] used percutaneous cementoplasty to treat 30 patients with acetabular lesions and found that 59% experienced complete pain relief while the remaining 41% experienced significant reduction in pain. Kelekis et al. [17] treated 14 patients and noted pain relief in 92% of patients. Several other studies report pain relief rates ranging from 81% to 100% [18–20]. Colman et al. [21], however, found that patients with Harrington II or III lesions treated with percutaneous cement acetabuloplasty had less pain reduction and less improvement in ambulatory status at 3 months than patients treated with acetabular reconstruction via surgery.

10.6 Tumor Histology

While multiple different primary neoplasms can metastasize to the pelvis, neoplasm origin can be important in tailoring treatment practice. For instance, while preoperative embolization is useful for many different tumor types, it is especially useful when treating patients with renal or thyroid metastases, which are known to be hypervascular in nature. Further, when treating myeloma, prostate cancer, and breast cancer, multiple bisphosphonates can be used and have been shown to be effective in reducing both pain and risk of future fracture.

In addition to its use in determining treatment pathway, tumor histology can also be helpful in determining prognosis for patients with pelvic metastases. Perhaps unsurprisingly, multiple studies have seen a difference in survival based on primary neoplasm type in patients with pelvic metastases. Patients with melanoma, genitourinary cancer, thyroid, or unknown primary tumor had a significantly worse prognosis (mean survival from surgery of 7 months) than other patients, whereas patients with hematopoietic malignancies had a significantly longer mean survival time from surgery (72 months). In the Tsagozis study, patients with prostate, breast, lung, and oropharyngeal cancer have a mean survival of 19 months. Additionally, Marco et al. [22] also found breast cancer to be a significant predictor of longer survival in comparison with other neoplasms. Similarly, a study showed that breast cancer as primary malignancy significantly predicted longer survival (mean survival 21 months) compared to other primary neoplasms (mean survival 9 months). The authors also noted that absence of visceral metastases was a significant predictor of longer survival.

Understanding whether a lesion is osteoblastic, osteosclerotic, or mixed is helpful in determining treatment. As mentioned above, some nonsurgical constructive techniques are only feasible with osteolytic lesions. Moreover, it has been suggested that solitary osteoblastic lesions outside of Enneking 2 zone require surgery much less frequently than osteolytic or mixed lesions in the same area Müller and Capanna [6]. Osteosclerotic acetabular lesions and lesions in the iliac wing and the anterior pelvis are commonly treated conservatively with external irradiation to reduce pain and as local control [6].

10.7 Sensitivity to Radiation and Chemotherapy

In the treatment of patients with pelvic metastases, first-line treatment includes radiation therapy. Report that 65–70% of patients with bone metastases experience some improvement in pain with radiation therapy alone. Between 20% and 50% of patients report complete pain relief. Radiation is able to help a significant amount of people, and surgery is avoided for these patients. Thus, a majority of surgical candidates are poor responders to initial radiation therapy. Moreover, if a patient has recurrent pain at a site already exposed to radiation, further radiotherapy may not be an option, thus making surgical intervention more appropriate.

Some authors have proposed divisions for neoplasm type based on response to adjuvant therapy. Müller and Capanna [6] categorize neoplasms into two groups: (1) responsive to adjuvant therapy and (2) nonresponsive to adjuvant therapy. The first group contains breast, thyroid, myeloma, lymphoma, and prostate cancers. The second group contains cancers of the kidney, gastrointestinal tumor, lung, uterus, and pancreas.

10.8 Functional Status of the Patient and Independence

As noted previously, the goals of surgical intervention in treatment of pelvic metastasis is the improvement in pain and increase in function. It is fairly widely accepted that surgical intervention provides pain relief and improves functional status as multiple studies have demonstrated an improvement in function after surgical reconstruction of the acetabulum. In one of the studies, 70 patients with acetabular metastases with protrusio cage, retrograde screws, and cemented THA were treated, and all patients showed reduced pain after treatment besides increased function except 3 patients. In another study, 16 patients with intralesional excisions of tumor and cemented THA were treated, and a mean change in VAS from 8/10 preoperatively to 2/10 postoperatively in their patient cohort was found, indicating a significant improvement in quality of life. A study adopted a modified Harrington procedure on nine patients and found that eight of nine patients achieved an improvement in the functional status after surgery, even in patients with preoperative ASA scores ranging from II to IV. When 62 patients with metastatic disease to the acetabulum underwent THA with acetabular reconstruction, these patients showed a mean ECOG score improvement of 2.6 preoperatively to 1.1 postoperatively. Kiatisevi et al. (2015) treated 22 patients with acetabular reconstruction and found an improvement in mean ECOG score from 3.1 to 1.7, an improvement in VAS from 8.4 to 2.2, and a mean postoperative MSTS functional score of 70. Twenty of the 22 patients were able to walk, with 8 of these 20 able to walk in the community and the rest able to ambulate at home.

While surgical intervention has proved to increase function in patients, consideration of a person's baseline function is important when deciding if surgery is the correct intervention. For example, it is not appropriate to perform acetabular reconstruction in patient who was of limited functional status prior to a diagnosis of cancer.

10.9 Life Expectancy

Patients with pelvic metastases are—in the vast majority of cases—considered to have terminal disease, and treatment is aimed at palliation of symptoms. In terms of clinical communication, it is important to make this clear to patients in the preoperative period in order to provide for realistic expectations. Survival reports from studies describing treatment of pelvic metastases support the notion that these patients' diagnoses are terminal. Thus, there is a tension between desire to improve patient quality of life quickly and not risking further morbidity or mortality as a result of intervention.

Clayer (2010) performed protusio cage insertion in 29 patients with metastatic disease to the acetabulum and found median length of survival to be 100 months. However, the cohort study showed a median survival of 12 months (70 patients). In other study, 81 patients after treatment had five-year survivorship of 5% with mean survival of 23 months and median survival of 15 months. Marco et al. [22] found a median survival of nine months in their cohort of 54 patients.

Interestingly, the initial work of Harrington [10] reported 52% survival at two years after surgery. This is likely due to a difference in patient populations, with Harrington treating patients at earlier stages of disease progression. Rather than treat patients based on level of acetabular involvement, Harrington operated on patients based on presence of pelvic metastasis and overall prognosis.

In their proposed scoring system, Müller and Capanna [6] divide predicted survival into three groups: (1) less than 1 year, (2) 1–2 years, and (3) greater than 2 years, based on source of metastasis. Less than 1-year survival was predicted for melanoma, lung, pancreas, undifferentiated thyroid, and stomach cancers. One- to two-year survival was predicted for colon, breast (unresponsive to adjuvants), liver, and uterus cancers. Over 2 years for patient survival was predicted for differentiated thyroid, myeloma, lymphoma, breast cancer (responsive to adjuvants), rectum, prostate, and kidney cancer.

Of note, Ruggieri et al. [23] found no difference in survival for patients with pelvic metastases treated with wide local resection or curettage/marginal resection. Moreover, this difference held for even patients with solitary lesions.

10.10 Conclusion

Care for patients with epithelial metastases to the pelvic requires a multidisciplinary approach that includes coordination of oncology, radiation oncology, and orthopedic surgery teams. When indicated, surgical treatment can offer significant pain reduction and functional improvement in a relatively short amount of time. Seldom does surgery improve long-term survival except in very selected patients with isolated disease discovered years after the initial presentation. In addition, nonsurgical treatments continue to improve in their ability to increase patients' quality of life. Surgery is most helpful in patients with lesions affecting the periacetabular region, and determination of proper acetabular reconstruction can be done by evaluation of location and infiltration of the pelvic metastasis. Other factors such as tumor histology, radiation sensitivity, functional status, and life expectancy should also be taken into account in order to determine a proper course of action.

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External Hemipelvectomy

11

Dasen Li and Wei Guo

In an age when internal hemipelvectomy is used widely, the limb-salvaging procedure is the first choice in most patients with malignant pelvic tumors. Although external hemipelvectomy (hindquarter amputation) is characterized by poor postoperative function and a huge cosmetic defect, there are still indications for this approach [1–3]. *Although posterior flap external hemipelvectomy is used much more frequently than is anterior flap external hemipelvectomy, the latter is a reasonable option when a posterior flap is unavailable.*

11.1 Indications

- Two out of three structures, i.e., the acetabulum (pelvic region II), the sciatic nerve, and the external iliac vessels, are affected by a malignant tumor or a locally aggressive tumor [4].
- Extensive local recurrence of a tumor after internal pelvectomy.
- Presence of a benign tumor with an extensive mass, as illustrated in Fig. 11.1 [5].
- Presence of overwhelming infections, such as gas gangrene or the consequences of trauma.

11.2 Classifications

"Classic or standard external hemipelvectomy" consists in the cutting off and ligation of the common iliac vessel, cutting off the pelvic ring through the sacroiliac joint and the pubic symphysis, and closing the wound using a gluteus maximus fasciocutaneous flap (Fig. 11.2a).

If the proximal bony cut line is through the ilium, instead of the sacroiliac joint, the procedure is called modified external hemipelvectomy (Fig. 11.2b). If the cut line is through the sacral ala, because the sacroiliac joint is affected by the tumor, the procedure is called extended external hemipelvectomy (Fig. 11.2c).

The blood supply of the gluteus maximus should be preserved by conserving the internal iliac vessels, so that the wound can be closed using a gluteus maximus musculocutaneous flap, which is also called modified external hemipelvectomy.

The performance of combined external hemipelvectomy implies that the pelvic viscera, such as the bladder, bowel, vagina, and/or the uterus, are affected by the tumor and have to be resected together with the limb (Fig. 11.3).

Based on the skin flap designed to close the wound, external hemipelvectomy is divided into posterior flap external hemipelvectomy and anterior flap external hemipelvectomy. The choice of the skin flap depends on the location of the tumor. Posterior flap external hemipelvectomy is used most commonly, with a gluteus maximus fasciocutaneous flap or a gluteus maximus myocutaneous flap. When the deep fascia cannot be preserved with the flap, flap necrosis tends to occur (Fig. 11.4). Occasionally, the posterior flap is invaded by the tumor (Fig. 11.5), and the anterior flap, or quadriceps femoris flap, is a perfect option if the iliac vessels and superficial femoral artery are intact. The quadriceps femoris flap has adequate blood supply and is large enough to allow the repair of a huge soft tissue defect. Myocutaneous flaps based on the obturator vessel

Fig. 11.1 External hemipelvectomy was to be performed in a patient with neurofibromatosis because of the huge and extensive mass in the right lower limb

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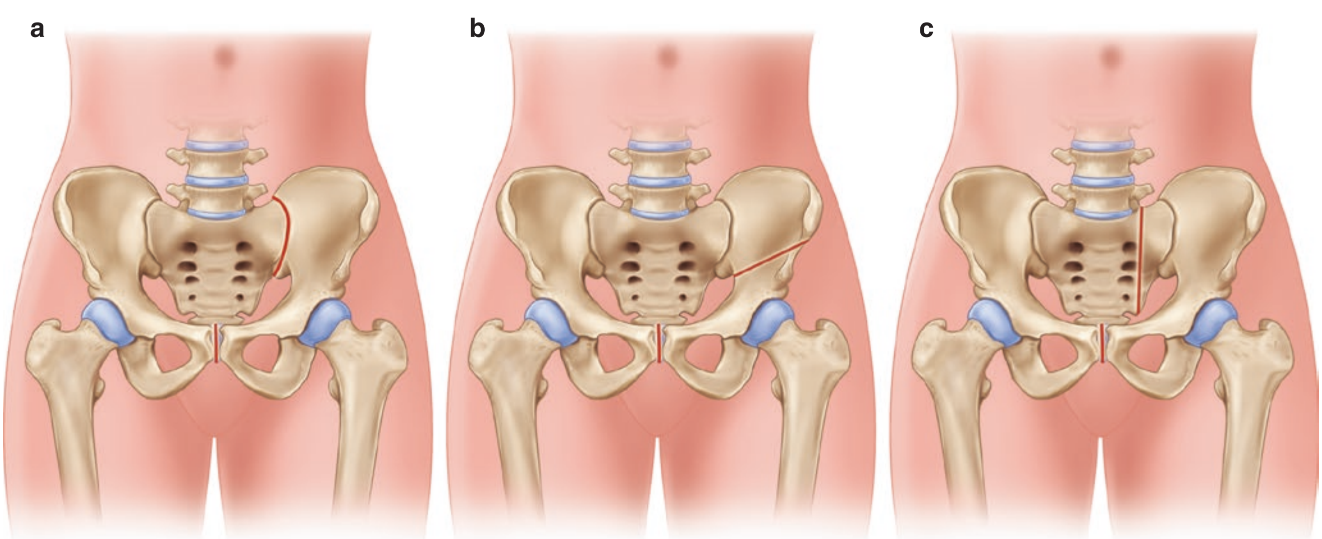

Fig. 11.2 Classic (**a**), modified (**b**), and extended external hemipelvectomy (**c**), based on the bony cut lines

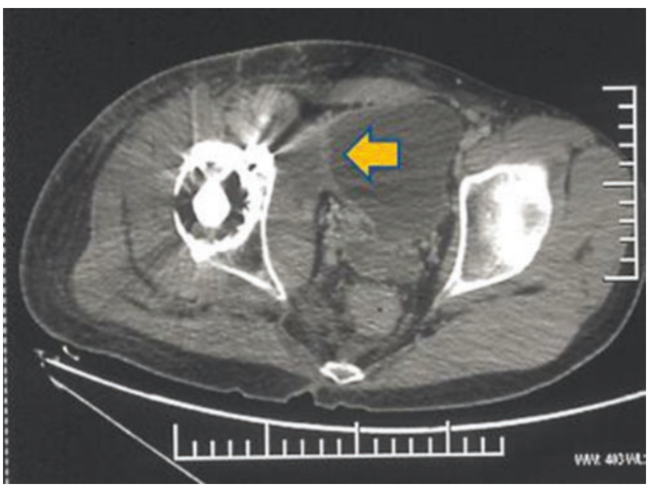

Fig. 11.3 Pelvic viscera (bladder) invaded by sarcoma

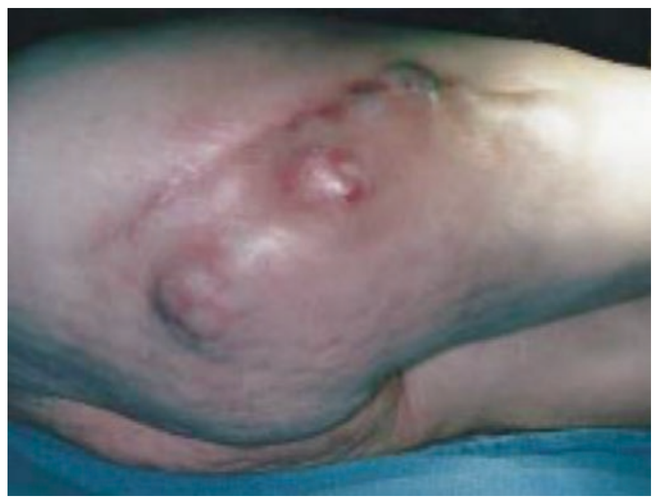

Fig. 11.5 Posterior flap invaded by tumor

Fig. 11.4 Necrosis of posterior flap

are applied rarely because they exhibit a high rate of flap ischemic necrosis.

Therefore, a meticulous preoperative workup using MRI and CT is necessary to plan a tumor resection with safe bone and soft tissue margins and, thus, to determine the type of external hemipelvectomy that is most appropriate. Occasionally, when an anterior flap is considered, arteriography is used to evaluate the superficial femoral artery.

In 2013, Grimer et al. reported 157 cases of external hemipelvectomy with a perioperative mortality rate of 1.3% over the past 30 years [2]. Hemorrhagic shock and septicemia caused by deep infection were the main causes of death. Based on our experience, blood control strategies, such as temporary abdominal aortic balloon occlusion and preoperative artery embolization, could be considered for tumors with a huge soft tissue mass and abundant blood supply. The incidence of wound infection or flap necrosis was reported to be as high as 45%. The use of carefully designed flaps can reduce the wound complication rate dramatically.

11.3 Preparations

- Prepare more than 2000 mL of blood according to the condition.
- General anesthesia.
- Insert a ureteral catheter for the identification of the ureter, when necessary, and drain the bladder using a Foley catheter.
- Close the anus temporarily with a purse-string suture or insert a rectal tube in the rectum for identification of the rectum, when necessary.
- Position the patient in the lateral decubitus position using sandbags, so that the patient can be shifted from a more supine to a more prone position easily. The lateral decubitus position can make the organs in the abdominal cavity move toward the healthy side.

11.4 Surgical Procedure for Posterior Flap External Hemipelvectomy

This is a three-step procedure.

- Step 1. Position the patient in a somewhat supine position. From the mid ilium, the anterior incision follows the iliac crest to the anterior iliac spine and then extends along the inguinal ligament to the pubic tubercle (Fig. 11.6a, b). Detach the abdominal muscles and the inguinal ligament from the iliac crest. Expose the retroperitoneal space. In men, retract the spermatic cord medially. Detach the rectus abdominis from the pubic bone, retract the bladder medially, and pad the Retzius space using gauze. Identify the common iliac vessels and the ureter, and doubly ligate and transect the external or common iliac artery and veins. Transect the femoral nerve and the iliopsoas at the same level.
- Step 2. The assistant should abduct the hip, and the surgeon should stand between the lower limb that is to be amputated and the table. Extend the incision from the pubic tubercle to the ischial tuberosity (Fig. 11.6a). Expose the pubic and ischial rami, and detach the perineal muscles and the corpus cavernosum subperiosteally. Protect the urethra and divide the symphysis pubis.
- Step 3. Position the patient in a more prone position. Flex and adduct the hip. The posterior incision starts from the mid iliac crest and passes over the greater trochanter, along the gluteal crease, to join the perineal incision at the ischial tuberosity (Fig. 11.6a, c). Incise the aponeurosis and the muscle fibers (when possible) of the gluteus maximus in line, along the skin incision. Transect the piriformis muscle. Sharply transect the sciatic nerve and allow its retraction. Ligate the small artery in the sciatic nerve. Detach the latissimus dorsi and quadratus lumborum from the iliac crest. Expose the greater sciatic notch and pass a Gigli saw through it (Fig. 11.6d, e). Disarticulate the sacroiliac joint and transect the ilium, or even resect a portion of the sacrum, as planned, to establish a surgical margin. Rotate the lower limb and pelvis externally, and ligate and divide the obturator vessels and nerve. Free the hindquarter by transecting the sacrotuberous, the sacrospinous ligaments, and the levator ani close to its origin on the ischium and the pubis.
- Place one or two suction drains and close the wound: begin closure by suturing the posterior flap to the lateral border of the abdominal muscles, and then close the skin loosely (Fig. 11.6f).

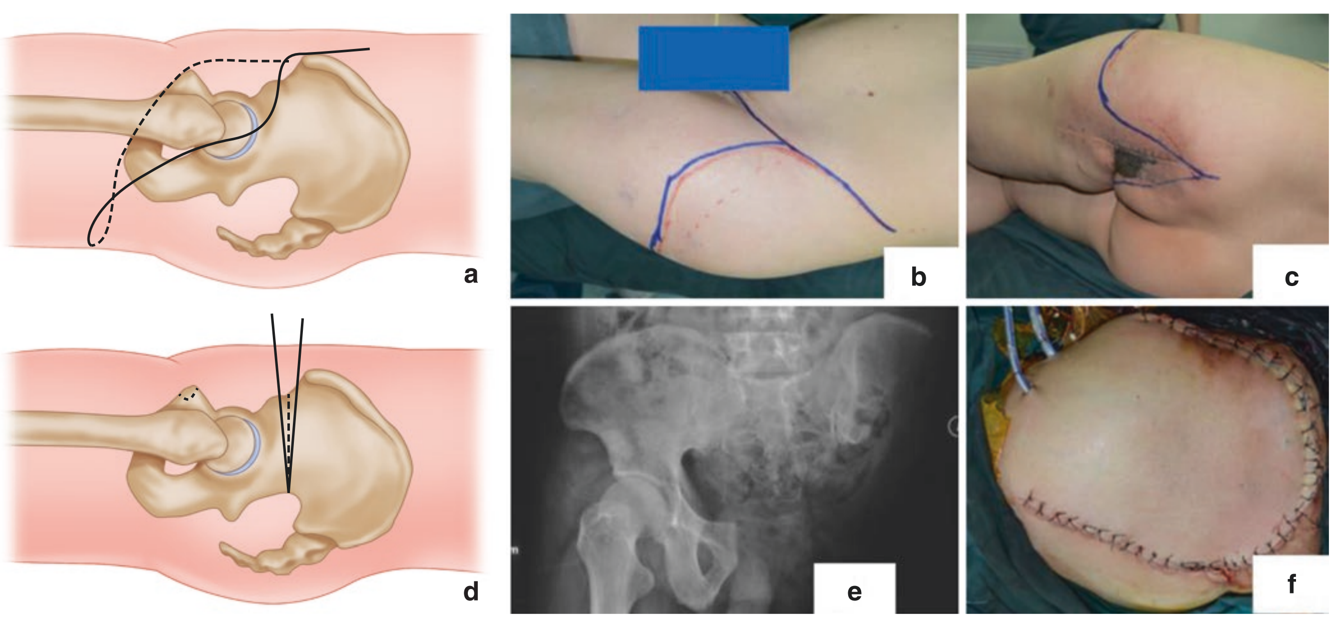

Fig. 11.6 Modified external hemipelvectomy (posterior flap)

Tips

- Preservation of the internal iliac artery should lead to a much better blood supply.
- Resection of the gluteal maximus fascia with the tumor, leaving only the posterior skin flap to close the wound, would lead to a much higher rate of complications of the postoperative wound.
- Protect the venous plexus on the surface of the bladder and prostate (in men), and leave the posterior urethra intact when dividing the pubic symphysis. Hemostasis by compression or suturing is effective in this area.
- When the sacral nerves are to be transected, cut and ligate them at 1–2 cm from the sacral foramina. Thus, the erectile nerve branch of the pelvic nerve plexus, which is derived from the sacral nerve, may be preserved.
- Postoperative management.
- Administer a low-fiber postoperative diet for 3 days. A laxative or enema can be used to help defecate on the fourth day postoperatively.
- A custom-made truss may be helpful to relieve the discomfort of the wound when the patient stands up.

11.5 Surgical Procedure for Anterior Flap External Hemipelvectomy

Design the quadriceps femoris flap according to the soft tissue defect (Fig. 11.7a–d). Usually, the incision begins at the anterior superior iliac spine and extends along the lateral aspect of the thigh to the level of the quadriceps tendon. Here, it extends transversely to the medial aspect of the thigh and then to the groin (pubic tubercle). Develop the musculocutaneous flap by elevating the quadriceps from the femur. Find the femoral vessels, ligate the profunda femoris vessels, and keep the superficial femoral vessels intact with the musculocutaneous flap (Fig. 11.7e).

Finish the external hemipelvectomy as described previously. Resect the posterior flap together with the lower limb (tumor), and close the wound with the anterior flap (Fig. 11.7f).

Tips

- Some elderly patients or patients with diabetes mellitus may have femoral vascular disease, such as atherosclerosis. The superficial femoral artery may be evaluated via preoperative arterial angiography.
- In some cases, this operation requires resection of the ipsilateral sacral ala.
- In some cases, the skin of the groin cannot be preserved, and an "island" flap pedicled with the superficial femoral artery is needed.
- When necessary, the posterior skin incision may exceed the posterior midline.

11.6 Replantation of Autologous Normal Tissue of the Ipsilateral Leg After External Hemipelvectomy

For patients with a large soft tissue defect, flaps such as a rectus abdominis musculocutaneous flap or a latissimus dorsi musculocutaneous flap may be used to close the

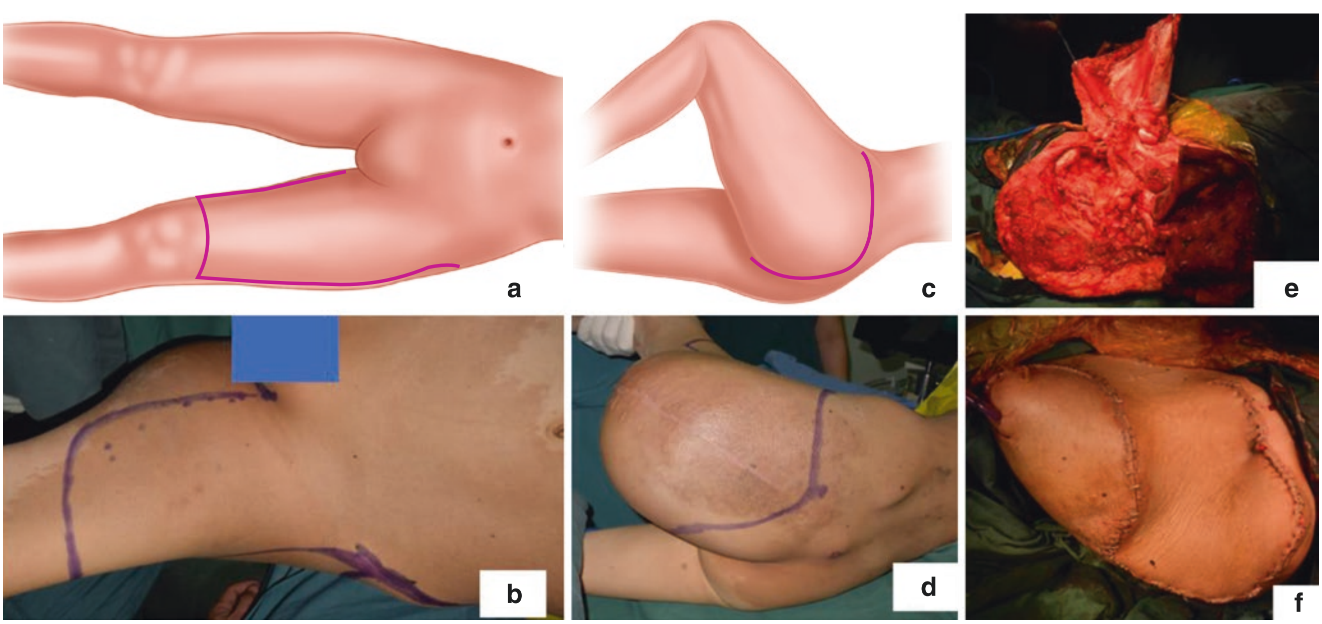

Fig. 11.7 Modified external hemipelvectomy (anterior flap)

wound after external hemipelvectomy [6, 7]. However, these procedures will bring additional trauma to the patient. Using microsurgical techniques, replantation of the free vascular pedicled calf musculocutaneous flap of the amputated lower limb could be used to close the wound [8].

Bramer reconstructed the ipsilateral ischial tuberosity using autologous tibial bone harvested from the amputated limb, which improved the patient's stability in the sitting position and artificial limb function [9]. Mei et al. suggested that the reconstruction of hemipelvic defects with femur may be a better choice, from a biomechanical point of view [10].

Acknowledgment *Source of all the clinical images and the diagrams*: All the clinical images were from the database of our center.

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Part III

Pelvic Tumors: Pelvic Reconstruction

12 Reconstruction After Ilium Resection

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The ilium serves two primary functions: it provides continuity between the acetabulum and the sacrum by means of the sacroiliac joint; it also provides a major soft tissue attachment site for the abdominal, gluteal, pelvic floor, rectus femoris, sartorius, and the iliacus muscles within the pelvis. Variations within this subset of resections include complete removal of the ilium and removal of partial ilium. Certainly, complete removal of the ilium "disconnects" the acetabulum from the ipsilateral sacroiliac joint and may cause significant destabilization of the remaining segment because it now hinges on the pubic symphysis. Partial resection of the iliac wing, by contrast, is a much less morbid procedure if continuity is maintained between the acetabulum and the axial skeleton.

Restoration of pelvic ring integrity has typically been achieved using bone allograft, autologous nonvascularized or vascularized fibular grafts, or iliac crest pedicle graft [1] because reconstruction of the pelvic ring and lumbar-pelvic junction is necessary to reduce rotational and translational instability essential for weight-bearing and ambulation. Failure to reconstruct the lumbar-pelvic junction and restore abdominal wall integrity may also result in symptomatic visceral herniation.

Regardless of the techniques chosen to reconstruct the posterior pelvis after creation of large bony defects that disrupt the pelvic ring and lead to instability, there are inherent difficulties in achieving sacroiliac arthrodesis. Occasionally, the distance between the remaining ilium and the sacrum is small enough that it can be closed primarily and wired in order to achieve direct bony healing. The remaining sacroiliac joint is denuded of cartilage, and a wire or suture construct may be used to "close down" the pelvis by hinging the remaining hemipelvis through the flexible symphysis pubis joint anteriorly [2]. Fixation of structural bone grafts to the residual supra-acetabular pelvis and sacrum also is problematic. Most of the patients required a posterior osteotomy through sacral ala to achieve safe margins of resection such that the amount of residual of sacrum available for fixation is limited.

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When the gap is too large to be closed using the direct apposition, a strut graft may be introduced to span the gap. Autograft fibular struts should be considered (with or without vascularity) to facilitate the bony healing. In younger patients with a favorable soft tissue envelope, allograft fibula [3] is a good option that has been shown to incorporate reliably and avoid donor site morbidity. Autoclaved autograft and vascularized iliac wing autograft have also been described. Biological reconstruction has the potential for permanent consolidation with bone and the avoidance of revision arthroplasties; however, prosthetic reconstruction has the advantages of early mobilization, acceptable cosmesis, long-term stability, and satisfactory function. With the advent of 3D printing technology (additive manufacturing or rapid prototyping), Guo et al. [4] reported a 3D-printed iliac prosthesis which can be used for ilium defect due to tumor resection. The prosthesis has features including preoperatively designed screw holes for acetabular side fixation, soft tissue reattachment anchorage, and structure connecting with screw-rod system. The bone-implant interface is porous on metallic side, which facilitates bone ingrowth, and osseointegration can be achieved between host bone and prosthesis.

12.1 Surgical Techniques

The patient is positioned in a semimobile lateral position. An extensile incision is made from the posterosuperior iliac spine extending anteriorly along the area of the iliac crest to the anterosuperior iliac spine and then directed distally along inguinal ligament, curving anteriorly, ending just proximal to the pubic tuberosity. The anterior abdominal wall is detached from the iliac crest, and the retroperitoneal space is exposed. The dissection then is developed between the iliacus and psoas. The iliacus muscle then is identified at the level of the intended iliac osteotomy just at the line of the anteroinferior iliac spine directly toward the sciatic notch.

When the inner and outer tables were thoroughly exposed, two Gigli saws then are passed through the notch with

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particular care of sacral nerve, and the osteotomy are performed just superior to acetabulum, normally at the level of the anteroinferior iliac spine. The other Gigli saw is then sued for osteotomy near iliosacral joint according to the preoperative plan. The remainder of the gluteus medius and minimus is transected along the line of intended resection from the sciatic notch up toward the anteroinferior iliac spine. Then the resection is completed, and there may be hemorrhage near superior gluteal vessels. Ligation and cautery are necessary.

Pedicle screws are placed into the lower lumbar spine, sacral pedicles, and vertebra body if necessary as anchors for the reconstruction using a spinal instrumentation system. Typically, we use a 6.0-mm titanium rod system rather than a 5.5-mm diameter rod system to perform the reconstruction as it may not be strong enough to support the reconstruction. Usually, a 7.5-mm diameter titanium multiaxial pedicle screw is placed in the sacrum through S1 pedicle. The screw lengths range from 35 to 45 mm depending on the size of the patient. We then identify another start point medially along the cut portion of the ischium and used a pedicle finder to probe in a lateral-to-medial direction distally in the remainder of both anterior and posterior columns. Fluoroscopy is used to ensure the trajectory not violating the notch or acetabulum. We then implant a 7.5-mm or 8.5-mm diameter titanium multiaxial pedicle or specially designed iliac screw after tapping the hole created by the pedicle finder. The screw lengths range from 50 to 75 mm depending on the size of the remaining ilium. Both screws are confirmed with fluoroscopy.

After confirmation of the screw position, titanium mesh, vascularized or nonvascularized fibular, or endoprosthesis was utilized to reconstruct the continuity of the pelvis keyed into the remaining ilium and sacrum. A 6.5-mm rod is measured to fit the distance spanned between the two screws, then contoured, connecting the two screws, and secured to the screws with the appropriate set screws. Compression is applied across the construct, ensuring that the graft is stable. Then the gluteus fascia is sutured to the abdominal wall. The skin was closed in layer fashion (Figs. 12.1, 12.2 and 12.3).

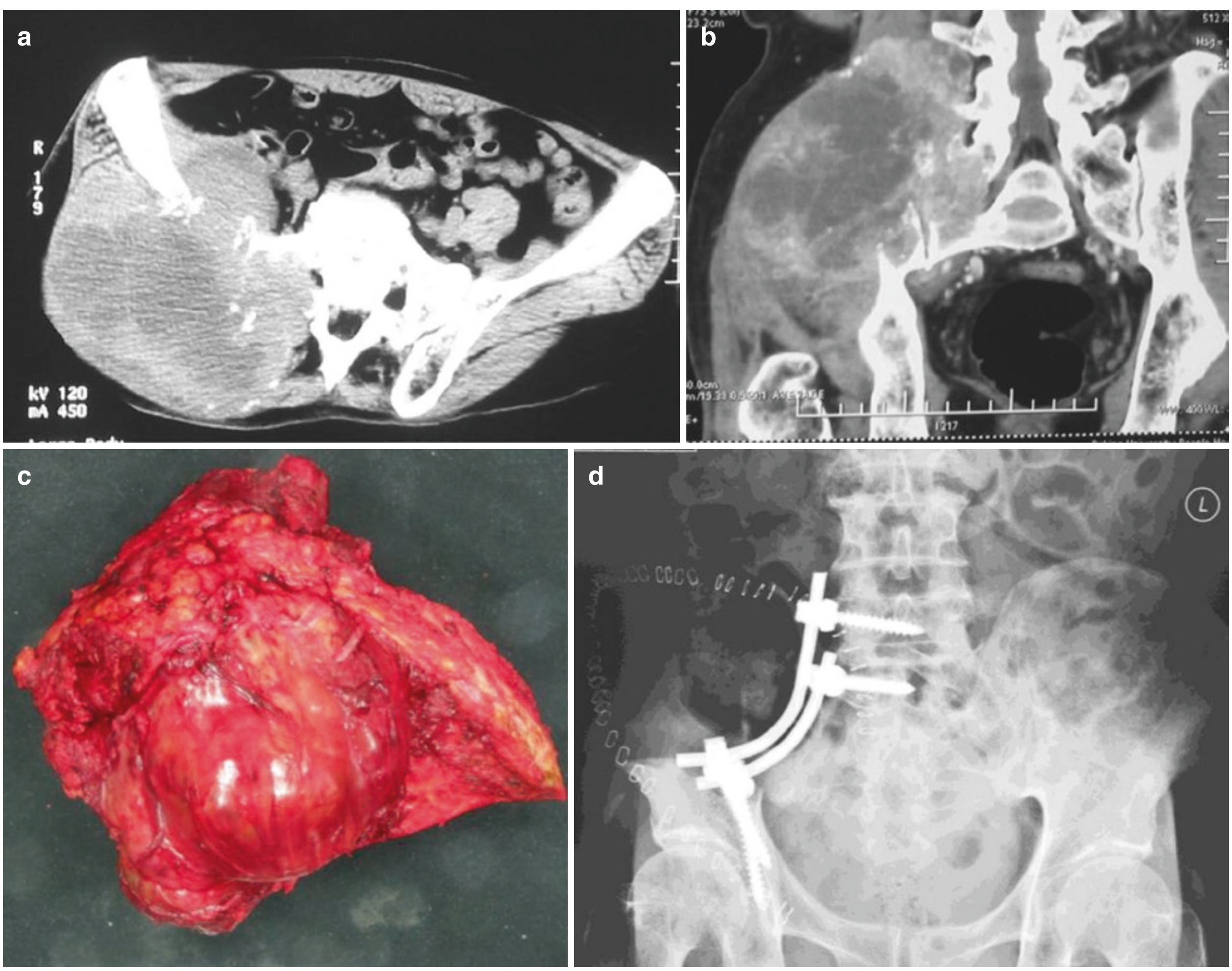

Fig. 12.1 The patient was a 72-year-old female with diagnosis of giant cell tumor of right ilium. Preoperative axial and coronal CT showed the lytic destruction of right pelvis with sacroiliac joint involved (**a**, **b**). (**c**) Tumor specimen after resection. (**d**) Postoperative X-ray showed the reconstruction with screw-rod system

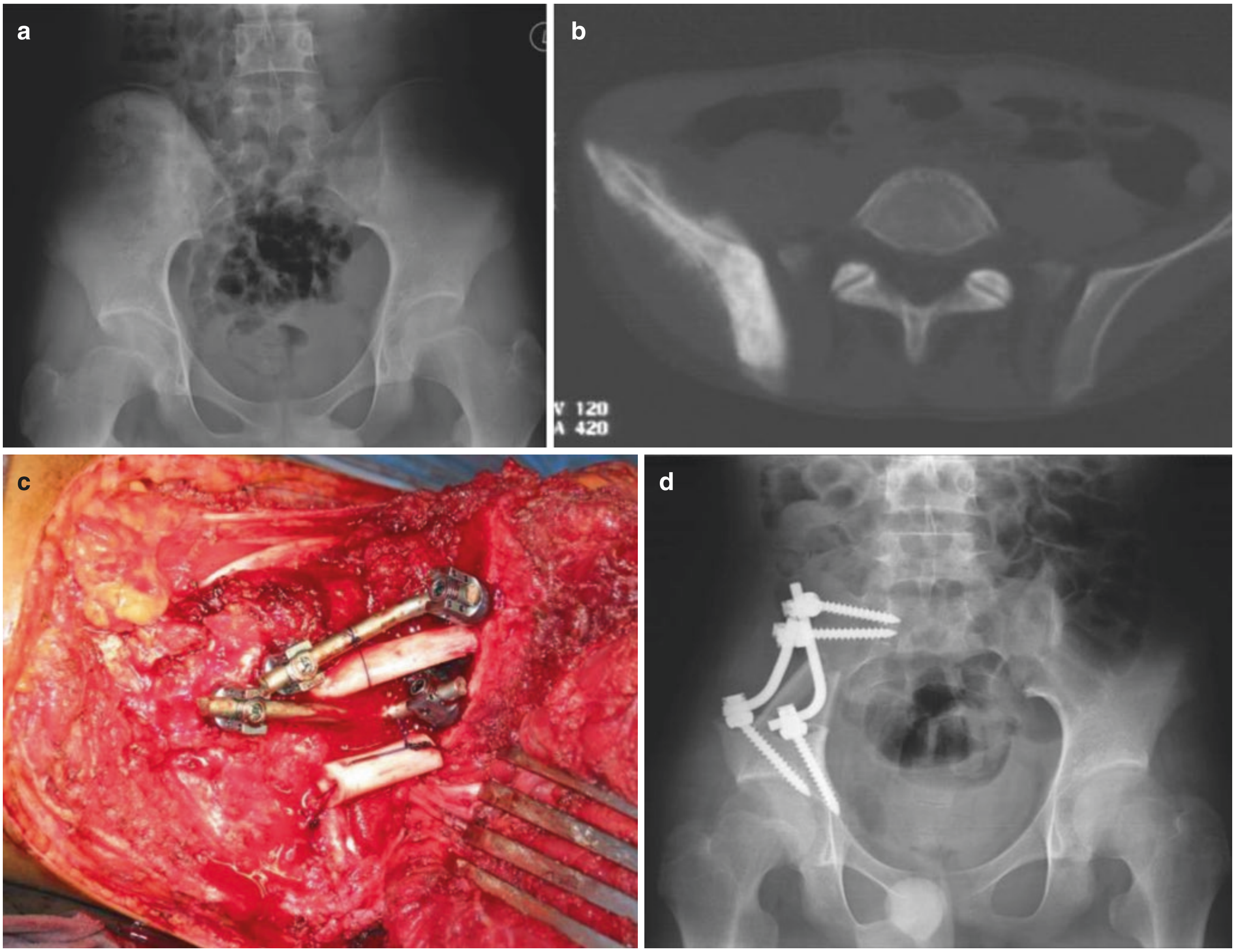

Fig. 12.2 The patient was a 14-year-old male with Ewing's sarcoma of right pelvis. Preoperative X-ray (**a**) and CT scan (**b**) showed the lesion. (**c**) Intraoperative photograph showed fibular graft combined with screw-rod system being used for reconstruction. (**d**) Postoperative X-ray

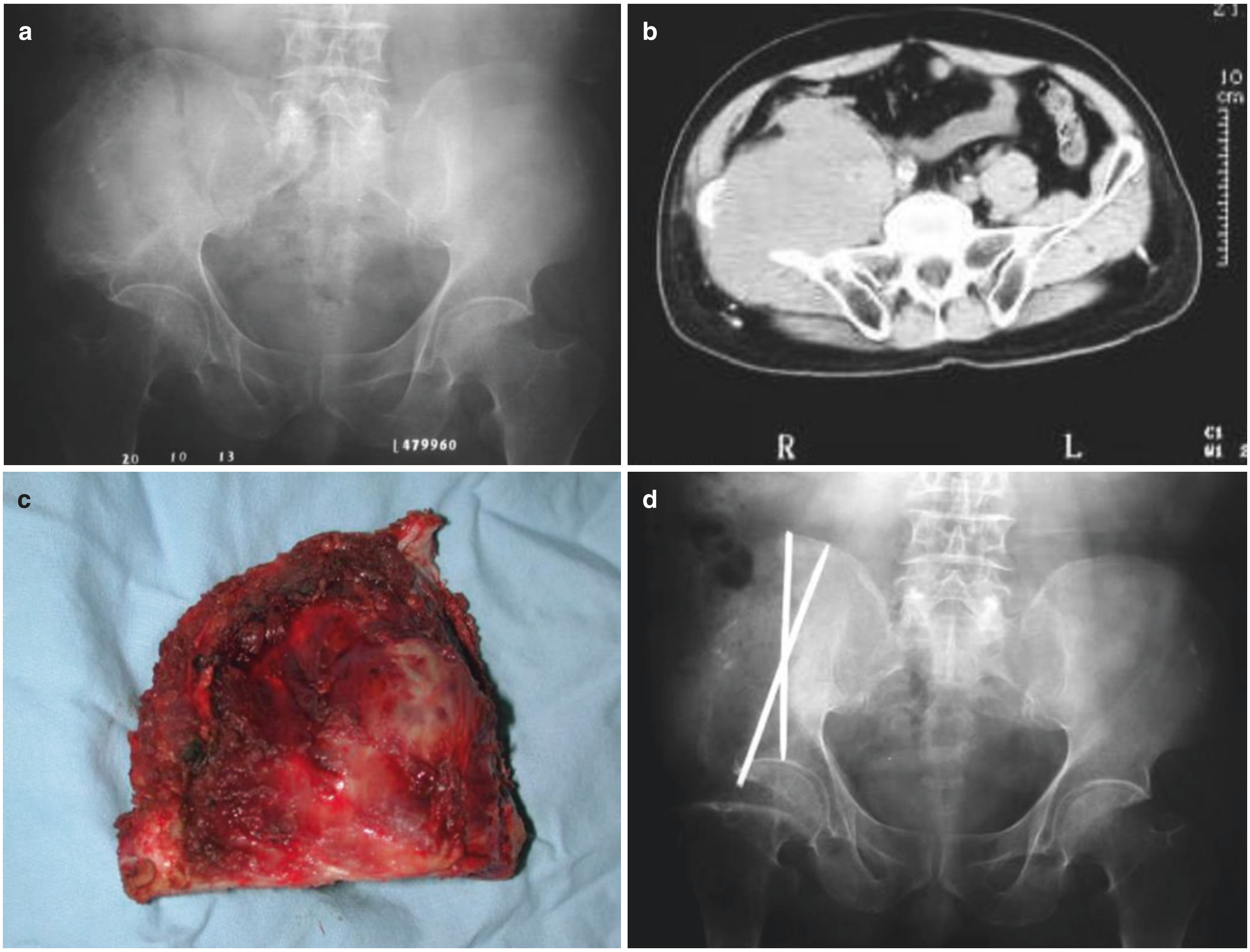

Fig. 12.3 The patient was a 58-year-old male with huge mass in the right pelvis. Multiple myeloma was diagnosed by core-needle biopsy (**a**, **b**). The tumor was resected (**c**), and Steinmann pins augmented with bone cement were used to reconstruct (**d**)

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13 Periacetabular Reconstruction

Tao Ji and Wei Guo

Approximately 10–15% of primary malignant bone tumors involve the pelvic girdle. The most common primary sarcomas in this location are chondrosarcoma, osteosarcoma, and Ewing's sarcoma. More commonly, 80% of patients with carcinomas have osseous metastases. Pelvic bone tumors are often large at presentation and proximal to organs, nerves, and vessels, which makes limb salvage and reconstruction difficult and challenging. Before the 1980s, hemipelvectomy was the standard surgical treatment for primary pelvic sarcomas. Such a procedure, however, removes a viable extremity to achieve local control. Additionally, hemipelvectomy is disfiguring and disabling. With the improvement in preoperative imaging techniques, neoadjuvant treatments, and surgical techniques, limb-preserving procedures have become more common in the past several decades. Enneking and Dunham proposed a tumor classification typically associated with four types of pelvic resections and/or reconstructions: Type I, the ilium; Type II, the periacetabulum; Type III, the obturator; and Type IV, the sacrum. Isolated resections of the ilium or ischium and pubis may not require reconstructive procedures to achieve excellent postoperative function. Type II resections require reconstruction to restore force transmission and weight-bearing along anatomic axes. The ideal pelvic resection would achieve an adequate tumor resection followed by a reliable and functional reconstruction with minimal morbidity. Adequate excision of Type II tumors often requires complete excision of the skeletal hemipelvis and large parts of the soft tissue of the pelvis. Several different reconstruction options have been proposed after this type of resection, including ischiofemoral arthrodesis or pseudoarthrosis, iliofemoral arthrodesis or pseudoarthrosis, massive allograft, autoclaved autograft, allograft prosthetic composite, custom-made endoprosthesis combined with hip arthroplasty, or the modular saddle prosthesis and 3D printing endoprosthesis. There are various options for reconstruction, each having advantages and limitations. Implantation of a megaprosthesis in early years has resulted in a high complication rate and a poor functional result. Major complications of megaprosthetic reconstructions, such as infection, loosening, and dislocation, occur frequently (approximately 25–35%).

13.1 Arthrodesis and Pseudoarthrosis

Iliofemoral and ischiofemoral arthrodeses or pseudoarthroses have been employed successfully for many years. The basic concept is to obtain continuity of the hip with residual pelvis by either fusion way or coaptation, achieving stability and weight-bearing potential. The method is associated with limb shortening. Another advantage of arthrodesis or pseudoarthrosis is the fact that resection can be done without looking at reconstruction. Hip transposition was developed to improve the stability of flail hip in pseudoarthrosis. By shortening of the leg, adequate soft tissue coverage can be achieved. The ideal candidate for iliofemoral fusion is a patient requiring periacetabular resection with or without pubis and ischium resection. Availability of most of the ilium minimizes limb length discrepancy and allows wider bone contact, enhancing fusion and stability. In arthrodesis, rigid fixation is usually achieved with a plate, and cable wires are used for pseudoarthrosis. Iliofemoral fusion is preferable for young and active patients because it provides stable and durable construct capable to withstand high-demand functional requirement. Iliofemoral pseudoarthrosis is indicated for more sedentary and older patients. The ability to flex hip seems to be appreciated by older patients with lower daily activity level. When the proximal osteotomy is above the neck of ilium, the iliac becomes thin which makes it difficult to obtain solid fixation and fusion. Pseudoarthrosis may be an option.

Ischiofemoral arthrodesis results in less shortening of the limb than an iliofemoral arthrodesis, and it is often associated with motion of the symphysis pubis. Pain in symphysis

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remained the main concern, and difficulty in achieving a fusion at the small area of bone contact limited the application. Overall, the fusion rate for iliofemoral or ischiofemoral was reported to be about 50–80%. The iliofemoral fusion results in good function while the result of iliofemoral pseudoarthrosis is unpredictable.

13.2 Pelvic Allograft or Devitalized Autograft

Satisfactory functional results have been reported after internal hemipelvectomy, despite at least a 50% prevalence of instability or shortening of the limb and complications such as failure of the arthrodesis. Several techniques have been developed for reconstruction of the hip and hemipelvis so that the patient retains a functional and cosmetically acceptable limb without shortening. These techniques include biologic and/or prosthetic reconstructions. Mnaymeneh and Mankin both reported cases of implantation of an osteoarticular graft after wide resection of the hemipelvis. The two cases showed the reconstructions did not fail within 5 years after surgery. Biologic reconstructions with allograft or replantation of the resected hemipelvis after it has been autoclaved have a number of advantages, including a readily available source, the ability to be size matched, and biologic union with host bone.

Early reports showed the allograft reconstruction after hemipelvic resection was feasible; however, postoperative x-ray showed evidence of localized resorption of bone at the proximal graft-host junction and progression. A comparison of the results of the use of long-bone allografts with that of pelvic allografts revealed that fractures of the former occur much earlier than those of the latter. No matter which fixation devices were used, none of fixation devices provided effective permanent splintage of the thin innominate bone. Many serious complications were associated with allograft including infection, nonunion, dislocation, fracture, nerve palsy, and local relapse. There were relatively few mechanical complications once the bone union was achieved. Although the rate of union after allograft reconstruction is reasonably high, and fractures of the graft in the pelvis are not common and may heal spontaneously, the high risk of other complications might suggest that this techniques should be used with strict indications. Rosenberg and Mankin [1] reported high rates of infection and recurrence after pelvic reconstruction with allograft. They reported that five of nine reconstructions with an osteoarticular graft failed, as did all of four reconstructions with an intercalary graft or an allograft-implant composite. Ozaki [2] reported that the allograft was removed usually because of infection after seven of nine reconstructions with an allograft-implant composite. Infection is a high risk of such type of surgery and is further increased by implantation of avascular biologic material such as allograft.

Reconstructions with autograft include devitalized tumor bone, fibular, or ipsilateral femoral autograft. Puget and Utheza described an option of reconstruction involving transportation of proximal part of the ipsilateral femur into the defect and implantation of a conventional total hip replacement in the autograft. The technique was further reported by Biau et al. [3]. Mechanical failure was the main complication and might be attributed in part to technical flaws and the so-called learning curve, also the inadequate fixation. They suggested the autograft to be stabilized with a plate and screws, with four cortices fixed at each extremity in the host bone, and a reinforcement acetabular ring should always be used. Femoral head autograft plus total hip arthroplasty has been reported to be used in reconstruction for periacetabular defect after tumor resection. In a pilot study of 13 patients who received femoral head autograft, the complication rate was reported to be 4 out of 13 [4].

There are several methods for recycling of the resected tumor bone, including autoclaving, freezing, pasteurization, extracorporeal irradiation, and alcohol devitalization, although the mechanical properties and osteoinduction may vary. Pasteurization is a proven effective method for devitalizing bones with tumor, and the devitalized bone usually integrates well with the host bone. Hypertonic saline (10%) can be used for pasteurization which can make a better preservation of protein. Surgeons from Asian countries usually choose the reconstruction using recycled tumor bone [5]. Complete union of pasteurized bone to the host bone was usually achieved at 1 year after surgery.

13.3 Endoprosthetic Reconstruction

A number of techniques have been described for the reconstruction of a periacetabular defect. Although associated with a significant reduction in range of motion, some authors prefer to perform biologic reconstruction including arthrodesis or pseudoarthrosis. However, failure to obtain a solid fusion is a frequent occurrence and results in a painful reconstruction with poor function outcome. The high failure rate of allografts and autografts due to nonunion, fracture, and graft resorption generates thoughts being given to endoprosthetic reconstruction, and a number of different types of endoprosthesis have been reported. Generally, dislocation and aseptic loosening are the two most common failure reason with prevalence of 12–22% and 3–12% retrospectively [6]. It is well accepted that reconstructing a pelvic defect with an endoprosthesis has the greatest potential to achieve a well-functioning limb. However, long-term mechanical failures are the major concern regarding endoprosthetic reconstruction.

The first attempts of endoprostheses to reconstruct resected pelvic bone and to restore the pelvic ring can be found in the early 1970s [7]. Scales and Rodney implanted a temporary spacer of acrylic cement and designed a steel prosthesis in the shape of resected iliac bone. The prosthesis was removed due to infection. The first case reported in literature about endoprosthetic reconstruction after pelvic tumor resection was in 1974 for a patient with chondrosarcoma. In early days, the preoperative plan was determined on x-rays. Later, attempts have been made to improve the accuracy of pelvic prosthesis design and production. The first report on endoprosthetic pelvis reconstruction was by Gradinger in 1993 [8]. The prosthesis was custom-manufactured from plain x-rays with low accuracy for intraoperative orientation of acetabulum. The anchorage into the remaining sacral ala or iliac bone was mainly provided by screws with additional plates or flanges, also the rigid fixation to the contralateral pubis with high shear forces. Ozaki et al. [9] reported a series of 12 cases of pelvic prosthetic reconstruction following tumor resection based on computer-aided design according to preoperative CT scan. Deep infection occurred in 3 of 12 patients. The overall survival of endoprosthesis at 3 years after surgery was 42%. Windhager et al. [10] reported a series of 21 consecutive cases of different reconstruction approaches. Nine of the 21 patients received a custom-made prosthesis with best functional results compared to saddle prosthesis and allografts (Fig. 13.1).

The main trend of pelvic endoprosthesis is modularity and iliac- or iliosacral-based fixation. With the advent of megaprosthetic reconstruction in pelvis, the custom-made fashion was the main design in early days. However, with development of modularity in endoprosthesis for limb salvage procedure in extremity, the modularity concept was introduced into pelvic endoprosthetic reconstruction. The symbolic design was reported by Guo et al. [11] in 2007.

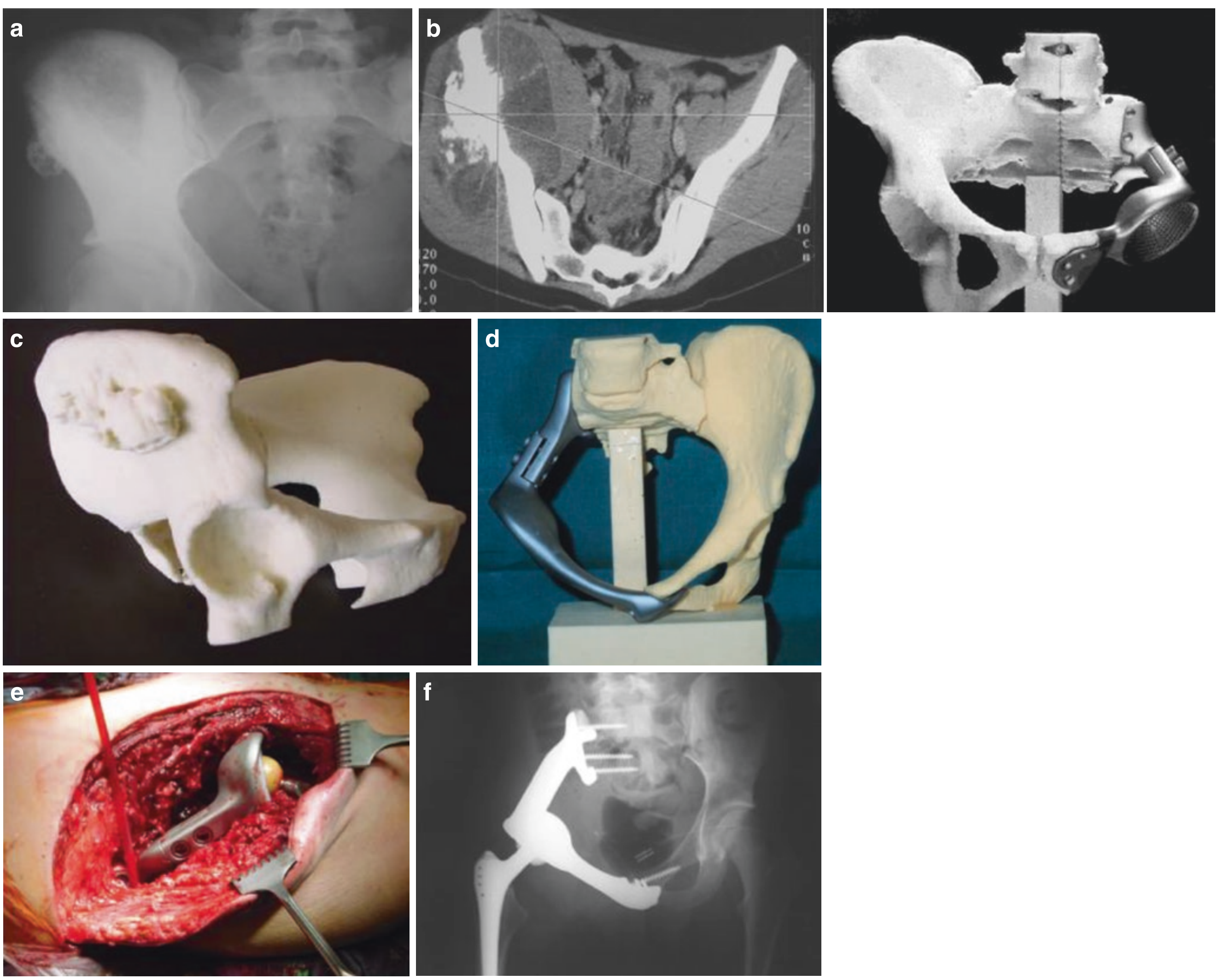

Fig. 13.1 The custom-made pelvic endoprosthesis by CAD approach. The planned resection area and custom-made endoprosthesis was fixed on the pelvic saw bone. The intraoeprative photo and postoperative x-ray was shown. (Copyright © Wolters Kluwer Health | Lippincott Williams & Wilkins)

There were three parts in the design: iliac fixation part that served as main fixation structure between prosthesis and residual bone, acetabular part which connected to iliac fixation part by Morse taper which is commonly used in modular limb reconstruction system, and pubic connection plate which was an optional part in reconstruction. The pubic connection part was rigidly connected to the contralateral pubis without any dynamic motion resulting in high breakage rate. The same results were found in custom-made endoprosthesis, and the high complication rate was approved by biomechanics study [12, 13]. In their midterm follow-up study, the pubic connection plate was seldom used; however, no higher fixation failure was observed [14]. Pedestal cup and ice-cream endoprostheses are both iliac-based fixation without structure to restore anterior pelvic ring. The advantage of modular pelvic endoprosthesis is the smaller size, which facilitates soft tissue coverage and may reduce infection rate. As for the fixation strategy, multiaxial fixation by screws is used in Guo's modular pelvic endoprosthesis which has a low breakage rate and loosening rate. Pedestal endoprosthesis has a cemented or press fit fixation technique, which is similar to megaprosthesis used for extremity. However, the lack of channel structure and cortical bone in ilium and iliosacral area brings difficulty in such fixation concept. It seems that it is well accepted that anterior pelvic ring should be left open in prosthetic reconstruction due to the rigidity at pubic connection area. Both intramedullary fixation (pedestal endoprosthesis) and extramedullary fixation (iliac flange fixation) exclude anterior ring restoration part (Figs. 13.2 and 13.3).

Both pedestal and Guo's hemipelvic endoprosthesis need at least a remaining part of the ilium and cannot be used for ilium removal. However, large pelvic tumor often involves both periacetabular and iliac part even with partial ipsilateral sacrum. Initially, Guo [15] attempted to reconstruct such defect by femoral head structural autograft to the sacrum and then the standard modular hemipelvic endoprosthesis. However, the early stability after surgery was inadequate for ambulation. Then pedicle screw system was used as adjuvant fixation to enhance the strength of the whole system (Figs. 13.4, 13.5 and 13.6).

The method achieved acceptable complication rates and favorable functional outcomes at a minimum follow-up of 15 months [16]. However, further follow-up of the patients identified a number of major complications, including rod breakage, prosthetic dislocation, and pedicle screw loosening, leading to substantial deterioration of lower limb function. As a result, a modified new generation of modular pedicle-hemipelvic endoprostheses with the aim of decreasing the complication rate and increasing the durability of the prosthesis was developed. The newer design is characterized by its enhanced fixation to the remaining sacrum aside from the simple connection to the lumbar spine. The newly designed prostheses achieve both fixation to the residual sacrum and connection to the lumbar spine. Additionally, the porous structure was three-dimensional (3D) printed on the medial surface to facilitate osseointegration and long-term stabilization. A double-axle component by sawteeth was also introduced to facilitate intraoperative adjustment of acetabular angle and acetabular anteversion angle. A series of 20 cases was reported recently. With the newly designed endoprosthesis, a favorable functional outcome (MSTS score 65%) was achieved with short follow-up. This appears to be better in comparison to the previous generation of pedicle-hemipelvic endoprostheses, which yielded an MSTS score of 58%. The new prosthesis offers enhanced modularity and more precise positioning of the acetabular cup compared with the previous design. The complication occurred in 3 out of 20 patients with one deep infection and two dislocations.

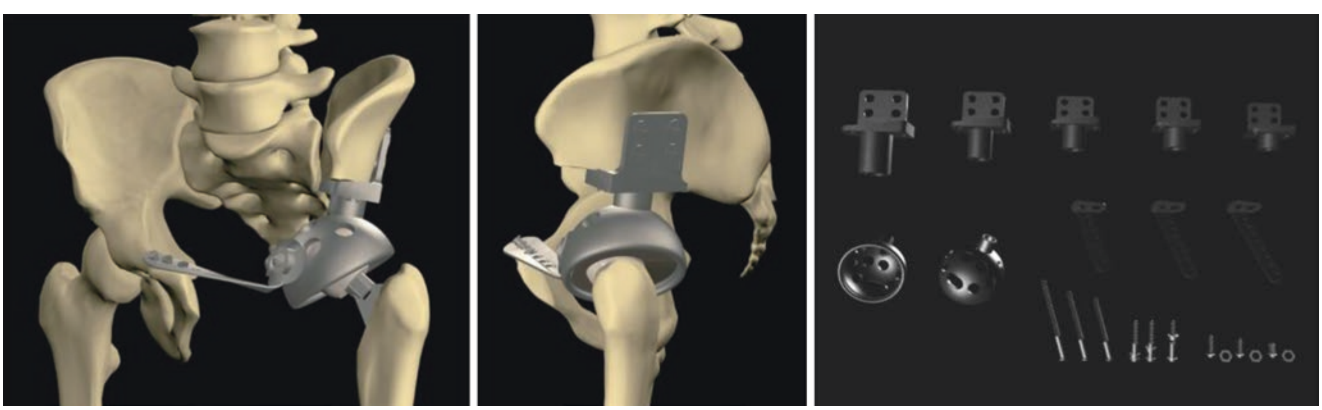

Fig. 13.2 The first-generation modular hemipelvic endoprosthesis which contains three parts: iliac fixation part, acetabulum part, and pubic connection plate

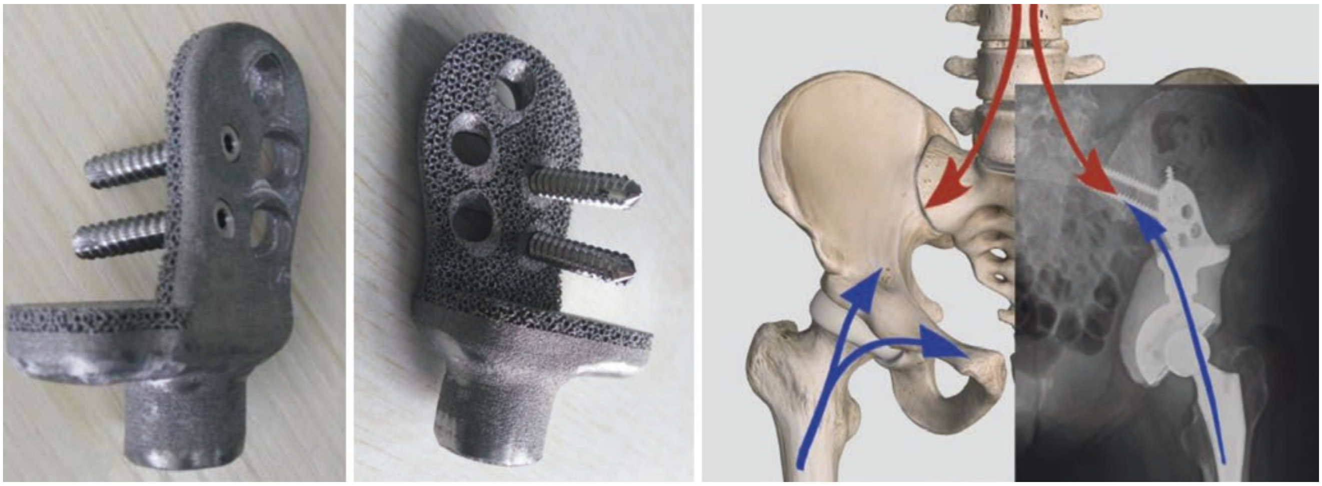

Fig. 13.3 The second-generation 3D printing-based modular hemipelvic endoprosthesis. The fixation location has been shifted from iliac osteotomy surface (first generation 2003–2015) to the iliosacral part with screws passing through iliosacral joint (second generation since 2016)

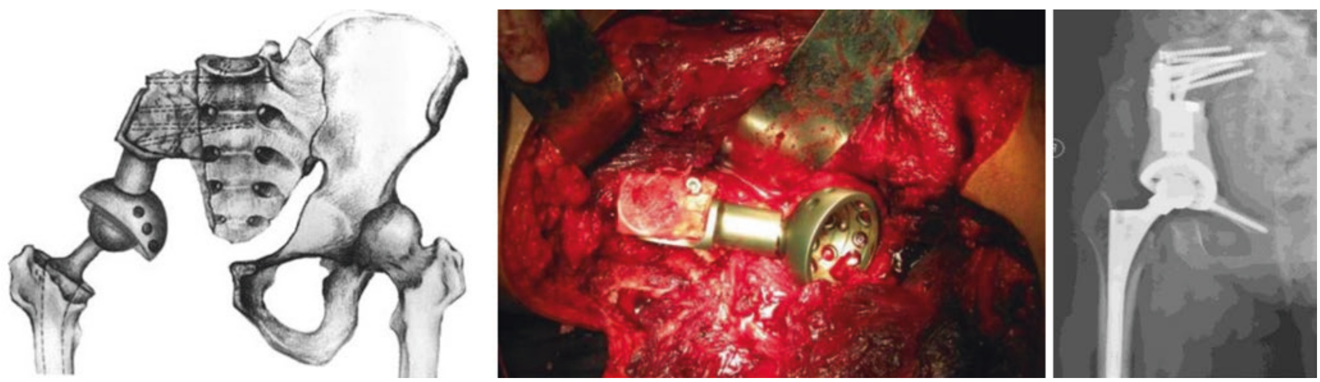

Fig. 13.4 Reconstruction with femoral head autograft and standard hemipelvic endoprosthesis after total internal hemipelvectomy

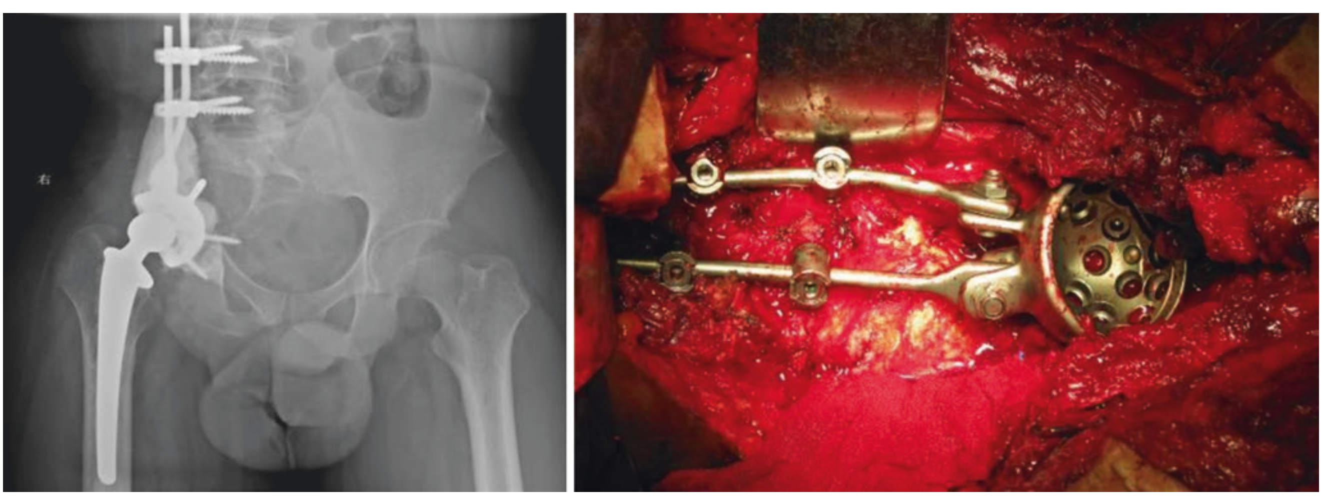

Fig. 13.5 Universal spine system was used to connect to the acetabulum to improve the stability of the reconstruction after ilium was removed

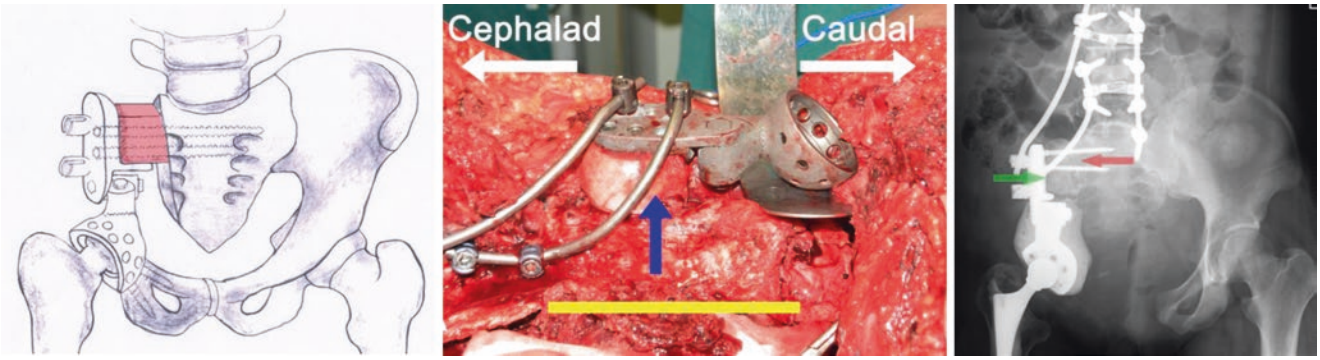

Fig. 13.6 The reconstruction after pelvic Type I + IV + II rsection. The autogenous femoral head was shaped into proper size and was fixed between the impalnt and osteootmy site at sacrum. The intraoeprative photo showed the connection of screw-rod system to the pelvic endoprosthesis. Also the postoepraive x-ray showed the whole reconstruction system

13.4 Saddle Prosthesis

The saddle prosthesis was designed by Nieder in Germany in 1979. Initially, this prosthetic concept was used for pelvic reconstruction of large acetabular defects following total hip arthroplasty. Since 1984, it is also indicated as replacement after resection of periacetabular tumors. The advantage of this method has been in the simplicity of its design, alleviating the need for an acetabular implant. However, the disadvantage is the need for postoperative immobilization to ensure soft tissue healing. Certain aspects of notch preparation are more challenging. Poor range of motion, dislocation, and progressive upward migration were common complications after saddle endoprosthesis reconstruction. Progressive erosion of bone and upward migration of the saddle resulted from the direct application of load and movement between metal and bone, but development of bone sclerosis at the interface led to stabilization of the saddle after a short-period migration. Migration was not found to be associated with osteoporosis, activity level, preoperative diagnosis, and site of implantation. Dislocation was reported to be ranging from 2% to 20% in literature. Heavy sutures will help secure the saddle component to the ilium. Also, optimal soft tissue tension balanced against excessive stretch on the neurovascular structures once a pseudocapsule forms and begins to ossify around the saddle component.

Stryker PAR (periacetabular reconstruction) endoprosthesis was designed to be secured with internal fixation and bone cement to the remaining ilium and support a reconstructed acetabulum. To address the previous mechanical complications found in Mark I and Mark II saddle prosthesis, loosening, migration, and dissociation, PAR endoprosthesis was developed which was a modular third-generation saddle prosthesis. The PAR endoprosthesis consists a wide iliac wing component that is secured to the ilium with cross bolts and cement, a constrained bipolar ball and socket joint, and a modular standard or endoprosthetic femoral stem. The complication rate of PAR endoprosthesis was reported to be 56%, and implant survivorship was 60% at 5 years. The dislocation rate was decreased to 12% [17] (Figs. 13.7 and 13.8).

13.5 Pedestal Cup

Pedestal cup (Zimmer, Freiburg, Germany) was originally designed for severe acetabular revision. It was firstly used in oncological condition since 2001 reported by Hipfl et al. in Vienna, Austria [18]. The so-called iliac stemmed-cone prostheses are effectively modified versions of the McMinn acetabular reconstruction component. In their retrospective review of a series of 48 cases reconstructed by stemmed pedestal cup (Schoellner cup, Zimmer Biomet, Inc.), a complication rater of 40% was found at a median follow-up of 6.6 years. Deep infection was the most common complication which affected 17% of the patients. The mean function score by MSTS 93 was 71%.

A modification type of pedestal endoprosthesis LUMiC® (implantcast, Germany) was introduced in 2003. The LUMiC prosthesis is a modular device, built of a separate cemented or uncemented stem with HA-coated and acetabular cup. The cup is also available with silver coating for anti-infection effect. The cup is connected to the stem by sawteeth allowing for rotational adjustment of cup position after implantation of the stem. A multicenter study during 6 years including 47 patients showed a dislocation rate was 13 for single time and 9% for recurrent dislocations. The infection was the most common type of complication which was 28%.

"Ice-cream" cone reconstruction of the pelvis was developed in 2003 by Stanmore Implants, and the system was named as coned hemipelvis. The concept was based on the old design of the McKee-Farrar stemmed hip replacement and has become known as the "ice-cream" cone prosthesis, as it looks like an inverted ice-cream cone. The prosthesis is inserted into the remnant of the pelvis and surrounded by antibiotic-laden bone cement. The overall complication rate was 37% with dislocation as the most common type (14.8%), followed by deep infection (11.1%) [19] (Fig. 13.9).

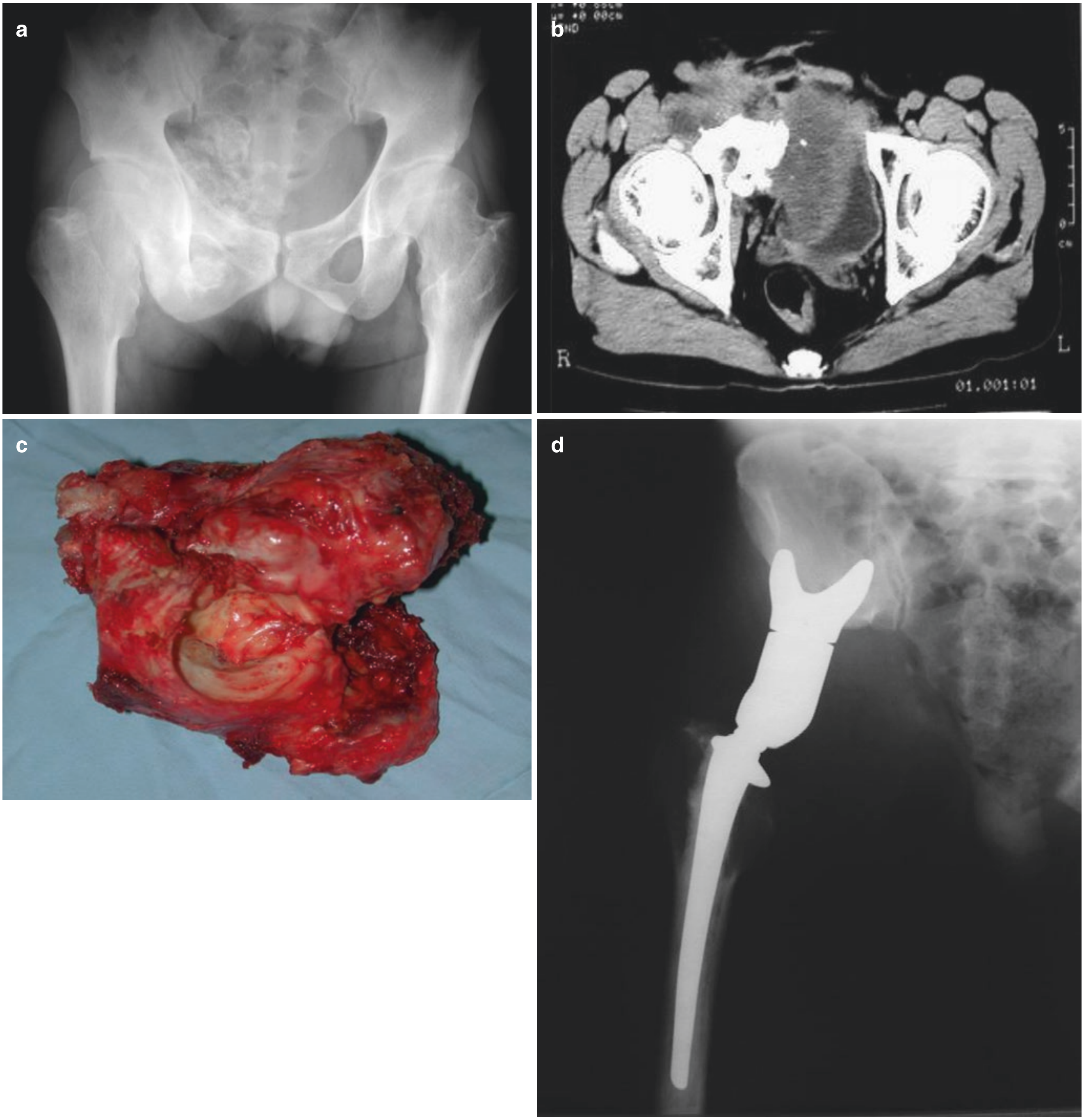

Fig. 13.7 The patient was a 34-year-old male with chondrosarcoma in the right pelvis involving the acetabulum, pubis, and ischium. Preoperative radiography (**a**, **b**) showing the tumor mass. (**c**, **d**) Intraoperative photos and gross specimen after type II + III resection. (**e**) Postoperative pelvis AP showing the link saddle prosthesis was in position

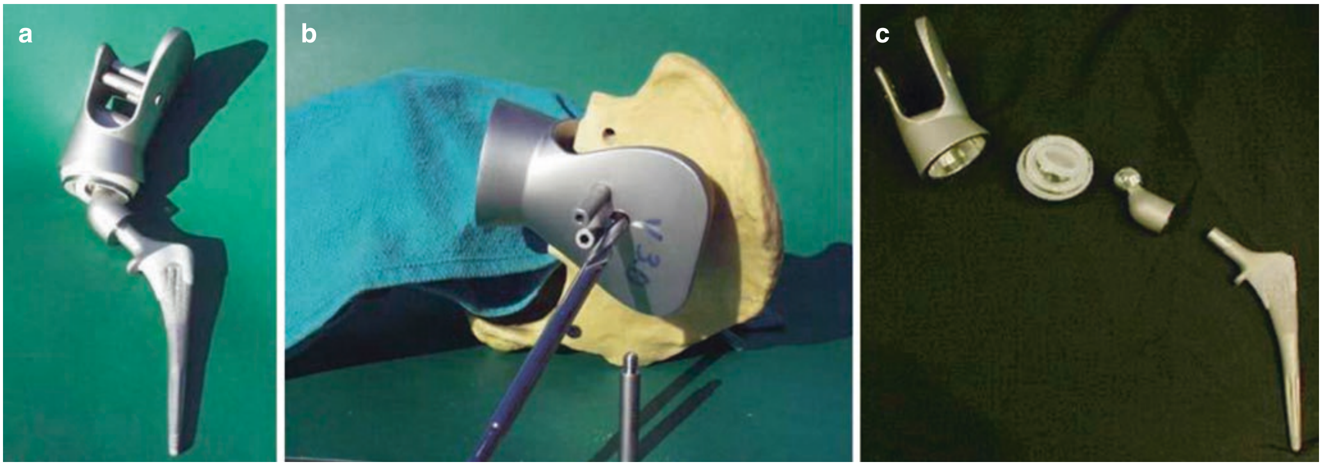

Fig. 13.8 The PAR endoprosthesis

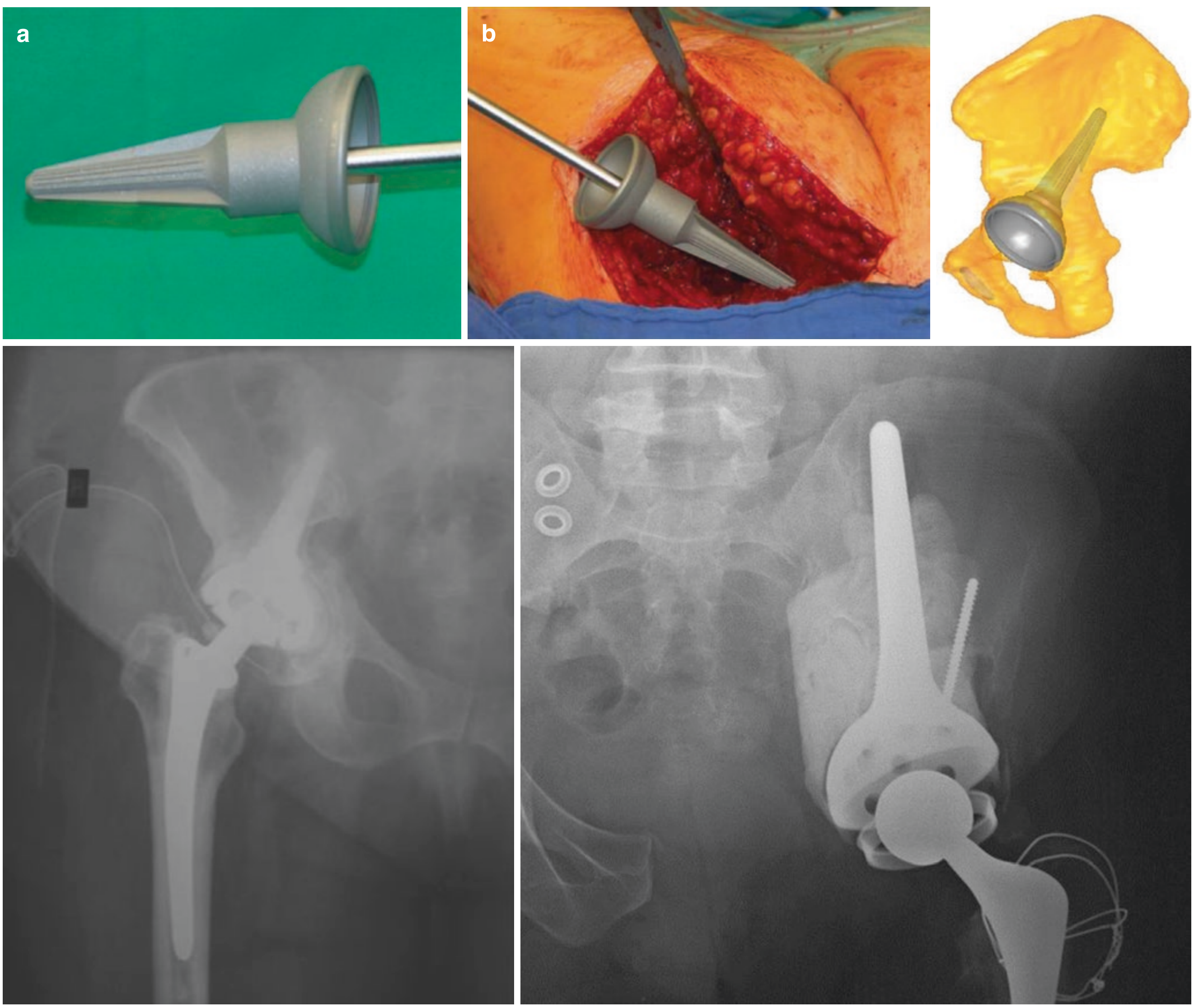

Fig. 13.9 The pedestal endoprosthesis reconstruction after periacetabular tumor resection (Copyright © Wolters Kluwer Health | Lippincott Williams & Wilkins)

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14 Reconstruction of Pelvic Tumor with Sacrum Involved

Yidan Zhang and Wei Guo

14.1 Fundamental Principles of the Reconstruction

1. No compromise to the surgical margins
   In pelvic tumor cases involving the sacrum, unexpected accidents including massive hemorrhage may prevent surgeons from keeping to the surgical margins. In addition, orthopedic surgeons are apt to emphasis too much on functional reconstruction when performing limb salvage surgeries. In that way, surgical margins are sometimes compromised to facilitate instrumentation and implant fixation. For example, when malignant pelvic tumor invades great proportion of the ipsilateral sacral wing, opening of the sacral canal and ligation of certain nerve roots are inevitable under these circumstances. However, these procedures may lead to severe destruction to the lumbosacral conjunction and functional loss of the sacral nerves. So intralesional curettage after bulk resection is sometimes adopted to save more bones, which are necessary to prosthesis or allograft anchoring. It might be applicable in benign tumors, but if the neoplasm is malignant, a second chance for surgical resection is unquestionably mere as recurrence occurs.
2. Recovery of limb length
   It is controversial whether reconstruction of pelvic girdle integrity and recovery of suffered limb length be beneficial to the patient. Some surgeons preferred not to reconstruction of pelvic girdle after iliosacral resection due to prolonged surgical duration and poor lower limb function led by injuries to the lumbosacral plexus. Other surgeons are used to iliofemoral arthrodesis or hip transposition to stabilize the salvaged limb [1, 2]. One of the major drawbacks of these procedures is that the suffered limb is dramatically shortened. And the patient has to face more difficulties in practicing standing and restoring normal gait function. Hence, although much of the muscle origins are cut off and the muscle innervations are destroyed by the nerve sacrifice, reconstruction of pelvic defect and limb length recovery can still benefit the patients' rehabilitation. Moreover, patients tend to participate in social life more easily with less mental disturbance.
3. Restoring of lumbopelvic continuity and three-column stabilization of lumbosacral conjunction
   Besides from the disruption of the sacroiliac joint, lumbosacral discontinuity and spinal instability caused by partial or total sacral defect are also challenging to the orthopedic surgeons. The lumbosacral conjunction is considered as a key structure responsible for strength loading of the body weight on the pelvic girdle. According to the three-column concept [3], the lumbosacral conjunction is unstable when the vertebral body of the sacrum, the anterior lumbosacral ligament, or the lamina and the spinal process of the sacrum is resected. Although techniques for lumbopelvic reconstructions after total or subtotal sacrectomy have inspired the surgeons in reconstructions after sacropelvic resections [4], the classic pedicle screw and rod system could not thoroughly make three-column stabilization to the major sacropelvic defects due to lack of screw insertion sites. Introduction of computer-assisted design and manufacture of sacropelvic implant may provide solutions to this problem.
4. Preservation of hip range of motion
   The acetabulum is frequently involved in massive tumors of the sacropelvic region. It is extremely difficult to restore the function of both the sacroiliac joint and the hip joint. Some surgeons believe that arthrodesis and pseudarthrosis repair should provide superior stabilization comparing to prosthetic reconstructions by sacrificing hip mobility. However, as great progress has been made in the prosthesis design and manufacture, range of motion of the hip joint could be preserved under most circumstances. This would not only prevent limb discrepancy but also facilitate muscle strength recovery of the lower limb

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through early rehabilitation. Complications including hip dislocation and mechanical failure may occur. Improvement in surgical techniques and prosthetic design should help preservation of the hip joint function in the future.

5. Avoidance of endoprosthetic-related complications
   Endoprosthetic-related complications are common. As for the sacropelvic reconstruction, endoprosthetic loosening, deep infection, dislocation, and neuralgia are all catastrophic events which can lead to a secondary surgery. Particularly, neuralgia is sometimes inevitable after resection of massive tumors and stimulation of lumbosacral bundles, while endoprosthetic-related neuralgia is evitable in most cases if neural interference with the prosthesis is successfully avoided. Herein, transient and persistent neuralgia could be prevented with subtle surgical procedures and dedicate prosthetic designs.
6. Prepare for revisions
   No matter the orthopedic surgeon choose reconstructions using allograft or prosthesis, revision may happen particularly in young patients, in whom physical demand is fairly high and structural complications including screw loosening and rod breakage are common. For the purpose of facilitating secondary revision surgeries, it is noticed to adjust mechanical conduction of the prosthesis or allograft-prosthetic composite and optimize the prosthetic design. To some extent, modular hemipelvic prosthesis has its advances in easy part changing, which could avoid unnecessary extensive exposure in a secondary revision surgery. Besides, introduction of computer-assisted design for a custom-made modular prosthesis may further help in a revision surgery.

14.2 Reconstructions Under Different Conditions

1. Partial sacral wing defect with continuous sacroiliac joint
   Such minor defect usually appears after intralesional resection of benign or invasive tumors involving the sacroiliac joint. Both ilium and sacrum are destructed, while anterior bone cortex and strong sacroiliac ligaments stay intact. Whether the mechanical reconstruction is necessary fully depends on the surgeon's experience and the patient's physical demand.

   Reconstruction methods include bone grafting with allogeneic or autogenic bones, bone cement filling with or without internal fixation such as cancellous screws, or pedicle-iliac screw and rod system. As for younger patient, biological reconstruction is preferable. Mechanical strength of weight-bearing tends to fully recover after ideal bone fusions (Fig. 14.1). As for invasive tumors or lesions with high recurrent propensity, bone cement filling is comparatively more feasible. Local recurrence is easily verified by distinct osteolytic appearance surrounding the bone cement in lesions such as giant cell tumor (Fig. 14.2). Moreover, the mechanical strength is enough for weight-bearing and physical activity shortly after the surgery.

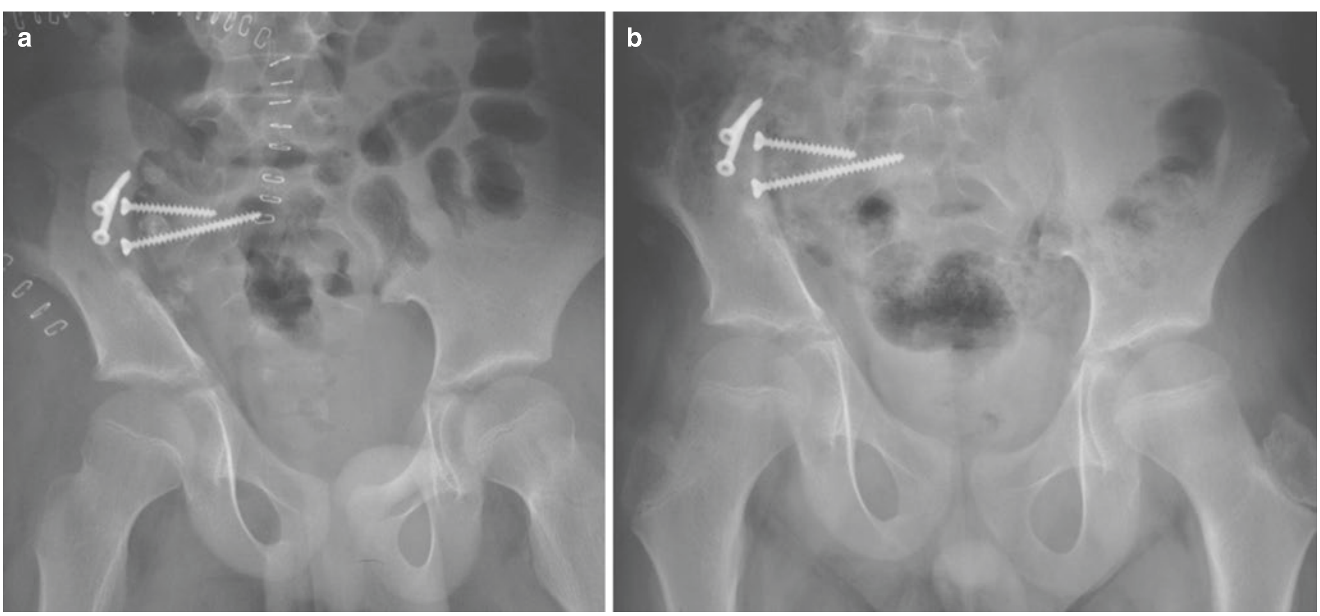

Fig. 14.1 A 12-year-old boy diagnosed with sacroiliac angiolipoma. After intralesional type I + IV resection, the biological reconstruction with the autogenic bone of the ipsilateral ilium was performed. Cancellous screws were also inserted for fixation of this structural reconstruction (**a**). Bone fusion was achieved at the 6-month follow-up (**b**). A PKUPH case

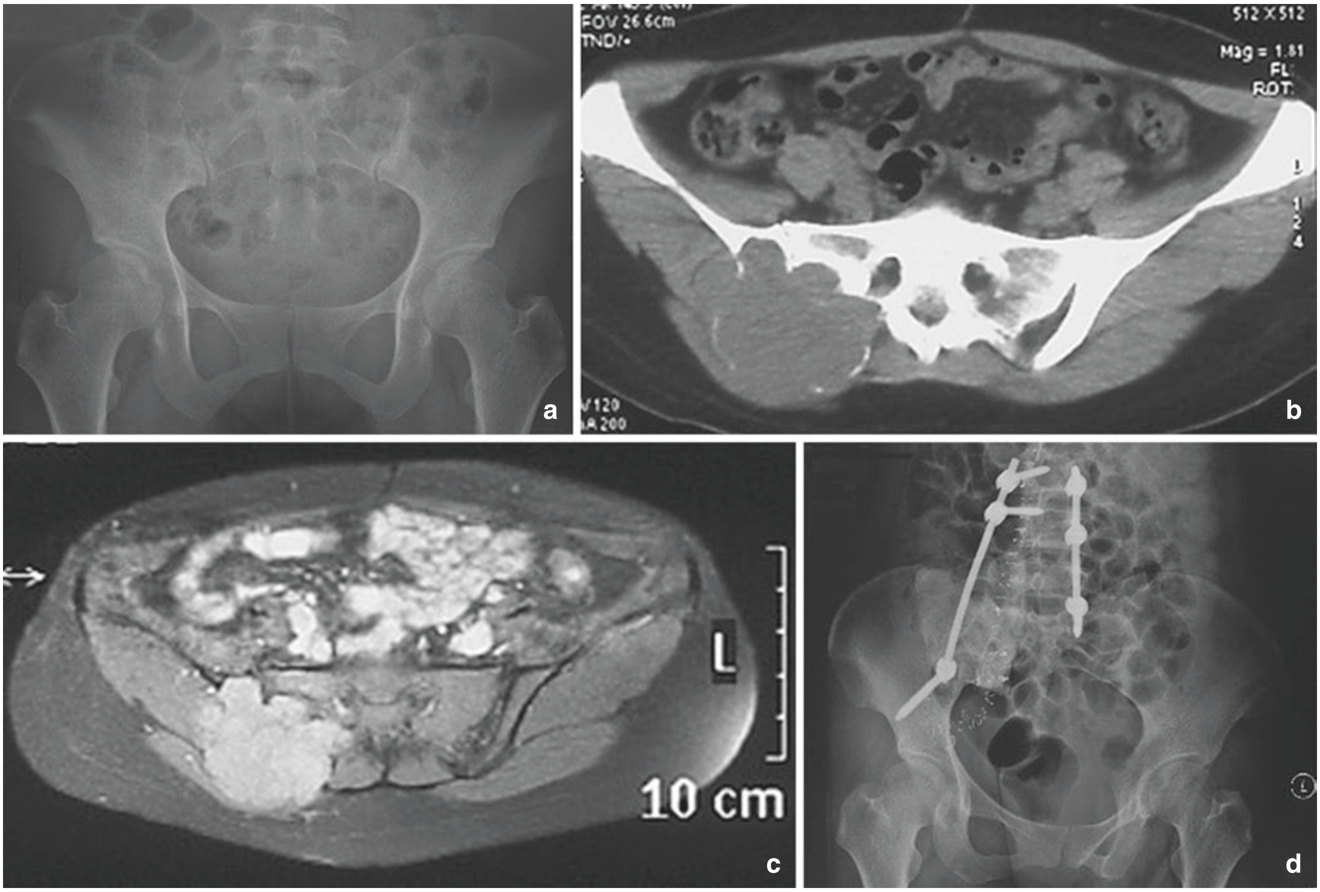

Fig. 14.2 A 38-year-old female diagnosed with sacroiliac giant cell tumor (**a**–**c**). After the intralesional resection and curettage, bone cement was adopted for the defect filling. Pedicle-iliac screw and rod internal fixation was also performed to enhance the lumbosacroiliac conjunction (**d**). A PKUPH case

2. Sacral wing defect with complete sacroiliac joint destruction

   Reconstruction of this type of sacroiliac defect is similar to that of partial iliac defect. The sacroiliac continuity or lumbopelvic conjunction should be restored by means of bone grafting and internal fixations. Nishida et al. prefer to adopt vascularized iliac bone graft for the reconstruction of iliosacral bone defect [5] (Fig. 14.3).

   Aydinli et al. chose autogenic fibula as the structural bone graft for reconstruction within the gap between the residual ilium and the sacrum. The screw and rod system was used to enhance the mechanical strength (Fig. 14.4) [6].

   Unlike the solitary iliac defect, difficulties in reconstruction for concurrent iliac and sacral defects include the following: (1) lack of anchoring sites for the screws in sacral ends due to major sacral bony defect and (2) injuries to the neighboring lumbosacral chunk easily causing loss of neural functions and neuralgia. Herein, although the lumbosacral conjunction is primarily intact, the direct lumbopelvic instrumentation is obligatory.

   Guo et al. led in the reconstruction using pedicle screw system for major sacroiliac defect of total iliac loss (Fig. 14.5) [7, 8]. Two screws were respectively inserted into the superior ramus of pubis and the ramus of ischium through the osteotomy plane above the ceiling of the acetabulum. By aligning these screws with the pedicle screws with double rods and enhancing the acetabular ceiling with bone cement, the mechanical strength for weight-bearing is fully achieved. The precision of screw insertion is essential. Besides from judging the screw direction by anatomical landmarks, intraoperative CT scan was also a powerful manner for precise screw insertions.

   If the residual sacrum is enough, a pedicle screw could also be inserted into the sacrum resulting a lumbosacropelvic fixation (Fig. 14.6).

   Ogura et al. also adopted lumbopelvic fixation with screw and rod system [9]. However, screws were not inserted into the pubis and ischium, while double-barrel vascularized fibula was counted on for combination with biological reconstruction instead of bone cementing. Bone union was achieved in five of eight patients (63%)

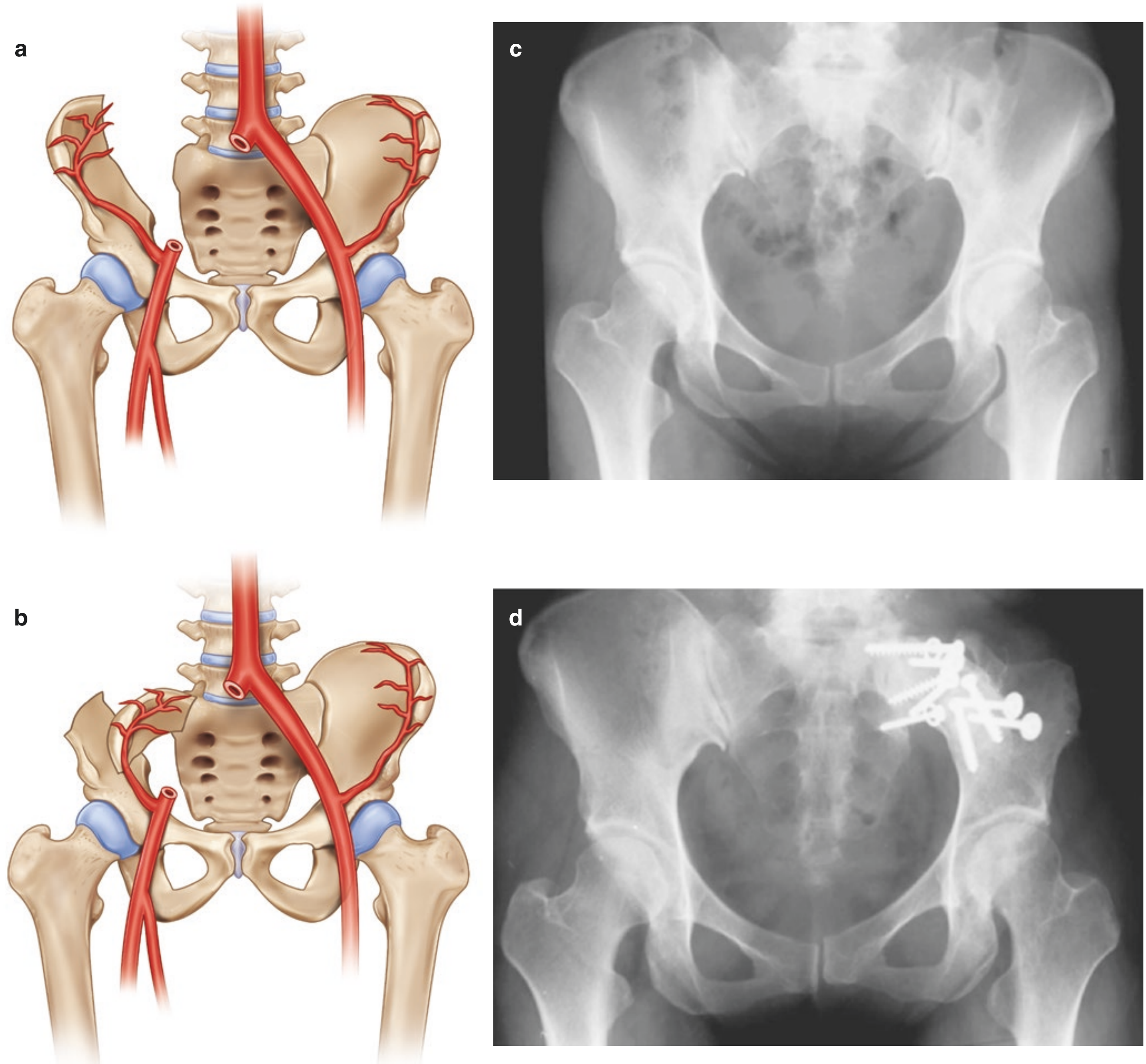

Fig. 14.3 The diagrams show reconstruction using a vascularized iliac graft (**a**) before and (**b**) after the iliac crest transfer. (**c**) A radiograph of the pelvis shows an ill-defined osteolytic lesion in the left ilium. (**d**) A plain radiograph taken 3.5 years postoperatively shows a well-united, transferred iliac graft

over a 1-year follow-up. There were three early postoperative complications: two deep infections resulting in graft removal and one implant failure resulting in nonunion. Among three patients, two developed scoliosis within 5 years. One patient developed lumbar disc hernia as a result of scoliosis, for which surgical intervention was required. At PKUPH, we adopted a 3D-printed sacroiliac endoprosthesis (Fig. 14.7) for reconstruction of sacroiliac defect and had demonstrated its durability in the short term.

Besides from instrumentations, C. Hoffmann et al. established hip transposition as a universal tool for periacetabular tumors excelling in small foreign parts and resulting in acceptable complication rates with good functional outcome [2]. However, its feasibility when the sacrum is involved needs further validation.

3. Subtotal sacral resection with complete sacroiliac joint destruction
   After a subtotal sacral resection, the lumbosacral conjunction is so weak and unstable that direct sacropelvic

Fig. 14.4 (**a**) Preoperative radiologic workup image suggests a mass involving the right iliac bone and sacroiliac joint. (**b**, **c**) Radiographs and 3D CT scans 3 years after type I + IV pelvic resection

reconstruction is difficult to achieve. Lumbopelvic reconstruction is the only method eligible. Instrumentation using bilateral pedicle-iliac screw and rod system remains predominant. Unlike minor iliac defect after sacrectomy, the iliac defect is major, and the residual bone ilium is usually lack of posterior iliac crest, which renders a great proportion of mechanical strength for screw anchoring and weight-bearing. Alternative methods of screw fixation in the residual ilium include inserting the screws into the iliac crest or more stably into the body of the ilium upon the weight-bearing surface of the acetabulum (Fig. 14.8). Transverse rod connectors or transiliac bars may be necessary for the prevention of the pelvic separation. And bone grafting is also preferable, aiming at lumbopelvic fusions, which could stop descent of the lumbar spine in the long term.

As for the hindquarter amputations along with subtotal sacrectomy, the contralateral sacroiliac joint needs stable reconstruction for weight-bearing of the salvaged limb. Ehud Mendel et al. chose to use autologous vascularized femur and fibula bone flaps harvested from the amputated lower extremity for lumbopelvic reconstruction [10]. At the 18-month follow-up, he was able to ambulate with the assistance of his custom-made prosthesis.

4. Bilateral sacroiliac discontinuity after internal hemipelvectomy and total sacrectomy

Extraordinarily massive tumor tends to invade the total hemipelvis and the total sacrum. En bloc surgery leaves the lumbar spine, and the uninvolved hemipelvis and the salvaged limbs totally separated. Though the nerves innervating the lower limb might be cut or damaged due to extensive surgical interventions, many patients still refuse to be amputated while the prognosis is unfortunately dim. Under this condition, the aim of reconstruction is to stabilize those anatomical structures in a manner that provides a one-stage solution for the defect and

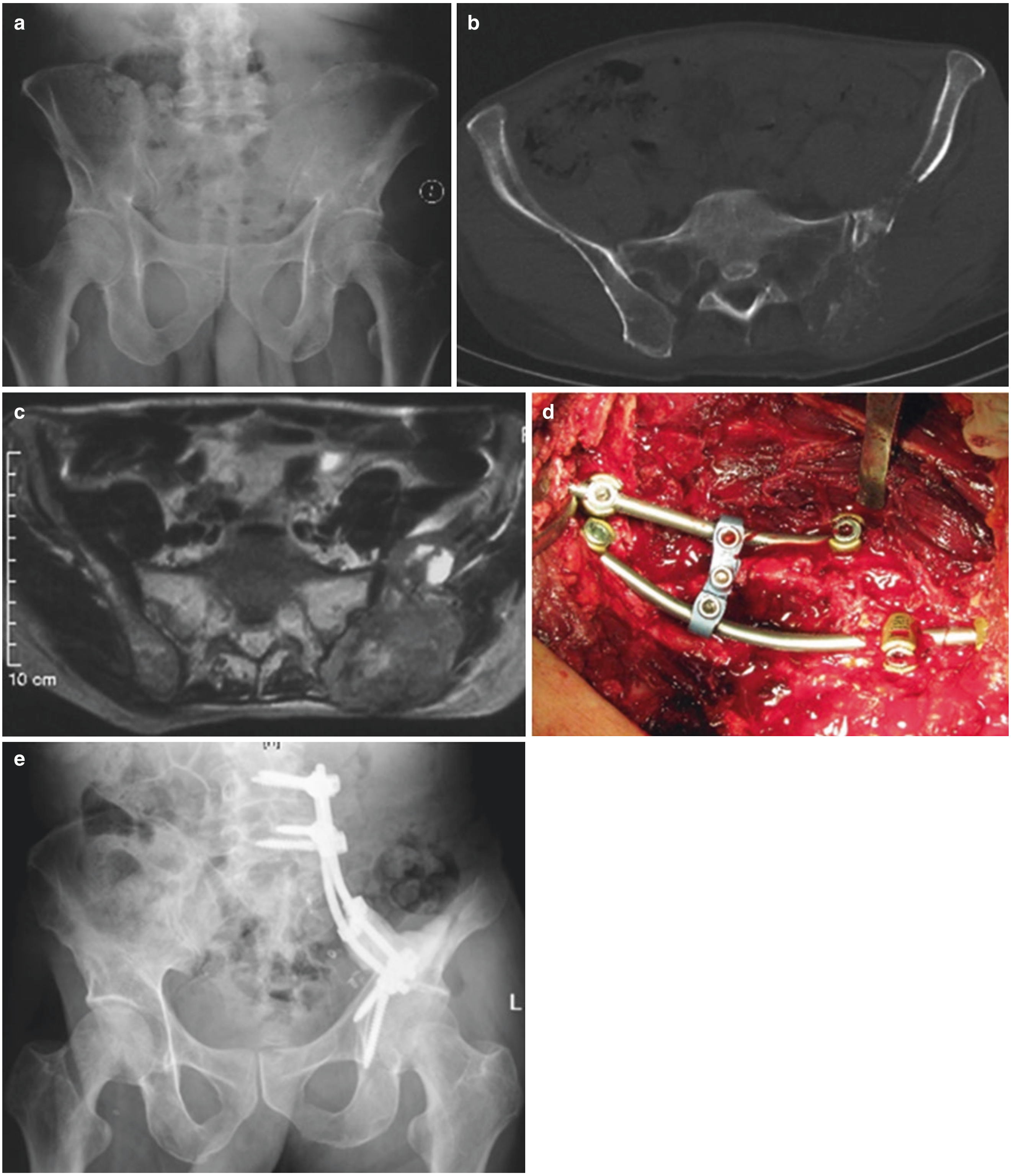

Fig. 14.5 A 69-year-old male diagnosed with poorly differentiated sarcomatoid carcinoma (**a**–**c**). After subtotal resection of the ilium and ipsilateral sacral wing, the lumbopelvic conjunction was reconstructed with pedicle-pubic-ischial screw and rod fixation system and bone cementing. Additional pedicle screws were also inserted laterally into the lumbar vertebra to provide additional anchoring sites for titanium rod (**d**, **e**). A PKUPH case

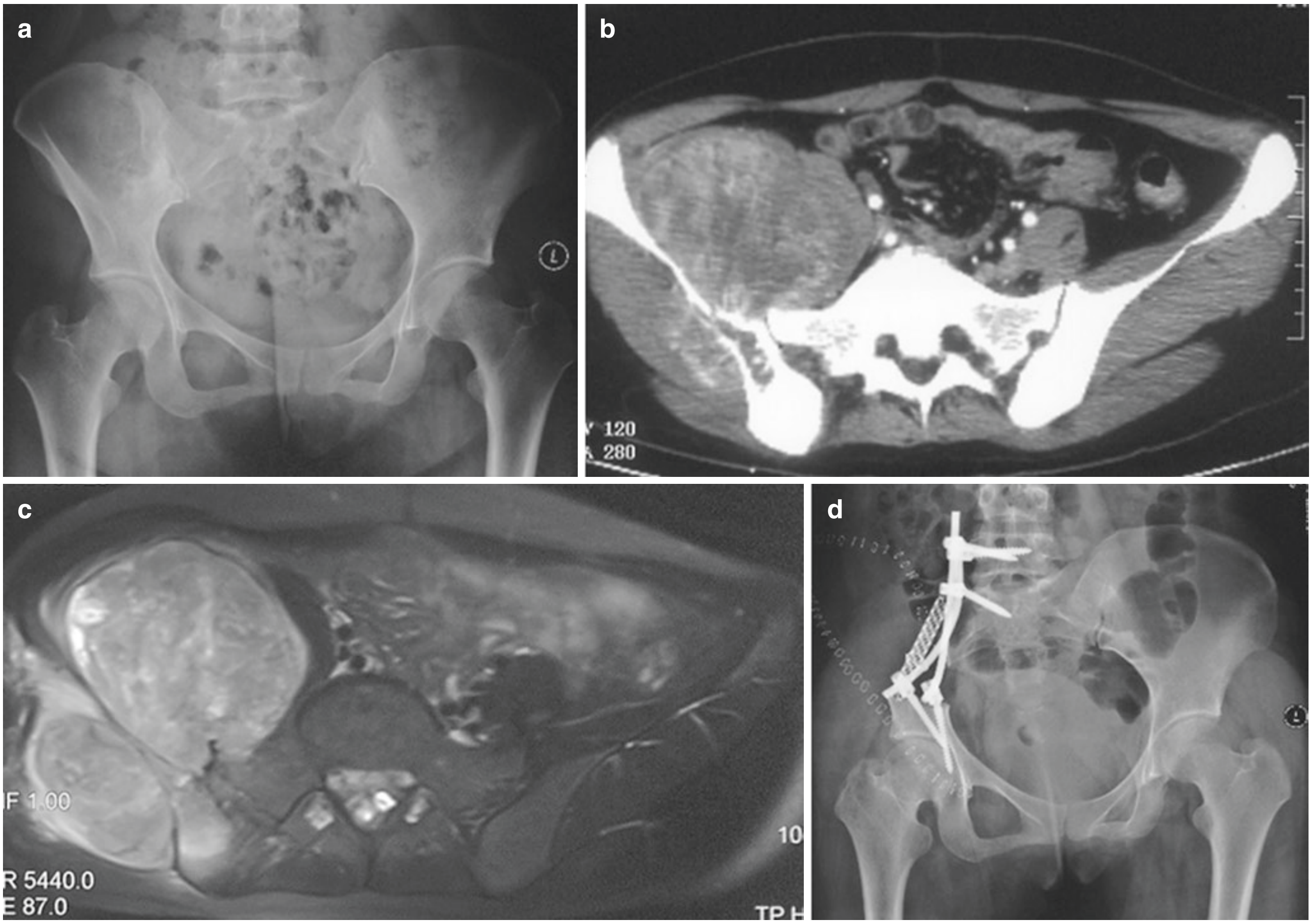

Fig. 14.6 A 28-year-old female was diagnosed with Ewing's sarcoma (**a**–**c**). After resection of the ilium and partial sacrum, the lumbosacropelvic fixation was performed and an allografted titanium mesh cage was stuck between the gap, in order to facilitate bone union (**d**). A PKUPH case

minimize the related complications as much as possible. Favorably, the reconstruction should relieve pains regarding the pelvic instability and give chances to a better rehabilitation (Fig. 14.9).

14.3 Development in Prosthetic Reconstruction of the Hemipelvis with Hip Involvement

The *pseudarthrosis*, femorosacral arthrodesis, and hip transposition all lead to irreversible limb discrepancies, which affect patient's normal gait significantly. Though the early periacetabular prosthesis such as the saddle prosthesis and the pedestal prosthesis could restore most of the hip range of motion, they are not eligible for reconstruction with major defect involving the sacrum, in that they both anchor to the residual ilium and rely on continuity of the sacroiliac joint. Herein, it relies on development in newly designed hemiprosthesis to achieve such major defect involving both the sacroiliac joint and the hip joint. Early solutions include caudal-flanged hip cage connected to pedicle screw and rod system (Fig. 14.10) and custom-made megaprosthesis [2]. Novel prosthesis used for hemipelvic reconstruction including the hip joint includes modular hemipelvic prosthesis connected to pedicle screw and rod system (Fig. 14.11) [11]. For more extensive resections when part of the sacrum is included, standardized surgical procedures were proposed (Fig. 14.12), and an updated version of the hemipelvic prosthesis was demonstrated to be durable in the short term (Fig. 14.13) [12, 13].

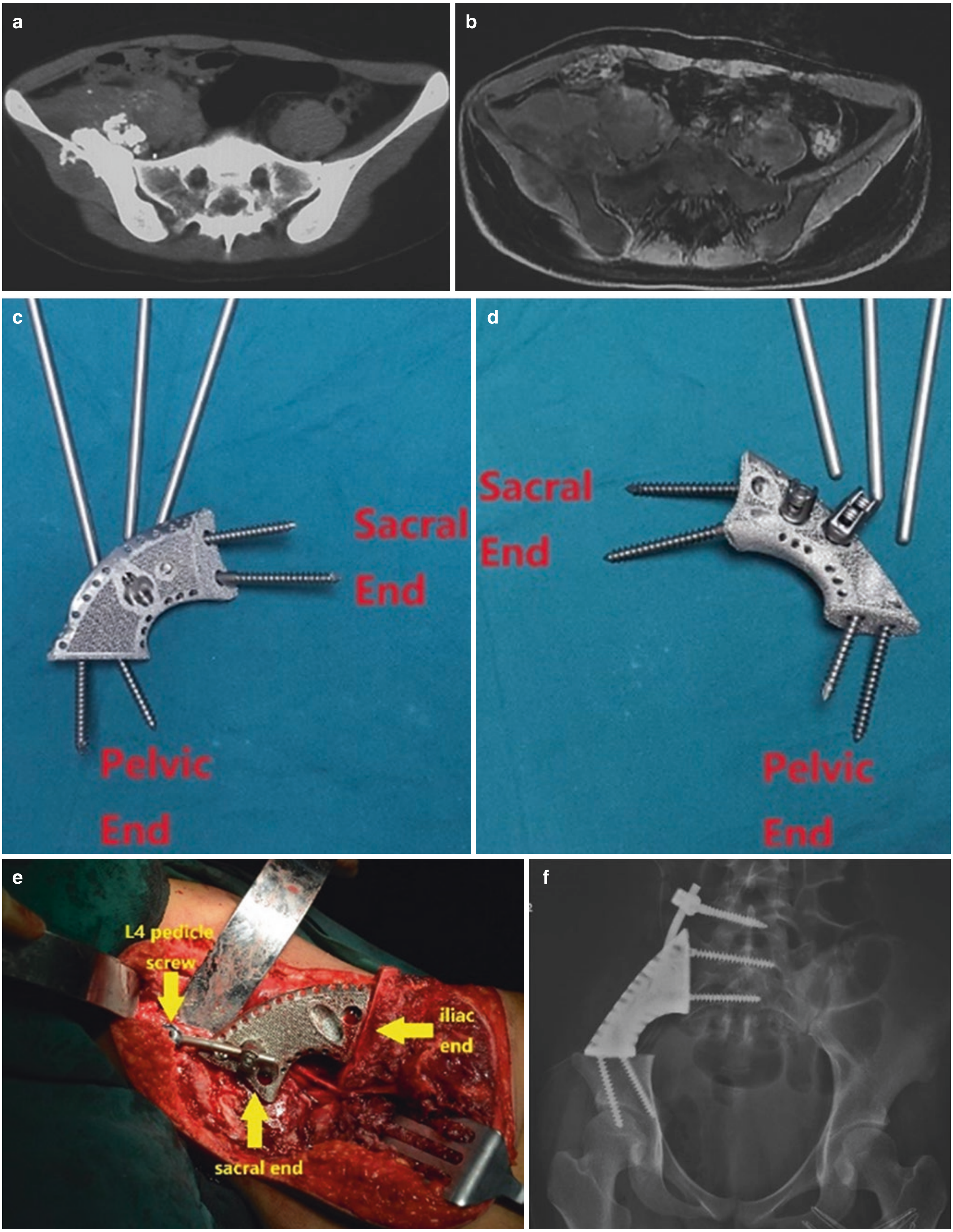

Fig. 14.7 An 18-year-old female was diagnosed with osteosarcoma (**a**, **b**). After resection of the ilium and partial sacrum, a 3D-printed titanium iliac implant (**c**, **d**) is fixed to the residual ilium and sacrum using four cancellous screws. Another lumbar pedicle screw was connected to a multiaxial screw within the implant to provide additional stabilities (**e**, **f**). A PKUPH case

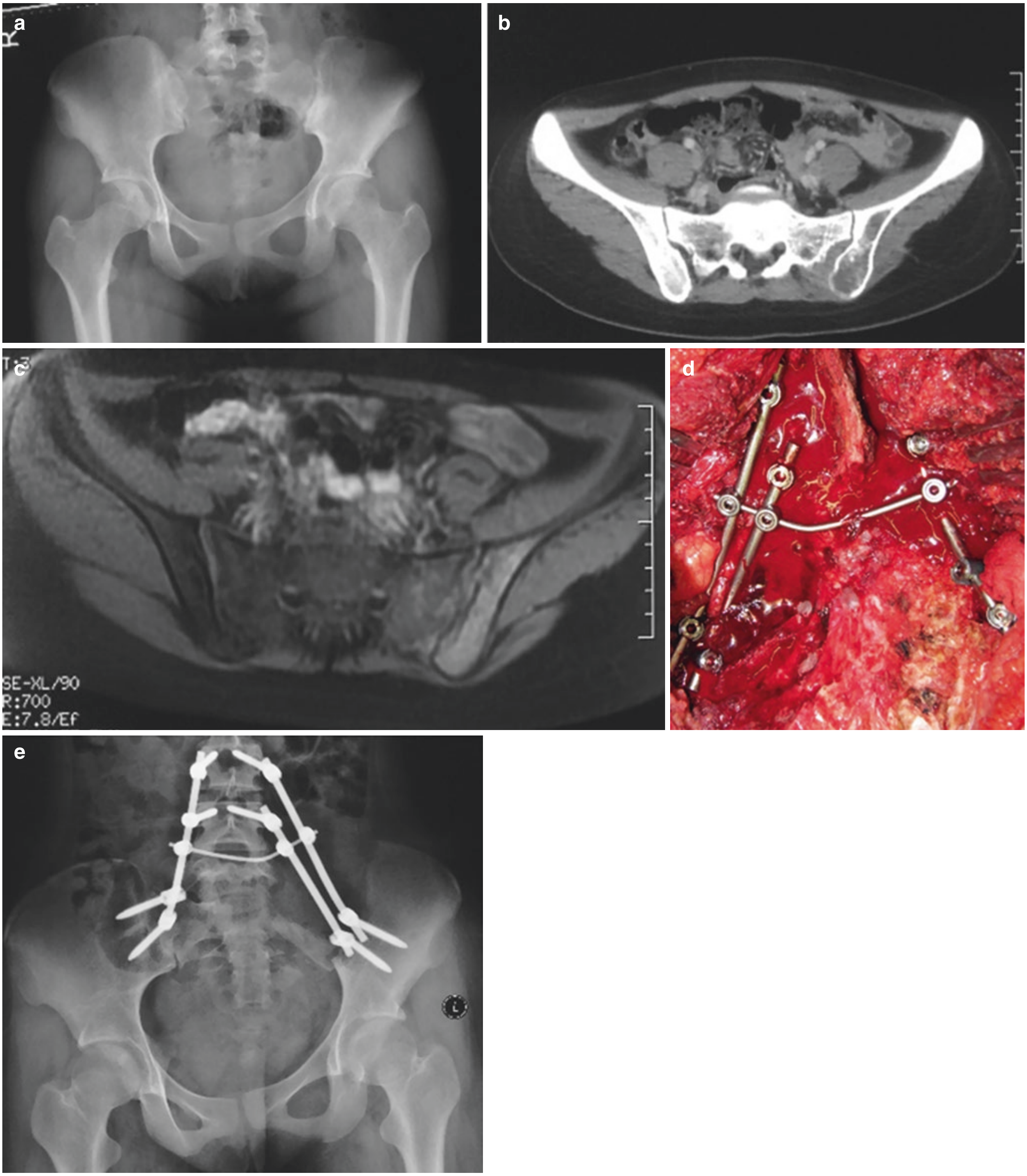

Fig. 14.8 A 15-year-old female was diagnosed with Ewing's sarcoma (**a**–**c**). After subtotal sacrectomy and partial iliac resection, bilateral spino-pelvic reconstruction using pedicle-iliac screw and rod was performed (**d**, **e**). A PKUPH case

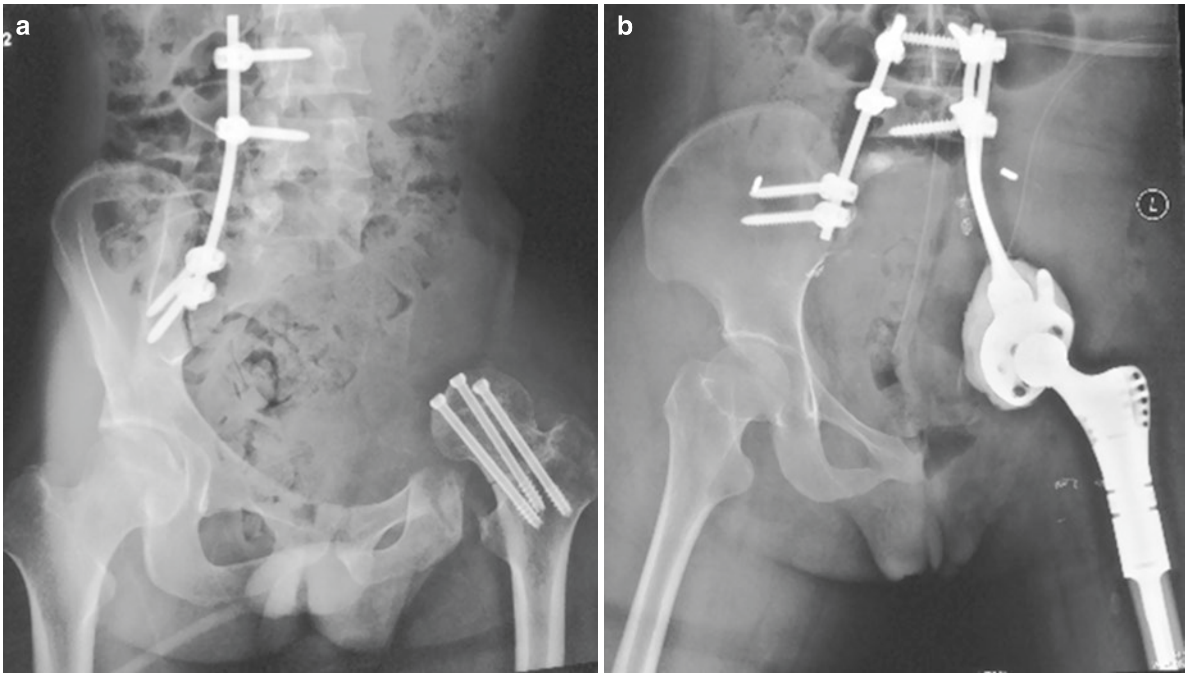

Fig. 14.9 (**a**) A 24-year-old male was diagnosed with chondrosarcoma. After total sacrectomy and internal hemipelvectomy, contralateral spinopelvic reconstruction using pedicle-iliac screw and rod was performed, leaving a spared left lower limb. (**b**) A 24-year-old female was diagnosed with osteosarcoma. After total sacrectomy and internal hemipelvectomy of the involved proximal femur, bilateral spinopelvic reconstruction using pedicle-iliac screw and rod connecting to a hemipelvic prosthesis articulated with a proximal femoral prosthesis was performed. Two PKUPH cases

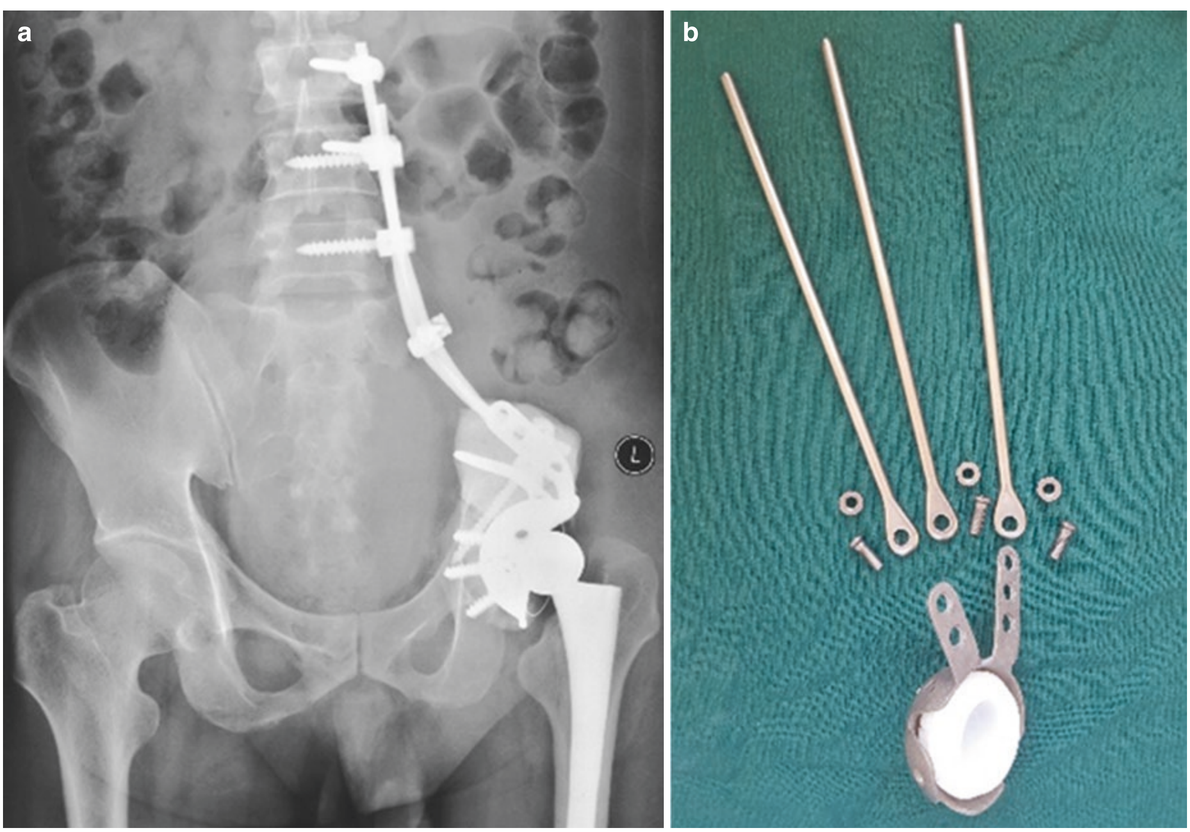

Fig. 14.10 A 43-year-old male was diagnosed with chondrosarcoma. After type I + II + IV pelvic resection, endoprosthetic reconstruction using a caudal-flanged hip cage connected to pedicle screw and rod system was performed with enhancement of periprosthetic bone cement. A PKUPH case

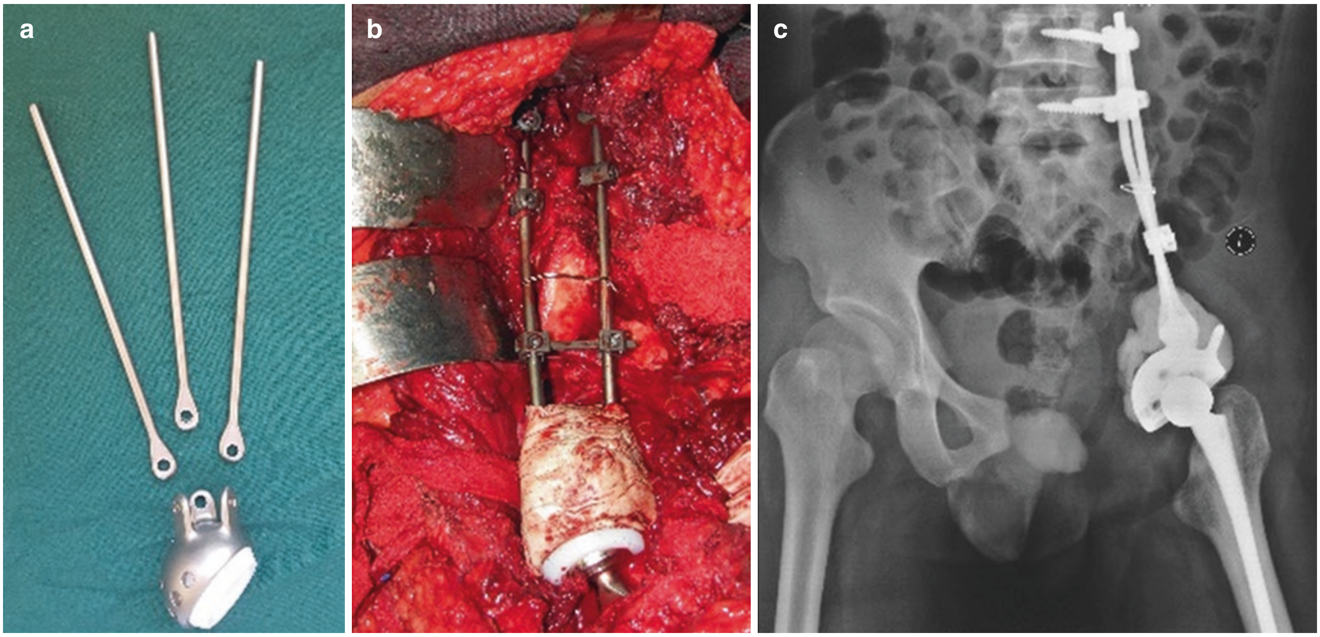

Fig. 14.11 A 33-year-old male was diagnosed as malignant inflammatory myofibroblastoma. After type I + II + III + IV pelvic resection, endoprosthetic reconstruction using a modular hemipelvic prosthesis connected to pedicle screw and rod system was performed with enhancement of periprosthetic bone cement. A PKUPH case

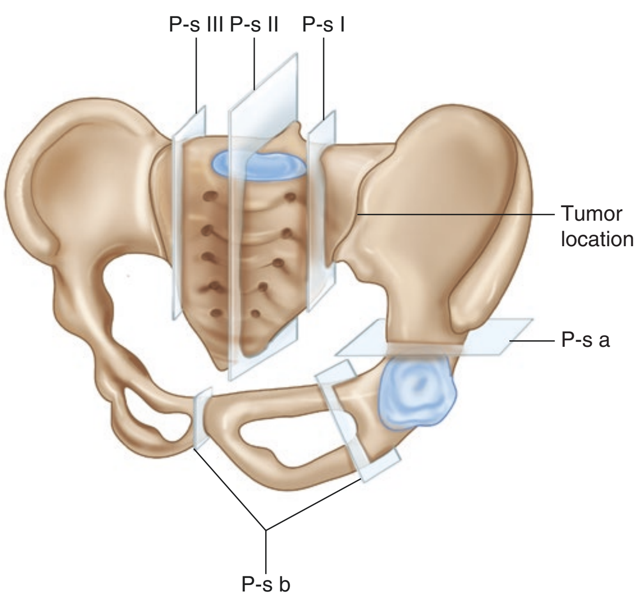

Fig. 14.12 Illustration of the new surgical classification for pelvic tumors with sacral invasion

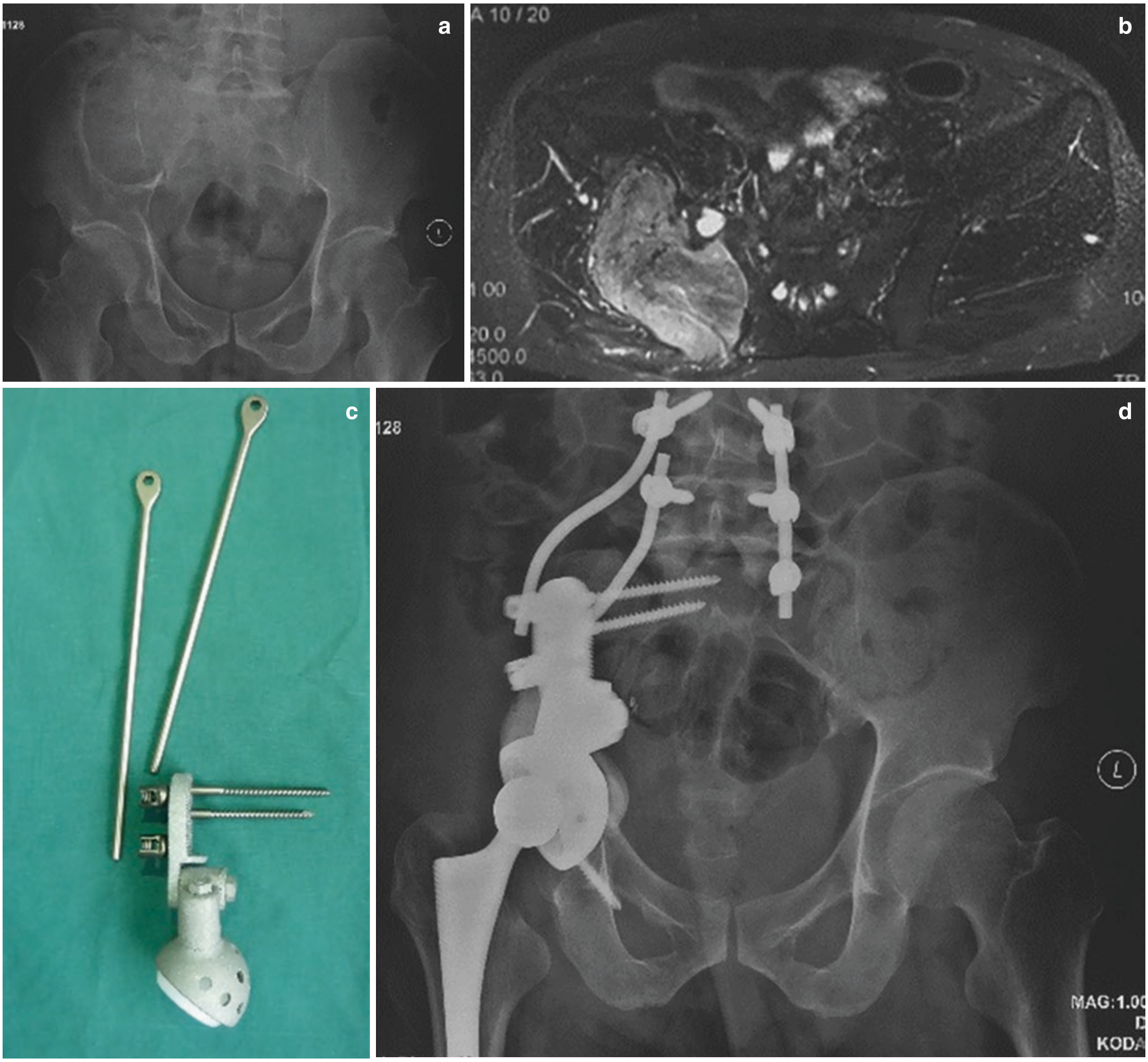

Fig. 14.13 A 35-year-old male was diagnosed with malignant peripheral neurofibroma (**a**, **b**). After type I + II + IV pelvic resection, endoprosthetic reconstruction using a newly designed modular hemipelvic prosthesis connected to pedicle screw and rod system was performed with enhancement of periprosthetic bone cement (**c**, **d**). A PKUPH case

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Part IV

Pelvic Tumors: Special Topics

15 Reconstruction with 3D-Printed Endoprosthesis

Wei Guo

15.1 3D Printing Technology in Bony Defect Reconstruction

In recent years, 3D printing techniques have been used widely in orthopedic surgery because of the special structure and characteristic of bone.

3D printing technique stays on the early research stage several years ago, but 3D printing techniques in clinical use have been improved quickly in recent years. It almost covers most of the orthopedic field including bone tumor resection and functional reconstruction [1], arthroplasty revision [2], spinal deformity correction [3], complex bone fracture treatment [4], and using 3D-printed bioceramics for antibiotic delivery to treat implant-associated bone infection [5], etc.

In the field of basic research, the maturity and reliability of 3D techniques have been further proved. Such differences and consistency of materials between 3D printing and traditional manufacturing have been evaluated [6], and a lot of studies on the surface treatment using 3D technology have been reported in the literatures [7–9]. Also, 3D printing technology being used in tissue engineering and bone regeneration has been widely evaluated [10, 11].

15.1.1 Reconstruction of Weight-Bearing Bone by 3D-Printed Prosthesis

In recent years, the most advance of 3D printing technology, also called additive manufacturing, is rapidly producing prosthesis. Weight-bearing bones are different from bones of upper limb; they need to bear weight at physiological status, so that the 3D-printed prosthesis has to be of strength in some degree. In addition, reconstruction of weight-bearing bony defect needs not only filling the defect but also long-term fixation. It could be ideal if the 3D printing prosthesis is of bone ingrowth characteristic. In general, titanium alloy 3D printing techniques by EBM or SLM could meet these demands.

Up to now, Guo [1] reported the largest series of using 3D prosthesis reconstruction of bony defect after pelvic tumor resection. The series including 35 patients received tumor resection and functional reconstruction with 3D-printed pelvic endoprostheses from September 2013 to December 2015. The methods of reconstruction included 3 patients with 3D-printed iliac endoprosthesis (Fig. 15.1), 12 patients with 3D-printed standard hemipelvic endoprosthesis (Fig. 15.2), and 20 patients with 3D-printed screw-rod-connected hemipelvic endoprosthesis (Fig. 15.3). The mean follow-up was 20.5 month. The average MSTS93 score was 19.1 (63.6%) in patients with 3D-printed hemipelvic endoprosthesis.

There was no deep infection, prosthesis loosening, or breakage found in the series, and also no osteolysis was found at the interface of the prosthesis. It proved that 3D prosthesis can meet the requirements of using for reconstruction of weight-bearing bony defect in mechanical strength, biocompatibility, and bone ingrowth aspects.

There were some case reports about using 3D printing technology in reconstruction of pelvic bony defect in the literatures. Wyatt [2] reported one hip revision case of using EBM 3D printing technique that repaired acetabular defect. Based on the patient's pelvic CT data, the engineer design a prosthesis for reconstruction of the pelvic bony defect, and then the doctor installed and fixed the prosthesis successfully for the patient (Fig. 15.4). Zoccali et al. [12] reported one case of using of 3D-printed prosthesis for reconstruction of the hip joint after pelvic tumor resection. The doctor designed the osteotomy level and made a shape-like prosthesis by 3D printing technique. Wong et al. [13] not only designed a 3D-printed pelvic prosthesis but also printed a guide plate for precise resection of the pelvic bone for a tumor patient, achieving the precise resection and precise installation during the surgery (Fig. 15.5). Arabnejad et al. [14] designed and made a titanium alloy femoral stem using 3D printing technique for total hip arthroplasty. Compared with

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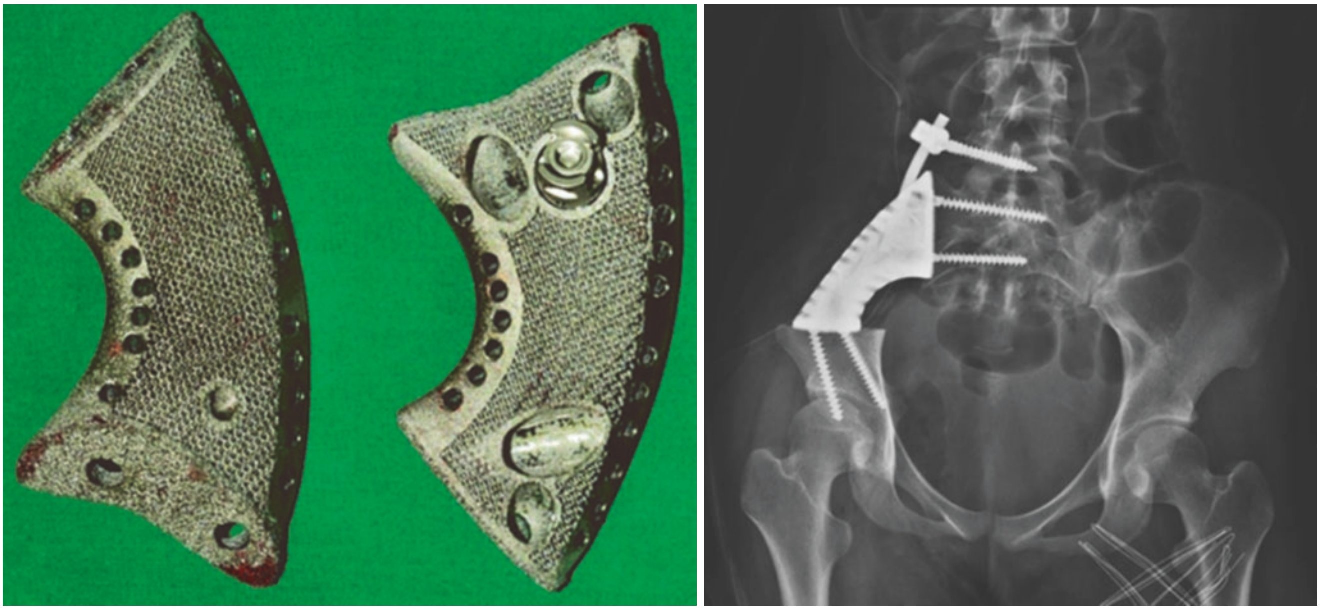

Fig. 15.1 3D-printed iliac prosthesis

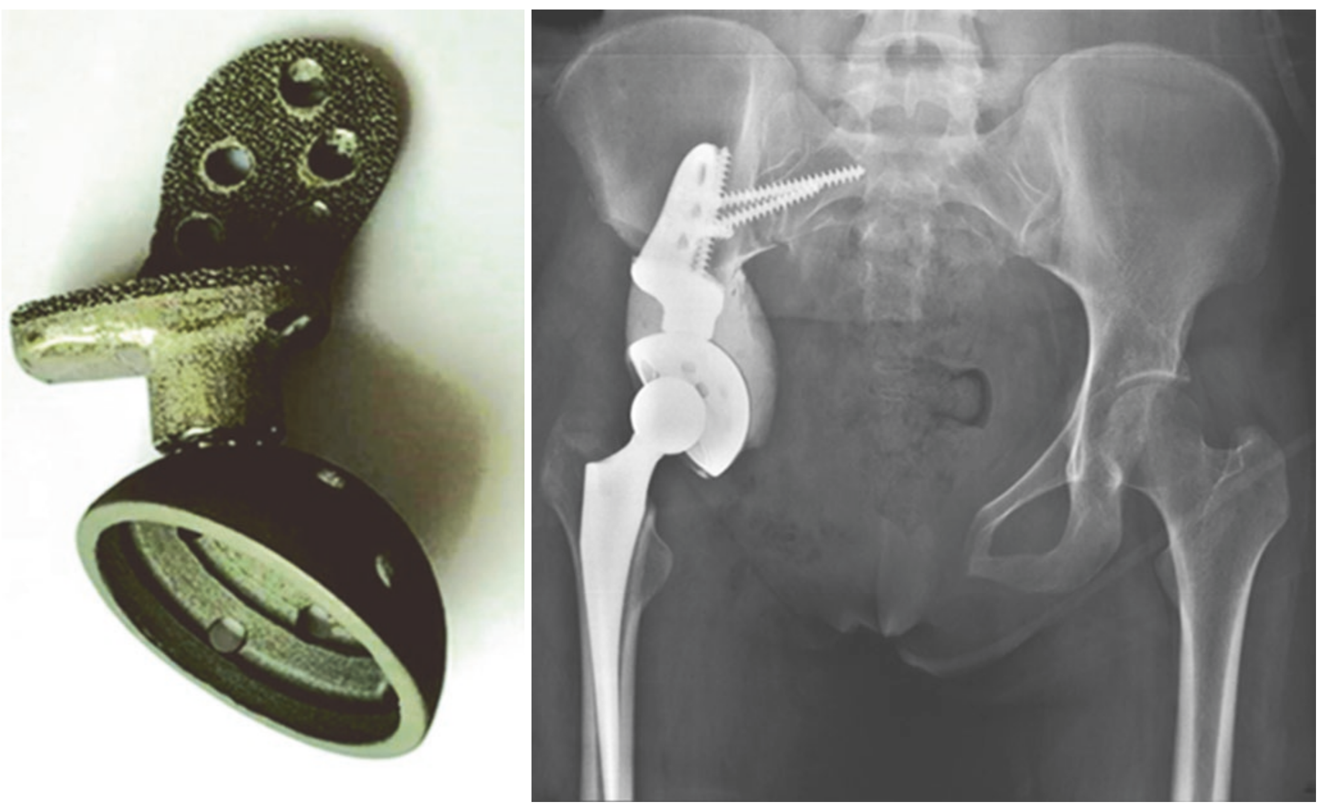

Fig. 15.2 3D-printed standard pelvic prosthesis

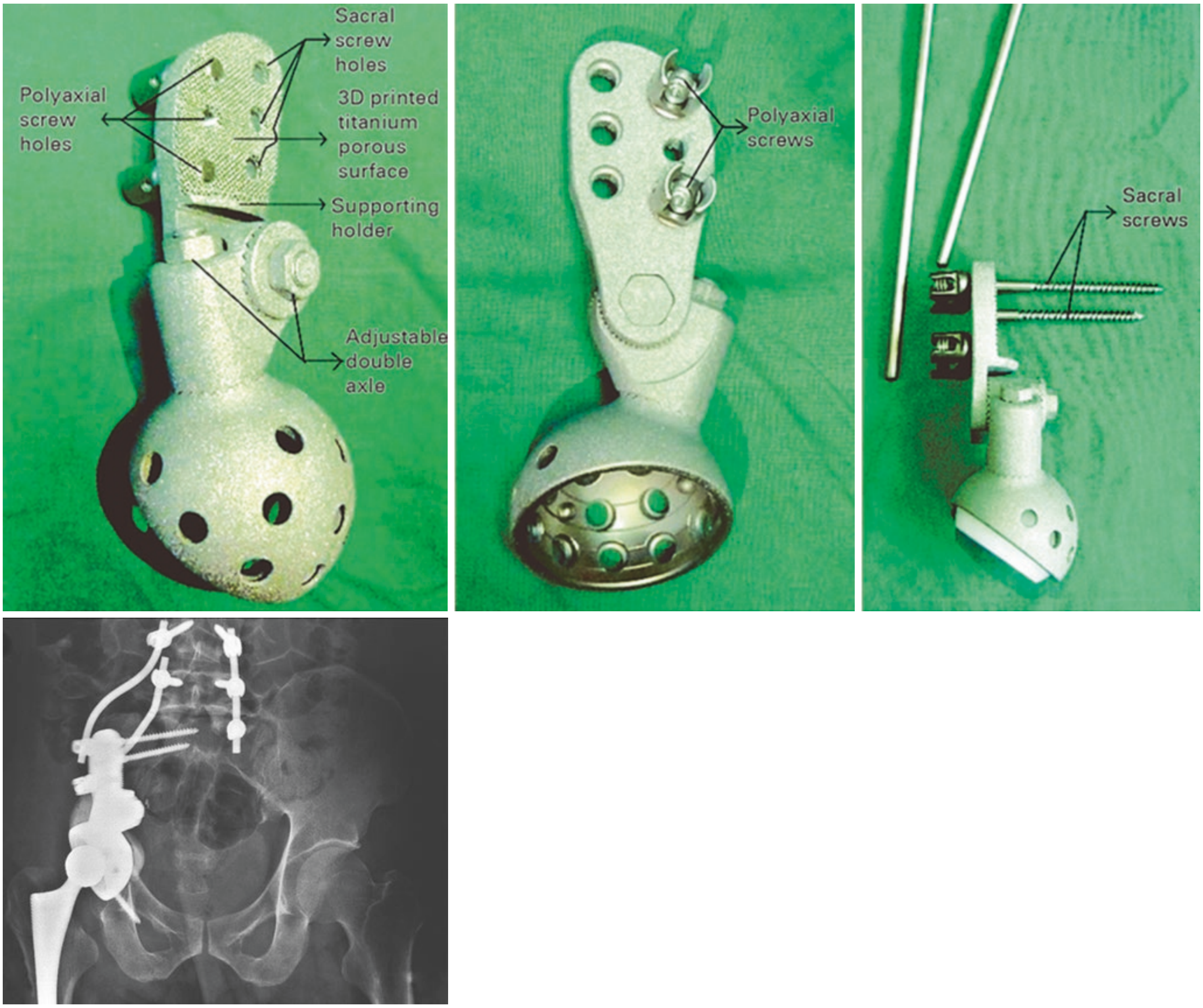

Fig. 15.3 3D-printed rod-and-screw-connected pelvic prosthesis

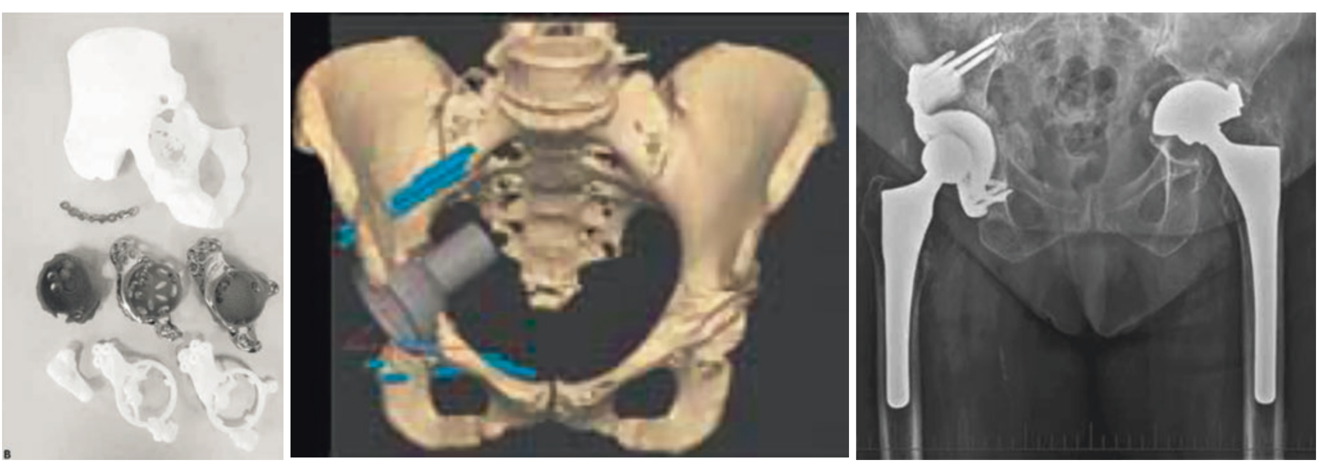

Fig. 15.4 3D-printed reinforced acetabular cup for hip revision surgery

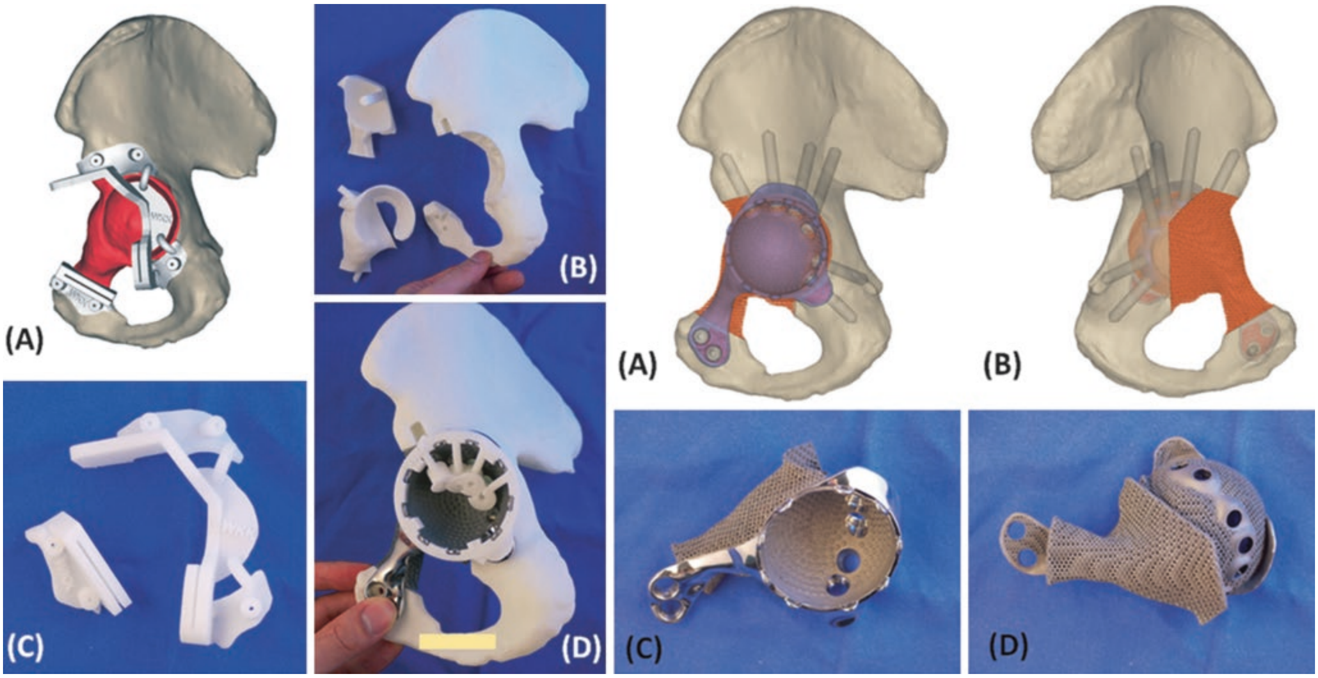

Fig. 15.5 3D-printed guide plate and prosthesis for pelvic tumor surgery

traditional manufacturing, this 3D-printed stem is of special porous structure which could reduce the stress shielding obviously to the femur.

15.1.2 Reconstruction of Non-weight-Bearing Bony Defect by 3D-Printed Prosthesis

3D-printed prosthesis also can be used in non-weight-bearing bony defect reconstruction because of the advantage of biocompatibility and soft tissue ingrowth characteristic of 3D-printed prosthesis. Fan et al. [15] reported one case of clavicle reconstruction using a 3D-printed prosthesis after Ewing sarcoma resection (Fig. 15.6). Based on three-dimensional imaging technique, the engineer designed and manufactured a total clavicle prosthesis by 3D printing technology. The two ends of the prosthesis were of several holes for fixation with sternum and scapular. The surface of the prosthesis was of porous structure which could be attached or grown by soft tissue. They also reported one case of scapular prosthetic reconstruction by 3D printing technology (Fig. 15.7).

15.1.3 3D-Printed Implants in the Spinal Surgery

In terms of the spinal implant, Xu et al. [16] reported one case of using 3D-printed implant for reconstruction of spinal stability after resection of C2 Ewing sarcoma. The doctor designed a 3D-printed implant which can be fixed to the C1 lateral mass by screws because regular titanium mesh could be fixed into the lateral mass of C1 (Fig. 15.8). de Beer and Scheffer [17] used rapidly manufactured patient-specific intervertebral disc implants for reducing subsidence risk. Ran Wei and Wei Guo reported using 3D-printed custom-made sacral prosthesis for reconstruction after total en bloc sacrectomy. The prosthesis provided an optimal reconstruction of lumbosacral and pelvic ring by integrating spinal pelvic fixation, posterior pelvic ring fixation, and anterior spinal column fixation in one step, and its porous surface could induce bone ingrowth and might enhance stability [18] (Fig. 15.9).

15.2 Reconstruction with 3D-Printed Pelvic Endoprostheses After Pelvic Tumor Resection

With the application of adjuvant chemotherapy, advancements of radiological studies, and especially improvements of surgical techniques during the past decades, limb-salvage surgeries for pelvic tumors have been justified worldwide in terms of safety and efficacy [19–21]. There are quite a few reconstruction methods for bone defect after tumor removal, but controversy remains on which method is optimal for selected cases [19–22]. Biological reconstruction has been noted for potential of permanent bone union and avoidance of prosthetic revisions during lifelong follow-up [23–27], while

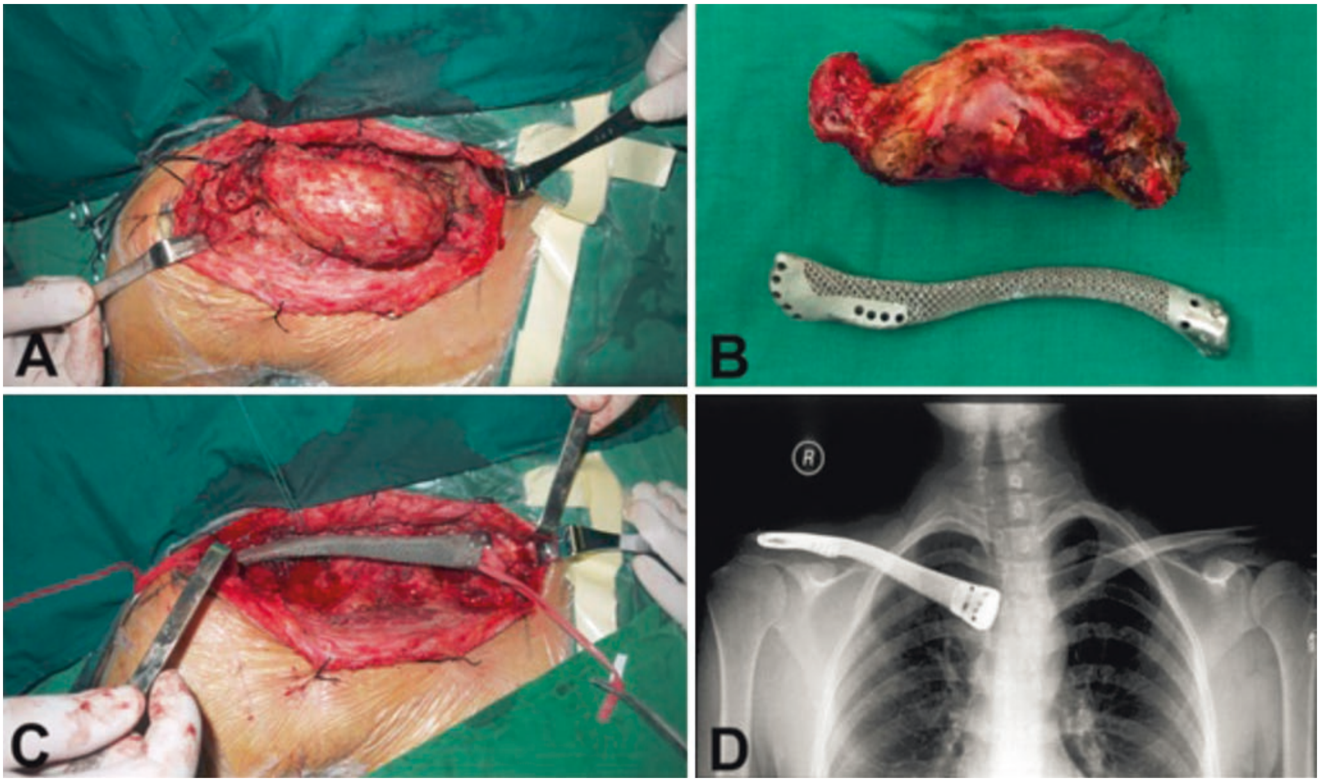

Fig. 15.6 3D-printed clavicle prosthetic reconstruction after tumor resection

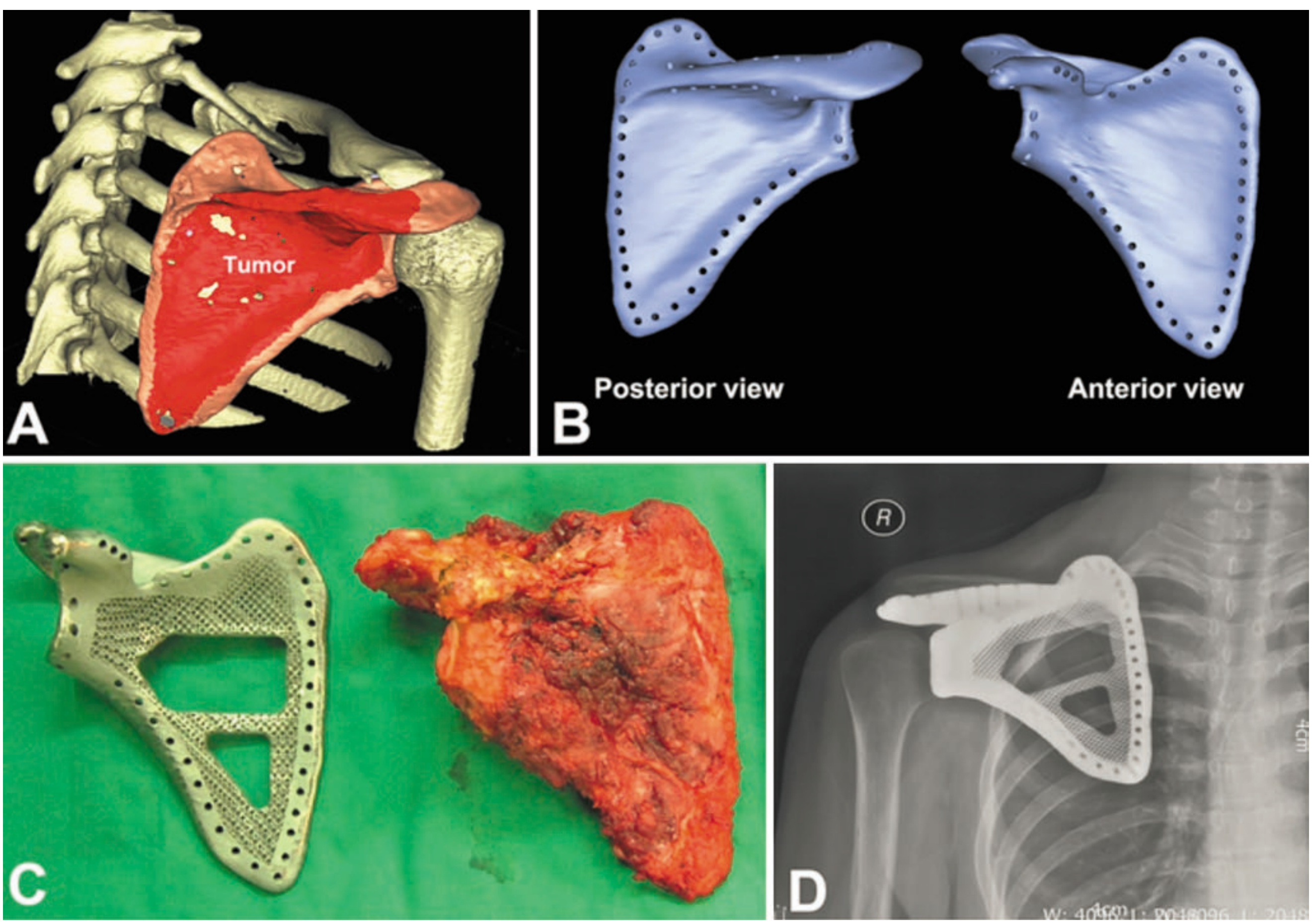

Fig. 15.7 3D-printed scapular prosthesis

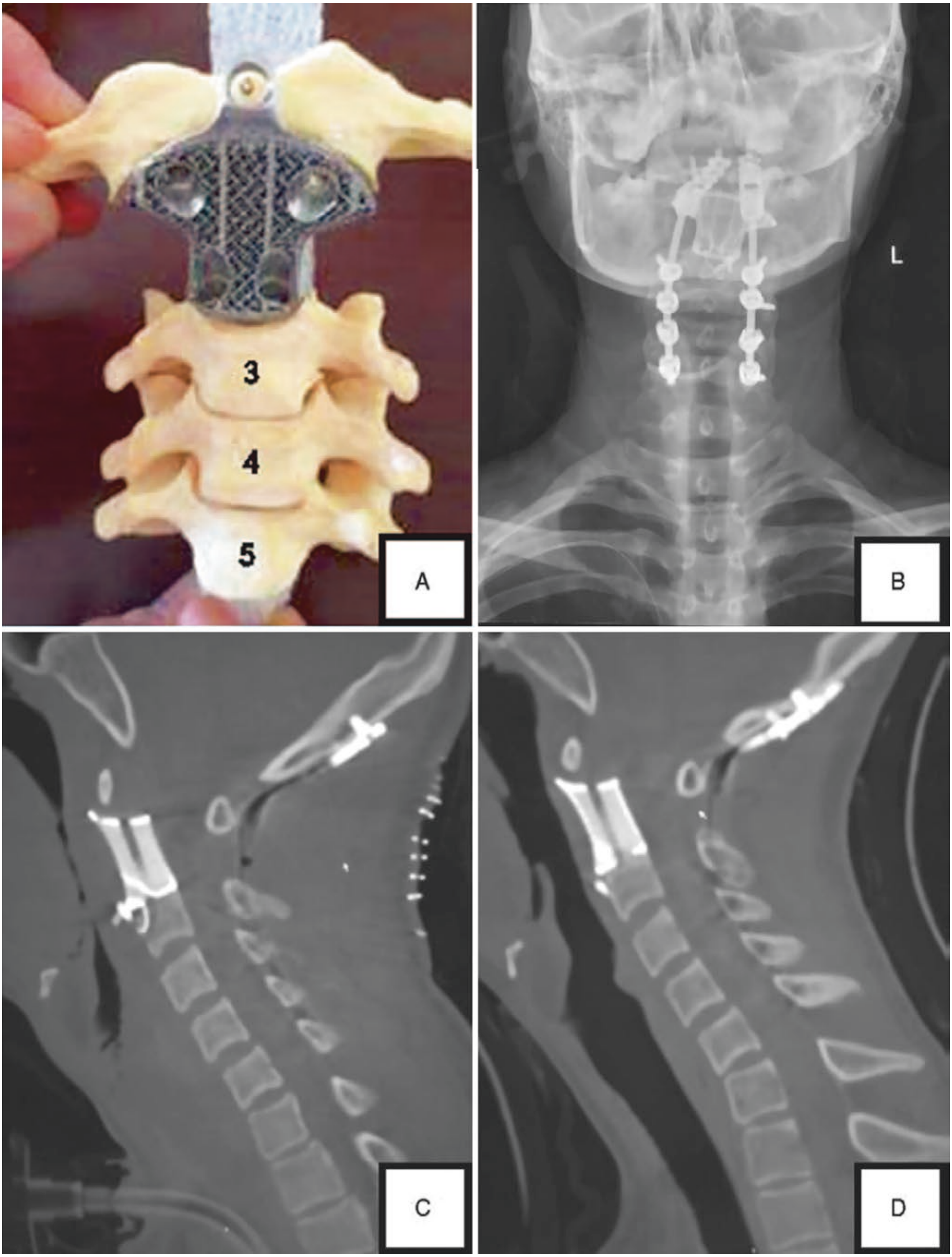

Fig. 15.8 3D-printed C2 vertebral prosthesis

Fig. 15.9 3D-printed custom-made prosthesis for reconstruction after total en bloc sacrectomy. (**a**) Dorsal view. (**b**) Lateral view. (**c**) Superior view

endoprosthetic reconstruction has the advantages of early ambulation, fine cosmetic appearance, prolonged stability, and satisfactory functional status [21, 22, 28–30]. The advent of 3D printing technology, also called additive manufacturing or rapid prototyping, makes it possible for integrating the advantages of both biological and endoprosthetic reconstruction into one implant. Theoretically, it could produce endoprostheses in any shape with porous surface structure that facilitates precise matching, stable fixation, restoration of normal mechanical loading, and osseointegration between the implant and host bone [8, 31–33]. To assess the feasibility of this technology in manufacturing endoprostheses for pelvic reconstruction, we performed a retrospective study of 35 patients who received pelvic tumor resection and reconstruction using 3D-printed pelvic endoprostheses.

15.2.1 Surgical Classification for Pelvic Tumor

Campanacci divided pelvic tumor into three surgical resection types and divided into four subtypes for each type based on the excision extension:

- Type A: Ilium resection; apply to ilium and surrounding soft tissue tumors.
  - Subtype A0: Wedge-shaped ilium resection. Iliac ring is not break off.
  - Subtype A1: Excision extension from superior border of acetabulum to the ilium side of sacroiliac joint.
  - Subtype A2: Excision extension from acetabulum to whole sacroiliac joint.
  - Subtype A3: Excision extension from acetabulum to half sacrum sagittally.

- Type B: Acetabulum resection; apply to tumors invading acetabulum and around soft tissue.
  - Subtype B0: Partial acetabulum resection.
  - Subtype B1: Total acetabulum resection (including intra-articular or extra-articular resection).
  - Subtype B2: Acetabulum and anterior arch excision.
  - Subtype B3: Extra-articular resection of acetabulum and proximal femur.
- Type C: Anterior arch excision (pubis and ischium resection).
  - Subtype C0: Single pubis or ischium resection.
  - Subtype C1: Single anterior arch excision.
  - Subtype C2: Single anterior arch and partial acetabulum excision.
  - Subtype C3: Both side of anterior arch resection.

Design excision extension should be according to MRI examination for malignant pelvic tumors. Osteotomy line should be 2–3 cm outside of the tumor. In order to achieve wide resection, above excision type can often be used in combination.

Enneking divided pelvic tumor into four surgical resection types based on the tumor location (Fig. 15.10):

- Type I: Ilium resection
- Type IA: Ilium combining with gluteus resection
- Type II: Acetabulum combining with femoral head and articular resection, proximal femur fusion with ilium
- Type IIA: The whole ilium and acetabulum resection, proximal femur fusion with ischium
- Type III: Obturator foramen resection
- Type IV: Iliac bone combining with partial sacrum resection

If the tumor is involving the region of several types, resection extension should be combination of several types, such as type I II, I IV, II III, I II III.

Although Enneking classification of pelvic tumors has been useful for the resection of tumors located in the innominate bone, it gives little information concerning the sacral involvement. Moreover, little is known with regard to the functional reconstruction from the Enneking classification, especially in this area of limb salvage. Therefore, based on the tumor location and the required osteotomy site, we proposed a novel categorization system for the pelvic tumors with sacral invasion. The standardized surgical procedures as well as recommended reconstructive methods are introduced for each type of resections.

The newly introduced system (P-s classification) was devised based on the sagittal extent of sacral invasion rather than the transverse level of sacral invasion of the tumor. Type P-s (abbreviation for "pelvic tumor with sacral invasion") I resection refers to osteotomy through the ipsilateral sacral wing when the tumor only invades the adjacent sacroiliac joint. Type P-s II resection refers to osteotomy through the sacral midline when the tumor invades the ipsilateral sacral foramina. Type P-s III resection refers to osteotomy through contralateral sacral wing when the tumor invades the contralateral sacral foramina. And type P-s IV resection refers to osteotomy through the contralateral iliac wing when the tumor invades the contralateral sacroiliac joint. Meanwhile, the extent of invasion to the innominate bone is categorized as type "a" or type "b" when they are confined to the iliac wing or invade the periacetabular region, respectively (Fig. 15.11).

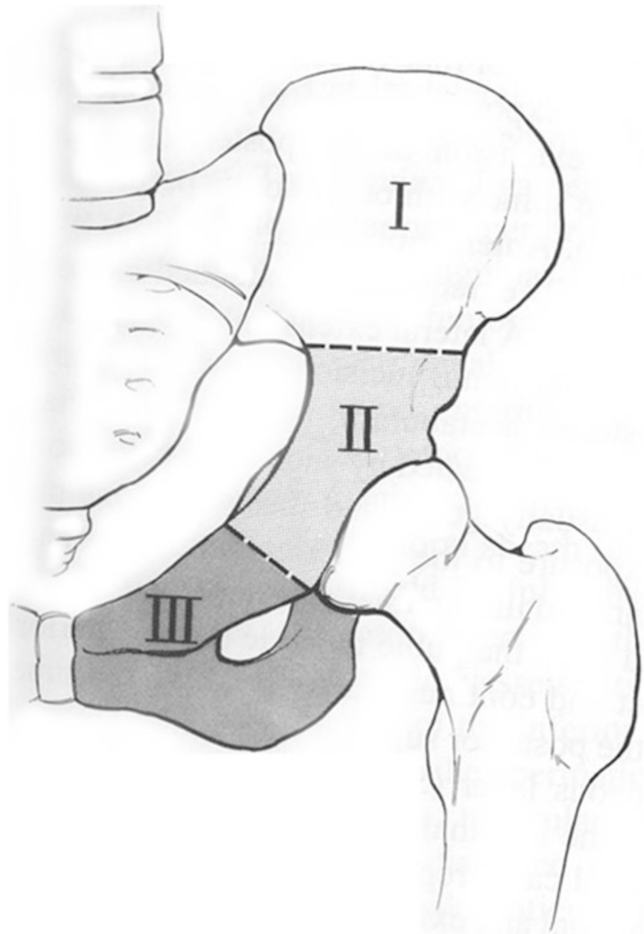

Fig. 15.10 Enneking surgical classification for pelvic tumor

15.2.2 Principle and Design of the 3D-Printed Pelvic Endoprostheses

Abundant clinical experience of application of modular hemipelvic endoprosthesis in hundreds of cases brings up ideas of modifications to improve implant survival [21]. All the three kinds of 3D-printed pelvic endoprostheses in this study were produced from titanium alloy by electronic beam

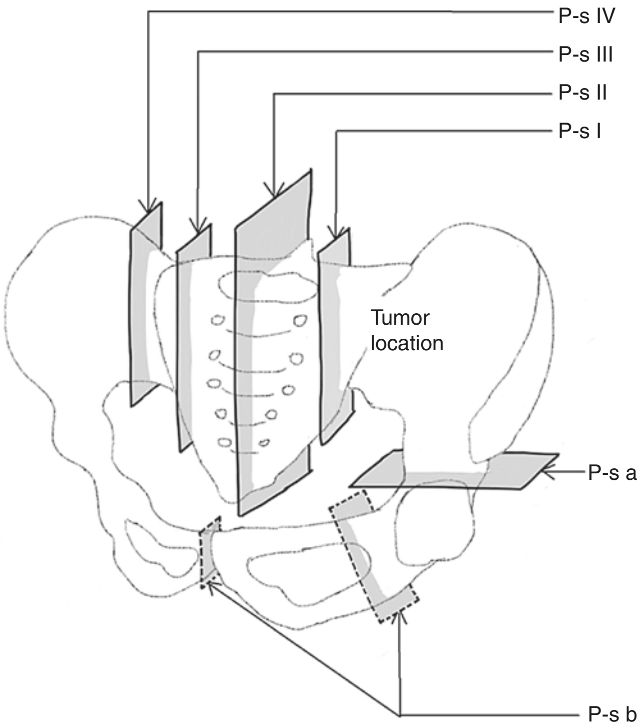

Fig. 15.11 Illustration of the osteotomy sites for different type of resections according to the new categorization system of sacropelvic tumors

melting (EBM) technology. The fundamental characteristic was the porous structure on the implant-bone interface that had osseointegration potential as proven by in vitro and in vivo studies [8]. In order to provide more flexibility during tumor resection and reconstruction, the endoprostheses were prepared in a modular way that each component had three sizes.

3D-printed iliac endoprosthesis (Aikangyicheng Co., Beijing, China) (Fig. 15.1): It was designed for type I and type I + IV resection that seated between the sacrum and roof of acetabulum. Its shape simulated the inner part of ilium with two reserved screw holes for fixation to the sacral and acetabular sides. It also had a polyaxial screw at the backside for connection to the lumbar pedicle screw. There were several reserved holes along the outer and inner edges of the endoprosthesis for soft tissue attachment.

3D-printed standard hemipelvic endoprosthesis (Aikangyicheng Co., Beijing, China) (Fig. 15.2): It was used for reconstruction after type II/II + III resection. It evolved from the previous reported modular hemipelvic endoprosthesis [21, 22] with modifications on the contour of iliac foundation, direction of the reserved screw holes, and structure of the implant-bone interface.

3D-printed screw-rod-connected hemipelvic endoprosthesis (Chunli Co., Beijing, China) (Fig. 15.3): It was used for reconstruction after resection of both acetabulum and iliosacral junction. This new endoprosthesis had great improvements compared with the previous reported screw-rod-connected hemipelvic endoprosthesis [34, 35]. It composed of a sacral holder and an acetabular ring that were linked by a double-axle mechanism. The vertical part of the sacral holder had three reserved holes for sacral fixation and two polyaxial screws for connection to the lumbar pedicle screws. The inner surface of the sacral holder was of porous structure, and the angle of the acetabular ring was adjustable.

15.2.3 Methods of Tumor Resection and Functional Reconstruction

1. Type I resection and reconstruction
   For cases of iliac tumors with/without involvement of sacral ala, operation could be done in a single lateral decubitus on the contralateral side. An extended ilioinguinal approach was used followed by dissection of the retroperitoneal space and mobilization of iliac vessels and femoral nerve. Then the glutei were dissected to expose the sciatic notch and sciatic nerve. Two Gigli saws were introduced through the greater sciatic foramen to make osteotomy through the ilium and sacral ala or sacroiliac junction, after which the specimen could be removed.

   A 3D-printed iliac endoprosthesis of suitable size was selected according to intraoperative measurement of bone defect. To fix the prosthesis, two cancellous screws were introduced through the reserved holes in the upper prosthesis into the S1/S2 vertebral body. Then another two cancellous screws were placed into the pubis and ischium, respectively. One or two pedicle screws were then introduced through the ipsilateral pedicles of L5 and/or L4. Finally, the fixation of the endoprosthesis was reinforced posteriorly by connecting to the pedicle screws with a titanium rod (Fig. 15.1).
2. Type II + III resection and reconstruction
   Resection of the acetabulum and obturator foramen was done as previously reported [21, 22]. After tumor removal, a 3D-printed modular hemipelvic endoprosthesis of suitable size was selected to fit for the contour of residual ilium. Before fixation, the angle of the acetabular ring could be adjusted. Then with the assistance of fluoroscopy, two long cancellous screws were introduced through the reserved screw holes of the endoprosthesis, across the iliosacral junction, strutting into S1/S2 vertebral body. Another two or three cortical screws would be used to strengthen the fixation between the endoprosthesis and residual ilium. Finally, high-viscosity bone cement with gentamicin was used to augment the endoprosthesis, and the hip joint was restored in similar way of conventional total hip replacement (THR) (Fig. 15.2).

3. Type I + II/I + II + III/I + II + IV/I + II + III + IV resection and reconstruction
   En bloc resection of the acetabulum, ilium, and/or obturator foramen, and/or partial sacral ala, was done as previously reported [21, 34]. After tumor removal, a structural graft was harvested from the intact femoral head and fixed to the residual sacrum. At times, the endoprosthesis could be fixed to the residual iliosacral junction without an autograft. Then the 3D-printed screw-rod-connected hemipelvic endoprosthesis was fixed to the graft using two long cancellous screws, strutting into the S1/S2 vertebral body. Afterward, two pedicle screws were placed in the L4 and L5 pedicles and connected to the reserved polyaxial screws of the endoprosthesis with two titanium rods (Fig. 15.3). The abduction angle and anteversion angle of the acetabulum could be adjusted by the two gear mechanisms. Finally, high-viscosity bone cement with gentamicin was used to augment the endoprosthesis, and the hip joint was restored in similar way of conventional THR.

15.2.4 Postoperative Functional Status

There were few cases reported in literature except our study using 3D-printed prosthesis for pelvic reconstruction [1]. The mean MSTS-93 score of the 30 survival patients was 19.1 (9–26). Three patients with iliac endoprosthesis reconstruction had a mean MSTS-93 score of 22.7 (20–25). Those with type II + III resection and standard hemipelvic endoprosthesis reconstruction had a mean score of 19.8 (15–26), while those with screw-rod-connected hemipelvic endoprosthesis reconstruction had a mean score of 17.7 (9–25) [1].

15.2.5 Complications

There were no cases of deep infection observed during follow-up except some cases of delayed wound healing that required debridement and few cases of hip dislocation. X-ray examinations were undertaken every 3 months postoperatively and showed no evidence of bone absorption or osteolysis at the prosthesis-bone interface. No bone absorption, prosthetic loosening, breakage, or displacement were found in radiological studies. In all of the 18 cases reconstructed with femoral head autograft, bony fusion was observed between the graft and sacrum [1].

15.3 Functional Reconstruction After Pelvic Tumor Resection: Techniques

With the improvements of surgical techniques and adjuvant therapies, the oncological outcomes of pelvic tumors have been improved to some extent that demands for more durable reconstruction [21, 36–38]. Although various reconstruction methods for internal pelvectomy have been proposed in literatures during the past decades [19, 21–27, 34, 35], improvements on implant/graft survival, complications, and functional status are still necessitated. The application of 3D printing technology in orthopedic implants provides greater freedom of endoprosthetic design and allows fabrication of more complex geometries including porous surface with determined pore sizes, which helps to increase the efficiency of intraoperative reconstruction, improve immediate and long-term stability, reduce operative complications, and facilitate osseointegration between prosthesis and host bone [8, 13, 14, 31–33]. In this study, we reported three novel 3D-printed endoprostheses for different types of bone defects after tumor resection, which showed satisfactory outcomes in terms of reconstruction convenience, perioperative safety, short-term implant survival, and functional status.

15.3.1 Reconstruction for Type I/I + IV Resection

Resection of tumors located at ilium, sacrum, or sacroiliac joint would cause discontinuity of the pelvic ring and disruption of loading transfer. Although it had been proposed that reconstruction after type I/I + IV was not necessary [39], an unconstructed sacroiliac defect could cause introversion and upper shifting of the acetabulum, discrepancy of lower limbs, and secondary scoliosis after weight-bearing [25, 39, 40]. Iliosacral arthrodesis with autografts or allografts has been attempted but showed just fair functional status (MSTS-93: 51–57%) [25, 26, 39] with significant shortcomings of prolonged immobilization, nonunion, infection, fracture, and secondary scoliosis. As a result, instrumental reconstruction, mainly sacroiliac screw-rod fixation, was adopted in addition to bone grafts implantation and showed permission for early ambulation and improved functional scores (MSTS-93: 57–75.4%) [25, 26, 40–42]. The combined biological and instrumental reconstruction methods, however, would increase operation time and could be still at risk of infection, nonunion, aseptic loosening, and breakage during long-term follow-up.

We used to reconstruct with a pedicle screw-rod fixation method to obtain immediate stability [35, 41], but long-term complications as above urged for more stable and durable reconstruction methods. Therefore, we designed the novel iliac endoprosthesis fabricated by 3D printing technology. With the reserved screw holes, the endoprosthesis could achieve one-step reconstruction of the iliac defect and simulate normal-loading transfer pattern through the posterior pelvic ring (Fig. 15.1). A posterior pedicle screw-rod fixation would add stability to the spinopelvic continuity, and the holes in the edges of the endoprosthesis could help reattach abductive muscles. The reconstruction procedures were simple and convenient, with mean operation time of 200 min and mean intraoperative hemorrhage of 1200 ml, which was quite comparable to those in literatures [26, 42]. Moreover, the porous surface of the prosthesis-bone interface increased the potential of bone ingrowth and long-term stability. Short-term observation showed excellent functional status (mean MSTS-93 score: 22.7; 75.6%) and no evidence of mechanical failures. One patient could walk by herself without evident Trendelenburg gait at 6-month follow-up, started swimming at 9-month follow-up, and kept doing well at final follow-up.

15.3.2 Reconstruction for Type II/II + III Resection

The highlight of reconstruction for type II/II + III resection has always been restoration of a functional hip with adequate stability. Although biological reconstruction including iliofemoral arthrodesis, ischiofemoral arthrodesis, pseudarthrosis, hip transposition, and flail hip had been reported to yield good functional score (MSTS-93: 58–62%), drawbacks such as prolonged immobilization, discrepancy of lower limbs, and limited hip joint movement warranted more meticulous reconstruction [23, 24, 27, 43]. Reimplantation of devitalized autografts with total hip replacement had also showed excellent functional outcome (MSTS-93: 75%) but could not be used in case of massive osteolytic destruction [44]. As a result, endoprosthetic reconstruction is a feasible choice after type II/II + III resection.

Saddle prosthesis was one of the earliest attempts in endoprosthetic reconstruction after periacetabular tumor resection but showed disappointed outcomes in terms of high incidence of complications (dislocation: 2–22%; departure of components: 0–12%; upshifting: 25%; iliac fracture: 30%; ectopic ossification: 35%) [45–48]. The failure of saddle prosthesis indicated that a total hip replacement plus a secure anchorage of the acetabular ring should be the fundamental principle for hemipelvic endoprosthetic design. Since then, various kinds of hemipelvic endoprostheses have been reported in literatures, and they generally differed from each other on the anchorage mechanism [21, 49–53].

Custom-made hemipelvic endoprostheses had been reported to have good function but compromised implant survival for pelvic reconstruction. Windhager et al. [54] once reported 21 cases of periacetabular sarcomas with different kinds of reconstruction in 1996. Nine cases underwent custom-made prosthesis replacement and yielded better function than saddle prosthesis and allograft implantation. Ozaki et al. [29] reported 12 cases of custom-made hemipelvic endoprosthesis in 2002, all of which were designed with computer-assisting technology based on preoperative imaging results. After a mean follow-up of 57 months, survival of the prosthesis was 42%, and the major complication was deep infection. Witte et al. reported the application of a MUTARS® custom-made hemipelvic endoprosthesis in 2009. The endoprosthesis achieved a mean MSTS score of 50% but with a high complication rate as 75% [50].

The pedestal cup prosthesis, also called ice-cream cone prosthesis due to its shape, showed disappointed outcomes in early stage of clinical application [51, 52]. Bus et al. reported the application of a pedestal cup in 19 patients with periacetabular malignancies with 3 cases of recurrent dislocation, 3 of aseptic loosening, 9 of infection, and 2 of local recurrence, which resulted in overall implant survival at 5 years of 50% and the mean MSTS score of 49% [52]. The Royal Orthopaedic Hospital in Birmingham also reported the outcome of a similar prosthesis in 27 cases treated from 2004 to 2009 with overall complication rate of 37% including 14.8% cases with dislocation and 11.1% cases with deep infection [51]. Recently, Bus et al. reported a modified pedestal cup prosthesis, called LUMiC endoprosthesis, for periacetabular reconstruction and showed improved implant survival (5-year failure rate: 17.3% for mechanical reasons, 9.2% for infection) and better functional status (MSTS-93: 70%) [53].

In 2007, our center firstly reported the outcome of a modular hemipelvic endoprosthesis in 28 cases [21]. In 2013, we published the midterm outcome of 100 cases of modular hemipelvic endoprosthesis replacement, which showed that mean MSTS score was 57.2%, the 3-year prosthesis survival was 81.8%, the deep infection rate was 15%, the rate of wound healing problems was 18%, hip dislocation rate was 9%, and the breakage rate was 5% [20]. This modular hemipelvic endoprosthesis had the advantages of convenience for implantation, simulation for normal-loading transfer, and short-term stability for sure [21, 55] but might experience loosening and decreased stability during long-term follow-up.

In this study, we modified the previous modular hemipelvic endoprosthesis with the 3D printing technology. Firstly, the contour of the iliac foundation, as well as the locations and directions of reserved screw holes within it, was designed based on enormous data from pelvic CT scans of previous cases. As a result, perfect fitness between the iliac foundation and outer curved surface of the residual ilium and precise screw paths from the implants through sacroiliac joint into S1/S2 vertebral body were guaranteed (Fig. 15.2). This provided more stable fixation and more similar to normal-loading transfer than the former endoprosthesis. Meanwhile, we utilized long cancellous screws in combination with short cortical screws to improve immediate stability. Moreover, the prosthesis-bone interface of the prosthesis was produced as porous structure with titanium alloy by EBM 3D printing technology, which could help facilitate bone ingrowth and achieve long-term stability. The procedures for installation of this new hemipelvic endoprosthesis were rather simple and convenient that did not cost extra time or additional intraoperative hemorrhage. By short-term follow-up, this

3D-printed standard hemipelvic endoprosthesis achieved a mean MSTS-93 score of 19.8 (15–26, 66%), better than those of the previous endoprostheses.

15.3.3 Reconstruction for Type I + II/I + II + III/I + II + IV/I + II + III + IV Resection

There were limited kinds of endoprostheses suitable for reconstruction after a type I + II resection. The concurrent deficiency of iliac wing, sacroiliac joint, and acetabulum could provide no anchorage for those common endoprostheses as mentioned above. We previously reported the application of modular hemipelvic endoprosthesis with a structural autograft fixed to the residual sacrum after type I + II/I + II + IV resection [20]. The functional result was fair (44.5%), and breakage of the sacral screws was observed after long-term follow-up. Therefore, we invented the first generation of screw-rod-connected hemipelvic endoprosthesis [34], which rebuilt the spinopelvic continuity and retained the function of hip joint. Patients could be fully weight-bearing and could walk with/without assistance. That endoprosthesis, however, still failed to provide a prolonged stabilization of spinopelvic and sacroiliac junction, which would experience loosening and breakage during long-term follow-up. Moreover, it could bring additional risk of neuralgia due to the utilization of lateral lumbar vertebral body screws for fixation, as revealed by our recent study [35].

To improve long-term mechanical stability, we modified our previous modular hemipelvic prosthesis and made it possible to fulfill rigid fixation to the sacrum and to preserve the posterior pedicle screw-rod fixation meanwhile (Figs. 15.3 and 15.4). The EBM 3D printing technology was also applied to generate a porous interface to facilitate bone ingrowth. In this cohort, 15 cases involving type I + II resection (5 cases of type I + II, 1 case of type I + II + III, 8 cases of type I + II + IV, 1 case of type I + II + III + IV, all reconstructed with screw-rod-connected hemipelvic endoprostheses) remained alive at last follow-up, and their mean MSTS score was 17.7 of 30, better than those of previous literatures. Moreover, no patients experienced postoperative neuralgia, and no evidence of bone absorption, osteolysis, prosthetic loosening or breakage, or shifting was observed at last follow-up. Nevertheless, it needs further observation to figure out whether bony fusion between the endoprosthesis and host bone could be achieved in the future.

15.3.4 Advantages of 3D Printing Technology in Reconstruction for Pelvic Defect

Advantages of our 3D-printed pelvic endoprostheses are listed in the following. Firstly, the contours of the iliac endoprosthesis and the standard hemipelvic endoprosthesis were designed based on intensive analysis of pelvic CT scans and could always match perfectly to the resection plane during operation. Secondarily, the endoprostheses were still fabricated in modular patterns with different sizes, not only to allow more flexible osteotomy but also to facilitate adjustment of the height and angles of the acetabular ring. As a result, we did not need intraoperative navigation for resection guidance but could still achieve precise matching between the implant and host bone. Thirdly, the size and density of the pores on the surface of the implants were determined according to previous biomaterial studies that could induce bone ingrowth and achieve long-term fixation [8, 31–33]. Moreover, we had no cases of deep infection in this cohort, which might be due to the short operation time that reduced intraoperative exposure, the small sizes of the endoprostheses that achieved good soft tissue coverage, sufficient wound drainage by two thoracic drainage tubes that minimized risk of hematoma, and prolonged prescription of broad-spectrum antibiotics that prevented perioperative infection.

The major limitation of our study was the relatively short duration of follow-up. Mechanical complications such as aseptic loosening and screw/rod breakage might not occur until several years after operation. The efficacy of osseointegration potential of 3D-printed materials would require further observation to confirm. Other prosthetic complications, implant survival, and oncological outcomes would also need longer follow-up to determine.

Based on the results of our study, we concluded that application of 3D-printed pelvic endoprostheses for reconstruction of bony defect was feasible with satisfactory safety and good functional status.

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16 Navigation in Pelvic Tumour Surgery

J. G. Gerbers and P. C. Jutte

16.1 Introduction

Computer-assisted surgery (CAS) is the term used to describe a relatively new concept of application of computers to enable pre-operative planning and provide continuous intra-operative orientation, instrument feedback and guidance. This concept enables surgeons to objectify the spatial position of anatomical locations, instruments or implants.

As it is a new technology, literature on CAS applications in bone tumour surgery is still relatively scarce. The field of pelvic and sacral osteotomies is however by far the most studied application of the use of CAS in the orthopaedic oncologic field. This is with reason as the treatment of deep-seated, often high-grade, lesions in an anatomically complex region is regarded as highly demanding. While there are advances in the development in chemotherapy in Ewing sarcoma, high-grade chondrosarcoma and osteosarcoma, radical surgical treatment of a solitary sarcoma remains critical for good clinical outcome [1–3]. Reports in the literature however demonstrate a relatively high rate of inadequate (intralesional or smaller than required) margins [4–6].

That this is not only due to the difficulties of creating adequate surgical exposure within a complex anatomical region, or characteristics of pelvic tumours, is demonstrated in experimental studies. Simulation of pelvic resections by Cartiaux et al. on plastic sawbones has shown that even experienced surgeons can struggle with orientation. Four surgeons could achieve a good 10 mm resection margin in only half of the resections, with 5 mm tolerance, on sawbones (without soft tissue) [7]. The authors called for larger surgical margins to compensate for the inaccuracy. However, larger margins implicate higher surgical damage to surrounding tissue that with more accuracy and precision would not be strictly necessary. A later follow-up study using 10 senior and 13 junior surgeons found 5 out of 23 intralesional resections in the freehand group [8], validating the smaller study. The lack of orientational accuracy may thus be a significant factor in the high number of intralesional resections and local recurrences in the surgical treatment of pelvic sarcoma [9–11].

Increasing surgical precision and accuracy can potentially have more benefits than only a decrease of intralesional resection rates. Through decreased required margins, it can allow for more a complex, tissue saving (and in case of pelvis even joint saving), surgical plan, fitting in the trend of limb or joint salvage surgery [12].

16.2 The Use of CAS

Most commercially available CAS systems use a stereoscopic optical device to follow the patient and the instruments. This is done using trackers attached to instruments and the patient, either active, emitting infrared light, or passive, reflecting infrared light from light sources in the camera mount (Fig. 16.1). The system can then calculate the position

Fig. 16.1 CAS system in the operating room. The stereoscopic camera is visible on the right. The surgeon is looking at the imaging feedback on the right. A battery-powered active tracker is visible on the curette

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in 3D space using the difference between the two camera images, pulse timing data and reflective/active array size and position. This position data can then be used together with imaging or models to provide feedback.

While functionalities in software differ between manufacturers, the systems can be divided into two basic categories: imageless and image-based navigation systems. The imageless mode, mainly used in joint replacement prosthesis surgery, uses computational kinematic and/or statistical models. This can be used for tumour prosthesis placement, for example, in large resections to reconstruct the joint line.

Image-based workflow uses three-dimensional imaging and is most often used in orthopaedic oncologic surgery. The datasets can be CT, MRI or PET-CT scans. High-resolution scans are required to achieve high accuracy with slice thickness optimally less than 1.0 mm. MRI and PET scans often lack sufficient resolution to be the primary dataset for use in navigation. Furthermore bone surface matching on MRI is, in our experience, not as precise as CT-based matching. A combination of datasets, for example, CT and MRI, enables the use of the better tumour delineation of MRI-GD on a precisely matched CT-based image. Most systems provide automated or manual matching for imaging datasets. Usually this is done before surgery, as a step before planning the resection planes and screw trajectories and marking points of interest.

To use this combined data in the OR, the imaging datasets have to be matched to real-world 3D coordinates. Most systems use landmark (point to point) and further surface matching but can also be performed using 2D (CT-fluoroscopy matching) or 3D (integrating an intra-operative CT using an Iso-C fluoroscope or CT in the OR) image acquisition. Pair point and surface matching are explained in Fig 16.2.

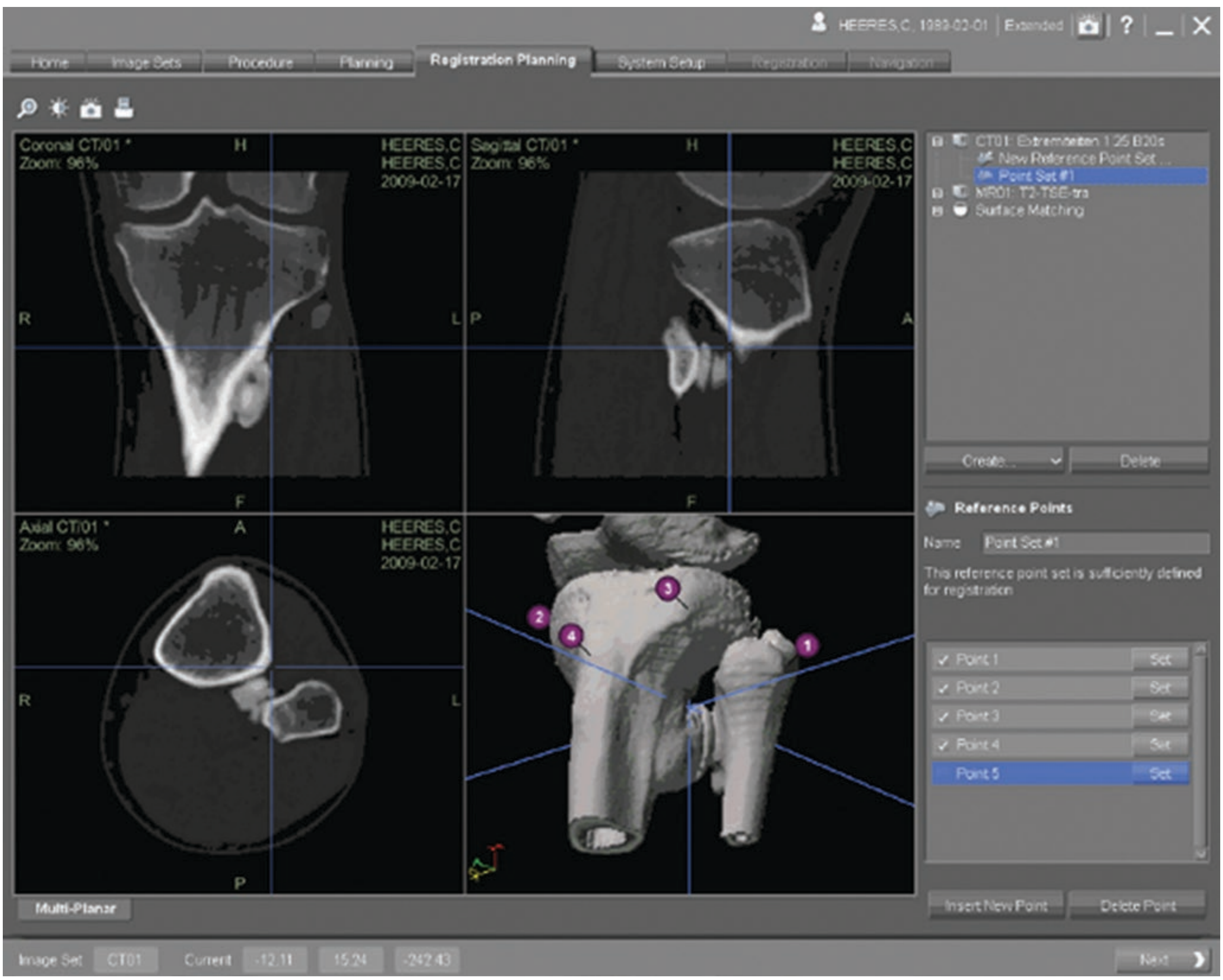

Fig. 16.2 The two phases of setup of a CAS system. (**a**) shows point-to-point matching with the use of landmarks that are registered both in software and on the patient. This rough fixed is improved by surface matching (**b**). Using the pointer tool drawn across the bone surface, registration points are collected that are then matched to the computed bone surface

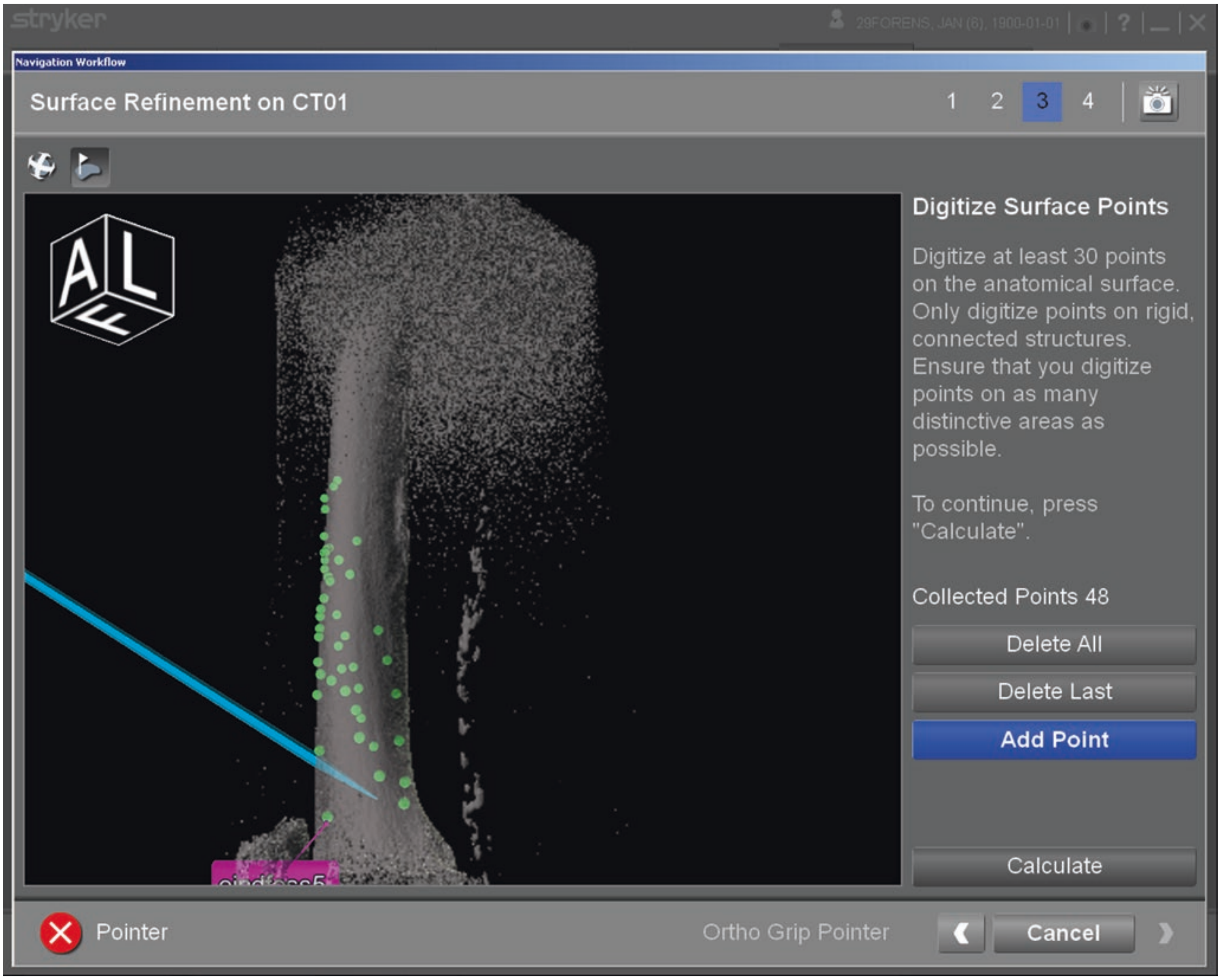

Fig. 16.2 (continued)

16.3 Literature on CAS Use in Pelvic Resections

The first uses of CAS for navigated pelvic and sacral resections were described by Hüfner et al., in two cases, and Krettek et al., in one case of periacetabular sarcoma, both in 2004 [13, 14]. All three were successful. Reijnders et al. reported the radical resection of two high-grade pelvic sarcomas [15]. So et al. described surgical workflow with CAS and reported two successful pelvic procedures with an average deviation of planes of 6 mm over three planes, as part of a larger case series [16]. Wong et al. were the first to describe the use of CT/MRI fusion to improve pre-operative planning and identification of margins [17]. The same group has also published papers on the successful use of integration of computer-aided design (CAD) planning and the use of custom prosthesis for reconstruction [18, 19]. Docquier et al. reported using CAS for both a successful pelvic resection and allograft reconstruction [20].

In recent years, larger case series with longer-term follow-up have been published. Cho et al. published about seven pelvic and sacral resections, part of a larger study of 18 patients. Clear margins were achieved in all resections; there were two local recurrences in the pelvis at a minimum follow-up of 3 years [17]. Wong et al. achieved wide resection margins in 16 cases and marginal resections in five, in a study that included 12 pelvic and sacral resections on a total of 21 cases. Local recurrences were found at a mean follow-up of 39 months in four cases that had had marginal resections; three of these were in the sacral region [19]. Young et al. described eight CAS-assisted pelvic sarcoma resections, all with clear margins and eight planned planes within 5 mm of the planning and one with 5 mm deviation due to surgeons' planning errors. All patients were alive and recurrence-free at a mean follow-up of 25 months [21]. In an overview of CAS use in 130 patients, Gerbers et al. described 17 pelvic resections; 15

had clear margins (R0 resection) with one intralesional R2 resection in bone and one R1 soft tissue margin. There were three local recurrences [12]. Jeys et al. have published the largest case series on pelvic resections, a study of 31 patients [10]. A clear resection margin was achieved in the bone in all cases; overall intralesional resection rate was 8.7%. At a follow-up of 13.1 months, four patients had a local recurrence.

The indication of CAS for pelvic and sacral resections, while not tested in a direct randomized controlled trial, appears clear. Out of a total of 91 reported procedures in literature, there was only one intralesional resection in bone reported (1%), and if soft tissue margins were accounted, there were five in total (5%). Long-term outcome is however not yet available.

16.4 Applications of CAS

Because the story of CAS is perhaps somewhat abstract and difficult to envision from the text, we will elicit the concept with some clarifying examples. The next cases will demonstrate different clinical and technical aspects of CAS use and help to envision its possible benefits. While tumour resection is the primary indication for CAS use in the pelvis, there are other applications where CAS can assist. Examples are curettages and placement of tumour prostheses.

Case 1: Joint Salvage Surgery

A 46-year-old woman presented with progressive pain in the right hip region in a first-line clinic. Four years earlier, during a presentation also for right hip pain, a pelvic radiograph showed a small lesion in the right os pubis, in the upper ramus. This was interpreted as a cyst. No follow-up was performed. Due to progressive stiffness in the right hip, she presented again, and a radiograph and later a normal MRI were made.

After referral to an oncological centre, a new MRI-GD demonstrated a lesion to both the rami of the right os pubis with expansion to the os ischium, involving part of the acetabulum. A biopsy demonstrated chondrosarcoma grade two. The tumour measured up to 9.3 cm in length, and volume was calculated as 92.69 ml (with one episodic and one cylindrical arm) using the method cited [22].

The surgical options were a joint-sparing local procedure or a larger segmental resection and saddle prosthesis placement. A CT scan was made to analyse the first option, demonstrating the possibility to salvage a little under two thirds of the acetabulum. There was no arthritis. A local resection, modified type 2/3 on the Enneking et al.'s classification, was planned in the CAS system on the fused images of CT and MRI (Fig. 16.3).

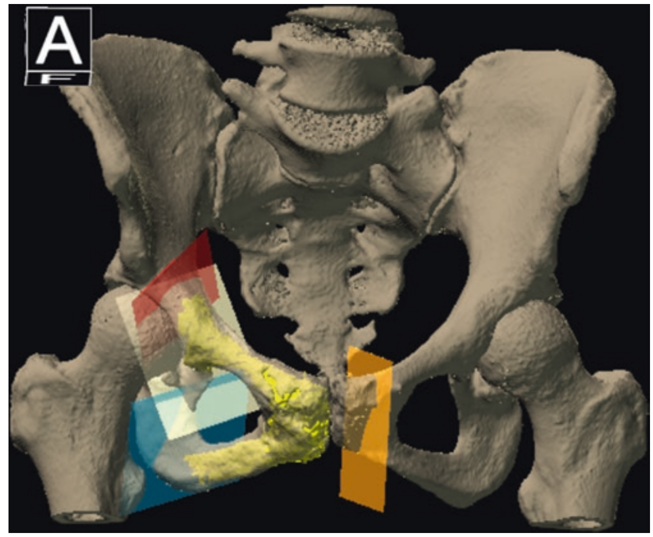

Fig. 16.3 3D view of the surgical planning of the resection planes in the Orthomap oncology module with tumour coloration on the fused MRI/CT image. The MRI is hidden, and only the tumour is selected. The planes are placed using minimum distance measurements

Two tracker pins were placed in the right crista iliaca as a stable basis for patient tracking. CAS setup using point and surface matching showed an acceptable mean system error of 0.5 mm. The deep resection planes were identified and checked for a safe margin using the system, and a K-wire was placed to mark the plane. The resection plane through the acetabulum was identified using the tracker tool and marked using diathermy. Using a navigated osteotome, the acetabulum resection was then performed, with intermittent checks for tumour margin. The same process was followed for the symphysis resection and ischial resection plane.

Pathological examination showed wide margins with a macroscopic and microscopic R0 resection. The acetabular resection plane had a minimum margin of 1 cm, as planned. Post-operative functions were excellent with immediate ambulation (Fig. 16.4). Rehabilitation time was short, and follow-up was event-free for over 10 years now. The use of computer navigation was vital to achieve the desired safe margins and provide the confidence to perform such a resections.

Case 2: Sparing Surgery

A 22-year-old patient developed moderate back pain after her first partus. A few months later during a physical activity, the pain in the back and leg suddenly worsened and could not be treated with heavy pain medication. The patient described no neurological symptoms, and the patient was referred to a rehabilitation specialist for analysis. A radiographic series of the lumbar spine displayed no evident pathology. Due to a positive Lasègue test, a MRI of the lower spine was made. This and the later MRI of the pelvis showed a large sacroiliac mass with involvement of the neural foramina on the left. When the patient presented in the tertiary centre, there was L5 neuropathy. Pathological analysis showed a high-grade conventional osteosarcoma. Neoadjuvant chemotherapy using doxorubicin and cisplatin was started, and a surgical plan was developed.

The two main surgical challenges were the involvement of the L5, S1 and S2 neural foramina (Fig. 16.5) and the reconstruction of the defect. A wide resection would not only involve resection of the nerve roots encased in the tumour but also likely severely limit the stability of the lower spine and pelvic girdle. Due to the complex shape of the tumour and the complex anatomical area, further (iatrogenic) damage to nerve or bone was something that should be prevented.

A safe but minimal surgical plane, to maximize post-operative stability, was chosen with surgical exposure from both the front and the back. The navigation system was first used for pedicle screw placement and placement of screws in the right sacrum. Then using a tracked osteotome, the pre-planned surgical planes were identified and followed (Fig. 16.6). A vascularized fibula autograft was used to reconstruct the left pelvic ring.

Again the surgical system provided the confidence of performing a highly complex procedure. Furthermore using the CAS system to achieve accurate screw and graft placement

Fig. 16.4 AP radiograph of the pelvis taken 3 months after surgery

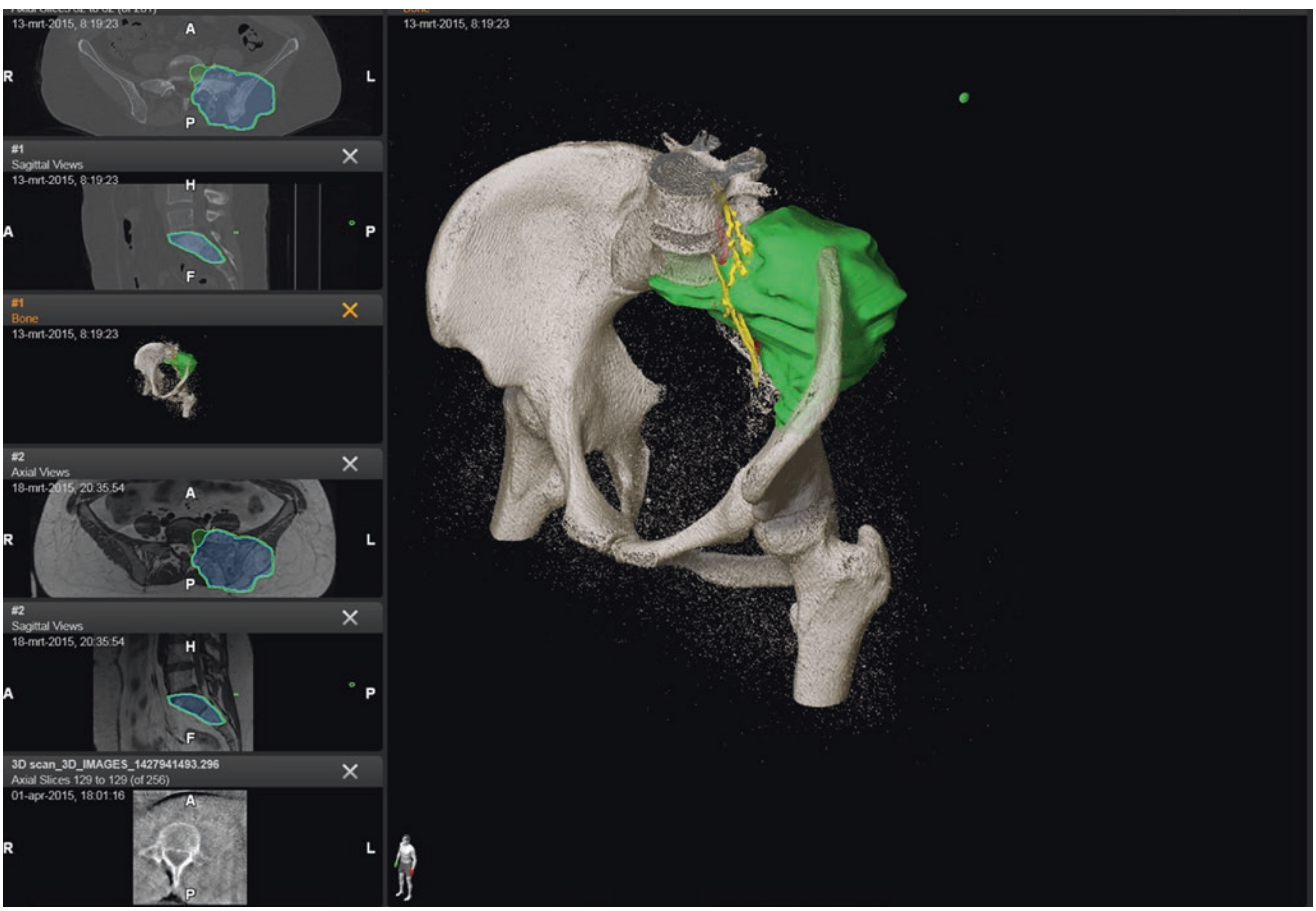

Fig. 16.5 Large 3D volume render on the right of the pelvis, the tumour and overlying critical structures. On the left side, views from multiple imaging sets are visible. The tumour was segmented on the MRI visible in the #2 screens

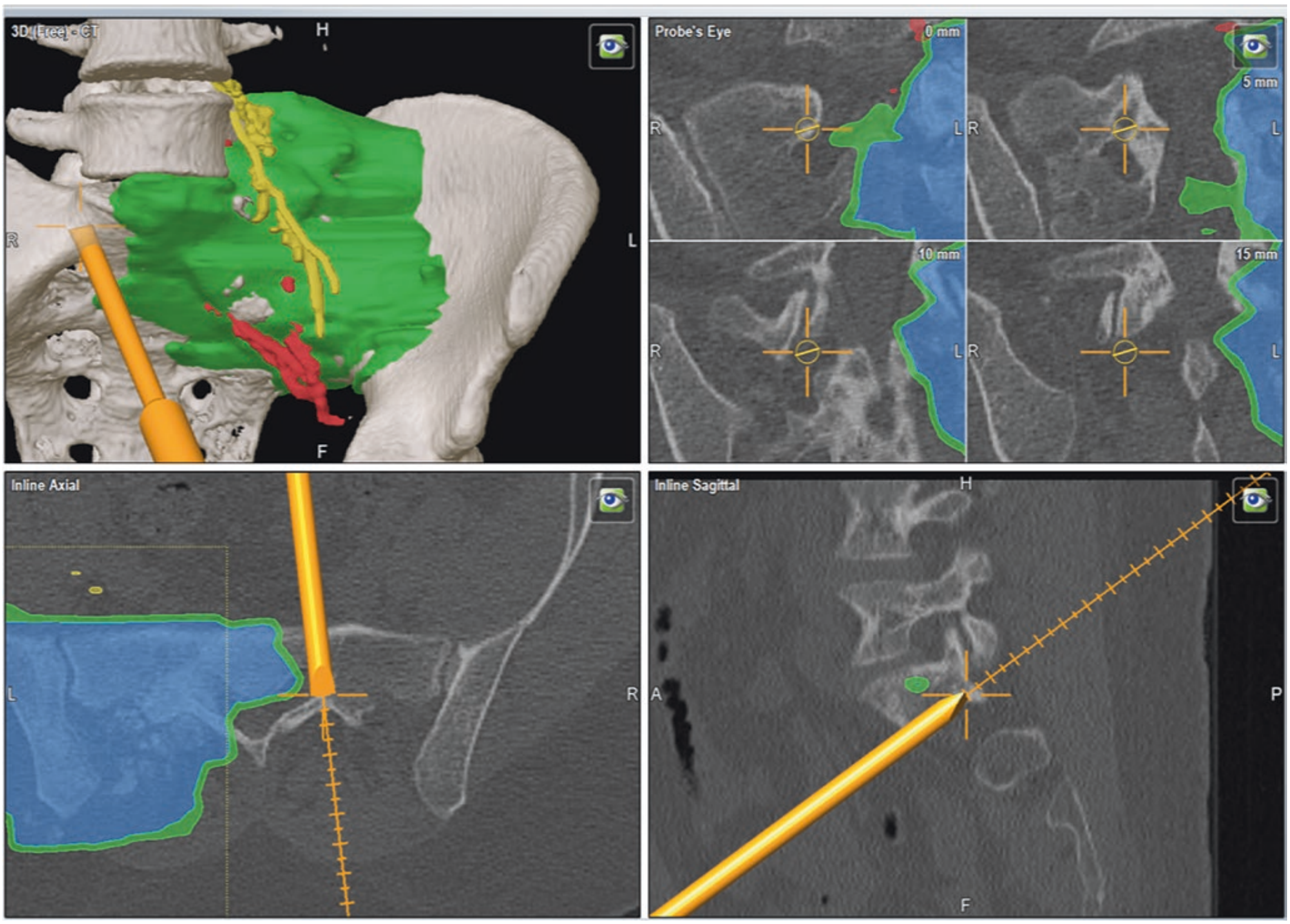

Fig. 16.6 Representation of the position and orientation of the osteotome as the surgeon would see it intra-operatively

provided a stable end result. At 3 months, the patient was training in a revalidation setting at 50% load-bearing capability.

Case 3: Intralesional Treatment

Another application where CAS can benefit is the curettage of pelvic tumours. A 39-year-old woman presented with intermittent pain of the right hip region depending on physical activity. Radiography in a tertiary clinic showed a large supra acetabular lesion with a multilocular and lucent aspect, and the patient was referred. The differential diagnoses included chondroblastoma, aneurysmal bone cyst, fibrous dysplasia or a simple bone cyst. As the lesion is deep-seated, intra-operative imaging is required for a successful procedure. The current standard for this type of procedure is the use of fluoroscopy.

A CT for CAS navigation was made and fused with a previously made MRI, and the tumour was segmented and coloured for easy identification. During the procedure, a tracker was attached to a curette, and the angled tip was tracked. Composition Fig. 16.7 demonstrates an intra-operative view of the curette positioning in the lesion. The tip has to reach the segmented edge or beyond for a successful curettage.

The CAS system allows for unlimited imaging feedback compared with the intermittent time-limited view of the fluoroscopy system. As the CAS setup was performed using point/point and surface matching, there was no intra-operative ionizing radiation use. Furthermore the technique provides 3D feedback and the possibility of defining regions of interest. Standard fluoroscopy is a 2D imaging technique and requires an external frame of reference (i.e. the surgeon's knowledge of anatomy) for precise instrument positioning.

Alternative CAS techniques include the use of intra-operative 3D imaging using isocentric fluoroscopy systems. This combined with image matching can provide a direct intra-operative check of complete curettage. This is something that is not possible in an image-based CAS procedure as the result is 'virtual'.

Case 4: Tumour Prosthesis Placement

A 57-year-old man presented with painless macroscopic haematuria over the course of 2 weeks. A year before, he had

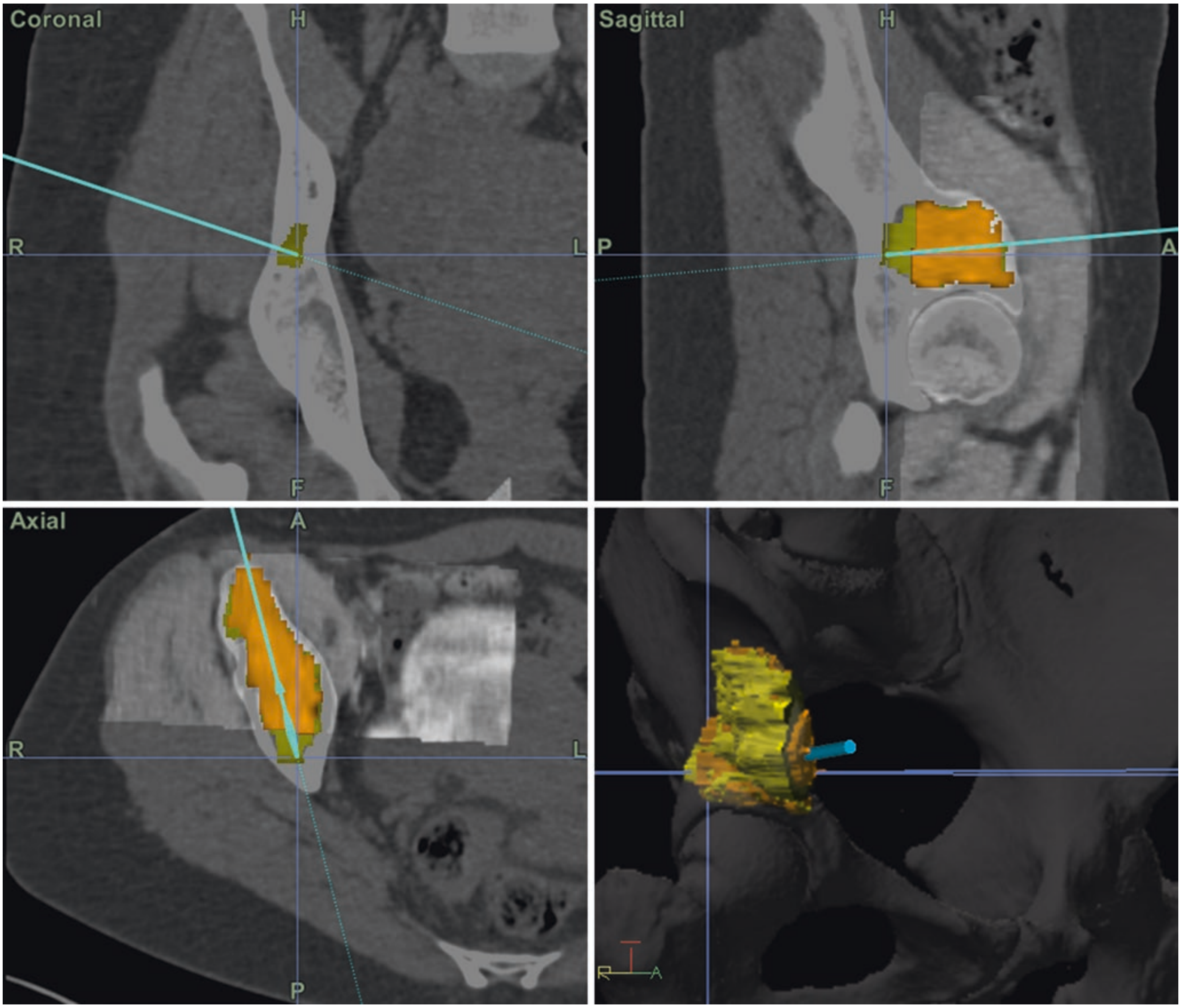

Fig. 16.7 View of the navigated curette inside the segmented tumour. The image is composed of a fusion of a CT and MRI scans. The tumour is segmented on both scans (orange and yellow)

already visited an orthopaedic surgeon for right hip pain. A radiograph showed only mild degenerative changes to the hip joints. As the patient history included prostate carcinoma, an MRI was performed that showed a large lesion above the right acetabulum. A biopsy showed a grade 2 chondrosarcoma. Due to the size of the lesion, a type 2 resection was required. Reconstruction would be done using a tumour prosthesis.

A pre-operative planning was made to minimize the risk of leg length discrepancy. A new joint line was planned using the known dimensions of the LUMiC modular ice cream cone prosthesis and an uncemented ABG stem and head. As the ice cream cone prosthesis is placed over a guide wire, a trajectory was planned in the centre of the desired position (Fig. 16.8). Resection planes were then planned at the correct angle and offset from the planned prosthesis.

The tumour was resected by first marking the planes using the pointer tool and then by using a calibrated osteotome. Then an electric drill (Colibri) with a tracker attached and calibrated using a universal calibrator was used to place the guide wire along the planned trajectory to the correct depth. The prosthesis was placed over the guide wire.

The use of CAS decreased the complexity of the surgical procedure by enabling easy identification of the correct resection height and plane orientation. Furthermore, the most critical aspect of the reconstruction, the correct tumour prosthesis placement, was reduced in complexity to following a planned line. This resulted in a tumour prothesis placement matching pre-operative planning, reducing post-operative complication risks as luxation, leg length discrepancy or early wear.

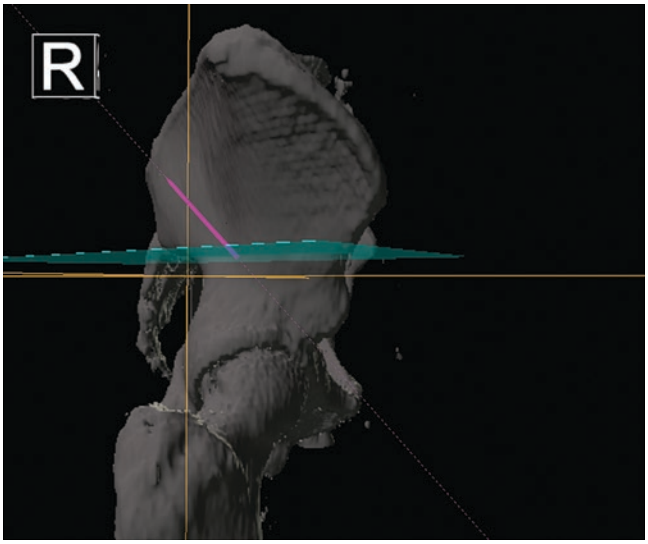

Fig. 16.8 Placement of the guide wire using the CAS system. The tool on the bottom right is similar in use to a horizon in an airplane. The cone has to be balanced over the cross to align the tool to the planned trajectory. Depth is checked on the sagittal, coronal and transverse views

16.5 Discussion and Implications for the Future

The cases above demonstrate the potential value that objective orientation in 3D can have in complex anatomy.

The main argument for the use of CAS in pelvic tumour surgery is the reported intralesional resection rate. Large overview papers on pelvic resections have reported intralesional resection rates, unknown if in bone or soft tissue, of at least 26% (Ozaki et al.) and 29% (Jeys et al.) [23, 24]. Fuchs et al. reported inadequate margins in 13 of 40 cases, or 33% [9]. The addition of CAS as far as reported in clinical setting showed much better results with a 1% intralesional margin and a 5% local recurrence rate in 91 cases.

Furthermore CAS allows for a more complex, tissue-saving, surgical plan, fitting in the trend of limb salvage surgery. Jeys et al. described that 'in several cases navigation allowed more complex surgical resections and reconstructions […] than […] possible using traditional methods' [10]. This resulted in the sparing of critical structures (nerve roots, avoidance of amputations), the ability to operate otherwise inoperable tumours and more functional reconstructions. Other authors described similar views [21, 25].

All papers' reporting cohorts do not yet draw conclusions on oncological parameters because of the length of follow-up. Wong et al. made the first careful conclusion in a review that results at early interval may be better than comparative studies [19]. It is however likely, considering the higher number of radical resections and the risk of recurrence in non-radical resections, that future reports will show clinical improvements.

While there is a possible bias, as likely only early adopters with positive CAS experience publish case reports, results reported are very satisfactory and surgeons performing pelvic and sacral resections may benefit from CAS introduction to their practice, as well as their patients.

Local ethics committee guidelines were followed, and all patients were provided with written consent for publication.

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17 Pelvic Tumor Surgery in Children

Rodolfo Zamora, Stephanie Punt, and Ernest U. Conrad III

17.1 Introduction

The care of pediatric pelvic tumors adds the challenges of caring for children to the challenges of caring for pelvic tumors. The pelvis is a less common and more complex anatomic location for tumor resections than extremity tumors, and pelvic sarcomas require more planning for resection than extremity tumors. Pelvic tumors are typically larger than extremity tumors and present more challenging resections because pelvic sarcomas may encroach upon the adjacent iliac vessels, sacral nerve roots, acetabulum, sacrum, or bladder and bowel. Surgical pelvic resection has historically been defined by the Enneking classification that recognizes tumor resections that are located in the posterior (sacroiliac, Type I), lateral (acetabular, Type II), or anterior (obturator, Type III) pelvis (Fig. 17.1) [1]. The most common osseous pelvic sarcoma diagnoses include Ewing's sarcoma (20–25% pelvic primary tumors) and osteosarcoma (10% pelvic primary tumors) [2, 3]. Chondrosarcoma and metastatic pelvic tumors are uncommon in children, while benign bone tumors in the pelvis are relatively common.

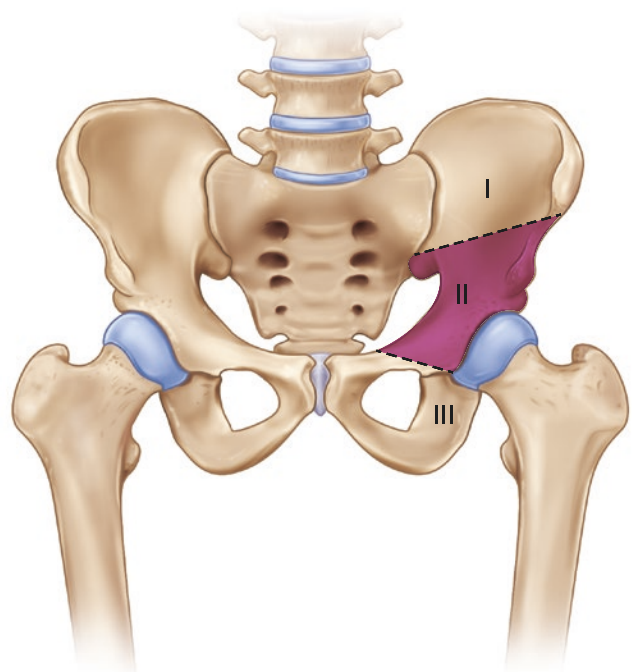

Fig. 17.1 Enneking classification of pelvic tumor resections

17.2 Pediatric Versus Adult Pelvic Resections

The resection of tumors located in the immature pediatric pelvis is sometimes less challenging to resect than adult pelvic sarcomas because of the greater sensitivity of many pediatric sarcomas to preoperative chemotherapy compared to adult sarcoma patients. That greater sensitivity to chemotherapy allows preoperative treatment that results in a tumor that is physically less invasive and more easily removed. This sensitivity also allows closer surgical margins and more effective surgery with better local control of their tumor for children younger than age 16–18 years. Assessing pediatric

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sarcomas or their preoperative chemotherapy response is an essential part of making surgical decisions regarding their surgical treatment.

Surgical resection in the skeletally immature patient also becomes less challenging if the acetabular and femoral epiphyses are spared by surgical resection. That option is easier to achieve in a younger patient who is more likely to have a good response to chemotherapy, indicated by greater tumor necrosis, and is more likely to have a resection with acceptable osseous margins. Making that decision is the essence of preoperative planning for pediatric pelvic sarcoma surgery. The resection of bone sarcomas in patients under the age of 14–16 years also requires accommodation for skeletal growth, planning for a life expectancy that may exceed oncologic implant survival, and an expectation for levels of postoperative physical activity that exceeds the usual adult expectations.

While preoperative chemotherapy is an important adjuvant therapy for both pelvic osteosarcoma and Ewing's sarcoma, preoperative radiation therapy has historically been utilized mostly with Ewing's sarcoma patients. Ewing's sarcoma is a relatively radiosensitive tumor, and radiotherapy can be employed pre- or postoperatively or in palliative situations as an effective treatment for local control. Osteosarcoma has historically not been considered to be a radiosensitive tumor. Surgical pelvic resections are not contraindicated in skeletally immature, unless the patient is younger than 10 years old and has acetabular involvement. Patients under the age of 10 years old with a pelvic Ewing's sarcoma involving the acetabulum should be considered for radiation therapy without resection for local control rather than surgical resection, if the patients have demonstrated a good response to preoperative chemotherapy on preoperative imaging. Patients of any age without a good response to neoadjuvant chemotherapy (based on preoperative imaging) are at a greater risk for local recurrence after radiation only and are at greater risk for requiring amputation. Nonsurgical treatment for local control (radiation without resection) of pelvic sarcomas does carry a greater risk of local recurrence than the use of both resection and adjuvant radiation therapy for local control [4, 5].

17.3 Predicting and Achieving Surgical Margins with Pediatric Pelvic Tumors

Adequate tumor resection margins are the most important goal for a successful tumor procedure in any patient. Resections should be based on a careful preoperative assessment of the response to preoperative (neoadjuvant) chemotherapy that is accomplished with careful review of preoperative MRI, CT, and positron emission tomography (PET) imaging that should assess the tumor size, soft tissue and osseous involvement, tumor inflammation, and proposed surgical margins [6]. The best assessment of the patient's response to preoperative chemotherapy involves a comparison of both MRI and PET scan imaging before and after the first 10–12 weeks of preoperative chemotherapy [7, 8]. That comparison of imaging before and after preoperative chemotherapy is a critical sign of whether the patient's tumor is responding to preoperative chemotherapy. The assessment of the neoadjuvant chemotherapy "response" with a comparison of MRI and PET imaging is an essential part of planning surgical margins in all patients treated with preoperative chemotherapy, because a good response to chemotherapy will allow closer surgical margins at resection and the possible preservation of important anatomic structures (Fig. 17.2). Close or smaller tumor margins (1.0–2.0 cm), however, are at higher risk for tumor contamination and subsequent tumor recurrence, whereas wider, greater margins (2.0 cm or greater), with the presence of a normal cuff of tissue, are theoretically at less risk of tumor recurrence [3, 9]. Evidence-based guidelines for determining the adequacy and quality of a tumor margin and the pathologic description of surgical margins are challenging and become more complex in the pelvis [10, 11].

The process of evaluating preoperative imaging should be determined with imaging guidelines. On T2 or STIR MR imaging sequences, the increased signal surrounding the tumor indicates inflammation and possible microscopic tumor at the tumor interface with surrounding normal tissues. With effective neoadjuvant preoperative therapy, this zone may disappear or shrink and allow a closer margin of resection [12] (Fig. 17.2). PET scan imaging is a quantitative assessment of the glucose uptake by the tumor as reflected in the standard uptake value (SUV). A 50% reduction in the PET SUV will typically reflect a good response when comparing pre- and post-systemic therapy PET scans.

A "good response" to preoperative chemotherapy may shrink the peritumor inflammatory zone and/or may result in a small decrease in tumor diameter as visualized on preoperative MRI [12–14]. A good response to chemotherapy may also result in apparent necrosis within the tumor mass and a subsequent reduction in pre- and post-chemotherapy PET scans. Multiple published adult sarcoma studies regarding the assessment of treatment response to neoadjuvant chemotherapy and radiation therapy using fluorodeoxyglucose PET show that a significant (50% or greater) decrease in tumor fluorodeoxyglucose uptake is predictive of patient survival [6, 8]. Criteria for acceptable or successful bony margins are difficult to interpret from the literature, but a comparison to extremity sarcomas is helpful [3, 9–11, 15, 16]. A review of pediatric sarcomas involving the distal femur by Zimel et al. demonstrated that resections with larger surgical margins compared to intercalary joint-sparing resections with smaller margins did not show a significant difference in local recurrence [17]. The role of local recurrence following resection

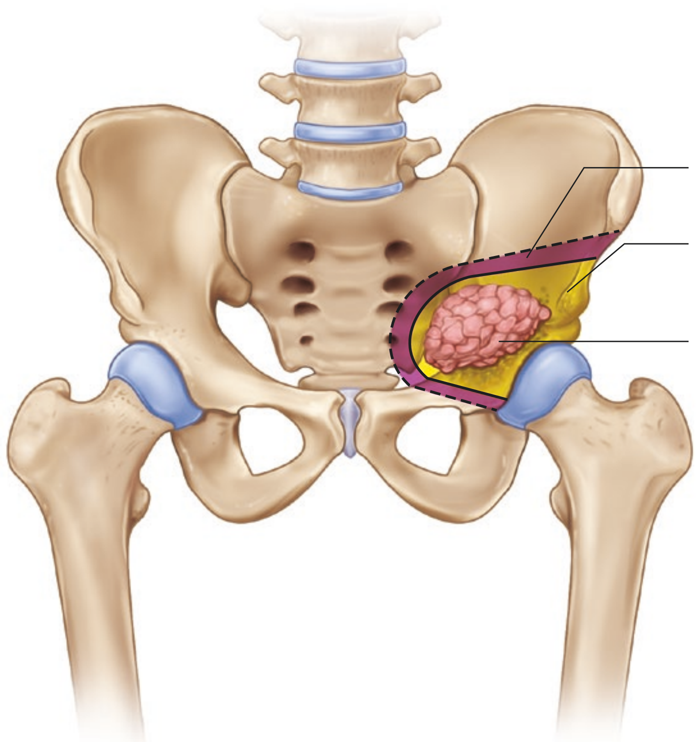

Fig. 17.2 Preoperative planning—predicting tumor margins by preoperative MRI

is associated with the adequacy of soft tissue and osseous surgical margins and should be carefully evaluated for all pelvic resections [18, 19]. The surgical goal for extremity surgical margins of 1.0 cm should be increased to 2.0 cm, if possible, for pelvic margins because of the higher risk of recurrence. In addition, recent margin assessments in osteosarcoma and Ewing's sarcoma suggest that soft tissue tumor margins are more challenging than bony margins and are more likely to contain microscopic, margin contamination, leading to local recurrence [3, 10].

Computer navigation assistance with pelvic resection is an attractive planning tool for pelvic resection because of its ability to improve and document the accuracy of resection in the pelvis and to improve the documentation of the resected specimen and the associated margins. The surgical experience with navigation requires accurate system "registration," reporting of surgical margins, and a comparison of local recurrence with or without the technique in order to demonstrate surgical efficacy [20]. Navigation requires documentation of the preoperative and intraoperative surgical margins that can be compared to the intraoperative and final postoperative plan and margins. Those comparisons represent important metrics for confirming the effectiveness of navigation for sarcoma resections. Demonstrating navigation accuracy with minimal radiation exposure (intraoperative CT "registration") is another issue to be resolved by future navigation protocols for pediatric patients [1, 21, 22]. Intraoperative navigation requires the "registration" or downloading of preoperative MRI and/or CT imaging and the intraoperative "registration" of the patient to that imaging [17–19] (Fig. 17.3e). The use of intraoperative navigation is able to provide a "GPS" function that will improve the accuracy and planning of both surgical tumor margins and reconstructions and is particularly useful in the pelvis.

17.4 Ewing's Sarcoma and Osteosarcoma of the Pelvis

Surgical resection of Ewing's sarcoma in the pelvis should consider multiple factors including the presence of metastatic disease, size of primary tumor, chemotherapy response, the anatomic location of the tumor, and patient age. Metastatic disease at presentation may serve as a contraindication for resection, although 20–30% of patients with lung metastases survive their disease (compared to 16% with bony disease) [4]. Patients with Ewing's sarcoma should be very carefully assessed at presentation for both pulmonary and osseous metastases. Patients with osseous metastases or multiple lung metastases should be considered for pelvic surgery with careful discussion of the risk of progressive disease.

Osteosarcoma of the pelvis is less common than Ewing's sarcoma and may be a more challenging tumor to resect with adequate margins. It has the same challenges as Ewing's of the pelvis with regard to surgical margins and is more common in older patients. Radiation therapy has historically not been effective for osteosarcoma, and preoperative chemotherapy is a critical pre-resection systemic therapy.

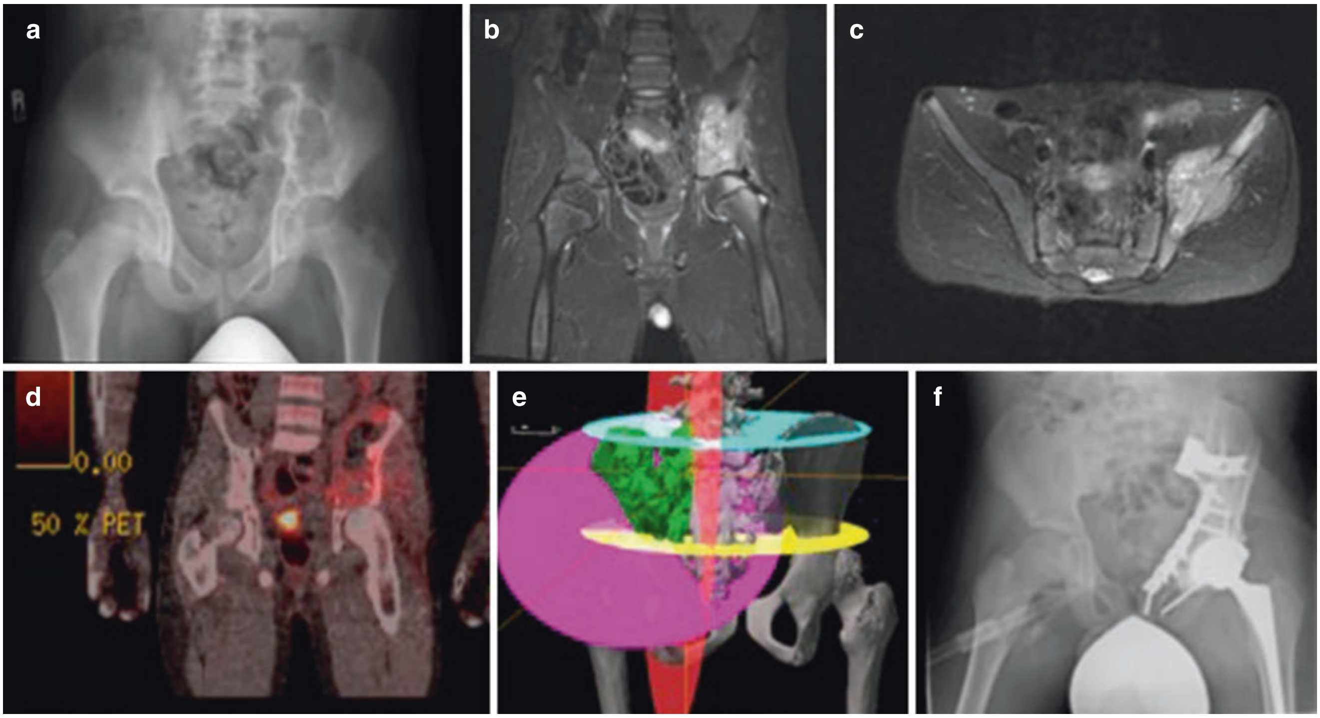

Fig. 17.3 Type II pelvic Resection and reconstruction—preoperative imaging (**a**–**c**), PET scan (**d**), navigation margin (**e**), postoperative composite allograft/THR (**f**). (**a**) Preoperative pelvic X-ray. (**b**) Preoperative pelvic MRI (coronal T2). (**c**) Preoperative pelvic MRI (axial T2). (**d**) PET scan image of tumor. (**e**) 3D image of navigated pelvic procedure. (**f**) Postoperative Type II alloprosthetic reconstruction

17.5 Pelvic "Composite" Alloprosthetic and Implant Reconstructions

Cadaveric pelvic allografts are an excellent reconstructive option for both Type I and II pelvic resections. Type III anterior, obturator resections without acetabular involvement are best managed with a Gor-Tex, allograft dermis, or other synthetic patch to reconstruct the inguinal canal and associated pelvic defect. When acetabular resection (Type II) is involved, a composite pelvic allograft and total hip reconstruction or custom implant should be considered to reconstruct the graft's acetabulum and patient's femoral head [23] (Fig. 17.3a–e).

Type II pelvic reconstruction can also be achieved with either custom or modular pelvic implants that incorporate a total hip reconstruction (Fig. 17.3f). Custom implants may be replaced by modular pelvic implants that incorporate sciatic notch fixation and modular acetabular components. "Saddle" prosthesis reconstruction has lost their popularity for pelvic reconstruction because of postoperative pain and instability at the "yoke"-patient osseous iliac interface [24]. While extremity sarcoma patients have been managed with "growing" implants, those implants remain associated with a high complication risk including implant aseptic loosening, sepsis, and failure of lengthening. There are significant limits regarding how large a segment these devices can lengthen (4–6 cm), and their use in the pelvis and hip joint is extremely limited. Flail reconstructions are a reasonable option for patients with large tumors and have demonstrated surprisingly good function postoperatively, and pelvic amputations should always be considered as a possible surgical option, especially in patients with large or recurrent tumors [5].

Type I and Type II pelvic allografts should be fixed to the remaining adjacent pelvis with plate and screw fixation at both the anterior pubic and posterior iliac osteosynthesis sites (Fig. 17.3f). That fixation should include at least six cortices of fixation at both host junction sites. Additional sacroiliac fixation should include two or three carefully placed large diameter sacroiliac screws if the graft involves the sacroiliac joint. If radiographic evidence of bony healing is not apparent on radiographs/CT imaging at the osseous junction site at 10–12 months postoperatively, fixation revision and autogenous bone grafting should be considered as an additional procedure [25–27]. Initial selection of pelvic allografts for reconstruction should be selected with consideration of patient and donor gender, age, and acetabular diameter. Pelvic allograft sizing can be achieved by the assessment of acetabular diameter for both the donor graft and, if possible, patient gender and age of grafts versus patients should be considered with graft selection [28]. Pelvic allografts carry a risk of postoperative infection but can achieve bony union at the host bone-graft interface and may offer better soft tissue attachment.

17.6 Surgical Exposure After Pelvic Resection

Pelvic resection, surgical exposure, and skin incisions are a critical consideration for a successful pelvic sarcoma resection and reconstruction. Choices for the surgical skin incision include the standard ilioinguinal incision from the pubis to the posterior lateral iliac crest. Patients may require more surgical exposure posteriorly or anteriorly depending on the location of their tumor, and their incision, exposure, and positioning should be adjusted accordingly.

Skin incision "extensions" of the standard ilioinguinal incision, transversing the groin and iliac crest, should include an anterior/longitudinal skin incision for obturator ring resection (Type III) with the associated exposure of the femoral vessels. A Type II resection with acetabular resection usually requires an anterolateral extension for total hip exposure and reconstructions. Posterior Type I resections may require a longitudinal sacral extension of the skin incision.

17.7 Postoperative Complications and Rehabilitation

Surgical complications vary by tumor size and location, the type of resection and reconstruction, age of patient, length of the surgery, extent of surgical blood loss, operative time, and surgeon experience and level of skill. Choices of pelvic reconstruction range from no reconstruction (flail limb) to nonanatomic endoprosthetic implants (i.e., saddle prosthesis) or anatomic composite pelvic allograft/total hip reconstruction. The most recent surgical trends for pelvic reconstruction favor "composite" alloprosthetic reconstructions, modular pelvic implant, or no reconstruction, frequently referred to as a "flail" pelvis. Pelvic amputation or hemipelvectomy is always an option for surgical resection.

One of the most significant factors in predicting surgical challenges and a higher incidence of postoperative complications is the inclusion of the acetabulum in the pelvic resection (Type II resections). Acetabular reconstructions require a decision to reconstruct the peri-acetabular ilium with a pelvic implant or alloprosthetic implant or a decision to leave the pelvis flail and unreconstructed. Acetabular reconstructions carry the risk of sciatic nerve injury, hip dislocation, and the challenges of fixation and graft healing. Pelvic amputation (hemipelvectomy) is usually reserved for patients with large tumors, neurovascular involvement, a poor response to chemotherapy, or a tumor recurrence after initial resection. When considering amputation as the primary resection, the surgeon should be aware of the recurrence risk for larger tumors because of the challenges of achieving adequate margins at the sacrum, spine, and/or other critical midline structures. Pelvic tumors with sacral or neurovascular involvement should be assessed preoperatively with caution for the adequacy of those surgical margins.

The risks of postoperative infections (20–30%), local recurrence, neurovascular injury, and massive intraoperative blood loss are the most common surgical risks with osseous pelvic resections [18, 19]. Critical surgical issues during tumor resection include the control of the external and common internal iliac and femoral vessels during operative exposure and resection (Fig. 17.4). The lack of adequate iliac vascular control is associated with a higher risk of massive intraoperative hemorrhage and perioperative death. It is also important to identify other critical structures including the sacral, sciatic, obturator, and femoral nerves and ureter. Damage to the peripheral nerve roots is most common for the femoral nerve, obturator nerve, and sciatic nerve. Those risks all increase with larger pelvic tumors, older patients, recurrent tumors, and revision surgeries.

Deep infections and delayed wound healing are the most common complications following pelvic resection. Those complications can be minimized with copious irrigation during surgery, shorter procedure times, lower blood loss, healthy wound closure, and attention to appropriate postoperative intravenous antibiotics. Approximately 50–60% of patients who undergo surgical resection and radiation therapy require a second operation, and approximately 15–20% of patients will have a major complication requiring multiple additional procedures. The risk of tumor local recurrence in pelvic sarcomas is 25–30% and related to tumor size and sensitivity to adjuvant chemotherapy. The incidence of revision surgery for pelvic allograft complications ranges from 25% to 50% with revision surgery most commonly occurring for graft infections, wound necrosis, nonunion of the pelvic allograft, instability of the hip arthroplasty, or tumor recurrence [15, 29]. The most common postoperative complication is postoperative infection, which occurs in 20–30% of patients within the first 4–6 weeks postoperatively and requires surgical washout with or without graft removal and extended antibiotics. Amputations are indicated for most tumor local recurrences and on rare occasions for postoperative complications. Wound infections can also be avoided

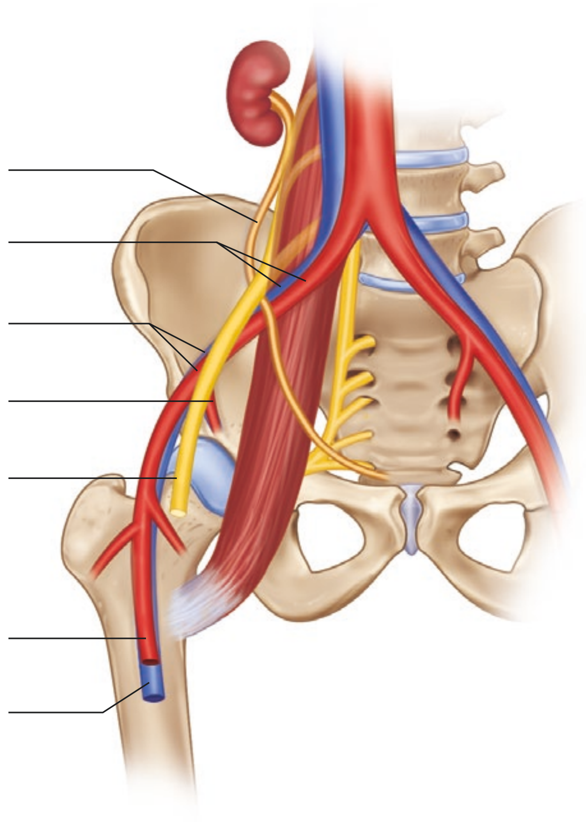

Fig. 17.4 Critical pelvic vascular anatomy

with careful consideration of appropriate surgical incisions and the judicious use of myocutaneous flaps for persistent drainage postoperatively. Published reviews for pelvic sarcoma resections demonstrate an overall risk of revision to amputation of approximately 10–15% [30, 31].

Postoperative rehabilitation following pelvic resections should be approached with a standard protocol specific to your practice and focus on preoperative and postoperative goals (Table 17.1). That protocol should be focused on the immediate postoperative management for the first 3 days, the first 3 weeks, and the first 3 months. The first 3 days should focus on a patient's blood loss during and after surgery, their vital signs, and nerve function. Patient pain control and neurovascular monitoring of the extremity should also be a high priority in the first 24 h and first 3 days. Routine Doppler vascular ultrasound should be carried out to confirm good arterial and venous flow in the iliac and femoral vessels. Venous Doppler to detect thrombosis should be repeated weekly for the first 4–6 weeks in all patients. Any problem with excessive postoperative bleeding at the surgical site in the first 48 h should be managed with intravenous embolization and appropriate transfusions.

Postoperative infection prophylaxis requires IV antibiotics to cover both gram negative and gram positive bacteria for the first 7–14 days postoperatively. All patients should be followed for possible infections with weekly CBCs/WBC for 3 weeks postoperatively to detect possible leukocytosis secondary to postoperative infections. Postoperative imaging should routinely include pelvic X-ray and a postoperative pelvic CT scan and venous Doppler. Pediatric patients are usually placed in a postoperative pelvic/hip brace for approximately 6–12 weeks with toe-touch ambulation with a walker or crutches for 6 months or until bony pelvic graft and implant healing is documented on postoperative radiographs. Transfer to a rehabilitation facility is typically considered at 2–3 weeks postoperatively.

Pelvic resection rehabilitation usually involves a postoperative hospitalization of 1–2 weeks. Patients are partial weight bearing for 3–12 months depending on the size of their resection and age. Patients receiving systemic therapies such as chemotherapy need to be able to resume their postoperative chemotherapy schedule in 2–3 weeks as a delay greater than 3 weeks can affect patient survival. Recovery for allograft reconstructions requires 6–12 months of partial weight bearing, while prosthetic reconstructions require 3–6 months. "Flail" pelvic resection patients and patients treated with hemipelvectomy also are able to recover their weight bearing status at 3–6 months.

Table 17.1 Pelvic resection preoperative, intraoperative, and postoperative checklists

I. Preoperative checklist
• First clinic visit—evaluate preoperatively, radiation therapy, chemotherapy, and surgical scheduling • Plan for surgical assistants (i.e., vascular, urology, general surgery specialists), preoperative embolization, and pelvic reconstruction (below) • Supplies—blood products including RBCs and platelets; check if implants and grafts are available • Medical consult—ICU reservation and preoperative medical consult • Imaging—MRI, CT, PET, surgical navigation, and intraoperative imaging • Pelvic implants/grafts—pelvic allograft order; THR system; plates/fixation
II. Intraoperative checklist
• Patient positioning and incision/surgical approach • Surgical assistants; anesthesia, central lines, and venous access • Transfusion—10 units RBCs, platelets • Operative implants, grafts, equipment, and instruments • Intraoperative navigation and imaging
III. Immediate postoperative checklist—first 3 weeks
• Overall medical status—CNS, cardiopulmonary, renal, metabolic • Vital signs—ICU, cardiopulmonary status • HCT, WBC, platelets, coagulation profile • Renal output/function, serum chemistry, and labs • Postoperative Doppler—femoral vessels on day 1–3, then once per week • Labs—CBC, pH, chemistry daily for 3 days, then once per week for 2 weeks • Postoperative plain X-ray/CT of the chest/pelvis • Estimated blood loss—OR vs. postoperatively vs. transfusions • Wound and drains—blood loss monitored for 72 h; wounds healthy • Pain control issues
IV. Extended postoperative checklist—first 3 months
• Vital signs—cardiopulmonary status • CBC/platelet • Renal output/function • Postoperative Doppler—days 7–14 • Labs—variable to rule out pelvic sepsis at 2–3 weeks with CBCs monitored • Wound drains taken out—postoperative days 10–21; wound healthy? • Chronic pain control • Pelvic brace for 6–12 weeks • Rehabilitation—toe-touch weight bearing for 6 weeks, partial weight bearing for 6 weeks, full weight bearing once bony healing is visualized on CT/X-ray (6–12 months); consider rehab/nursing home transfer • Oncologic follow-up imaging—chest/pelvic CT at 3-month intervals

17.8 Growth in the Pediatric Patient

The onset of puberty in children (age 10–14 years) is a critical milepost signaling the beginning of the last phase of skeletal growth. Younger preadolescent children have 10–20% of their overall skeletal growth and 15–25% of their femoral growth remaining at age 10 years. Girls 8–10 years of age have 6–10 cm of remaining femoral and proximal tibial growth, and boys have 9–12 cm remaining. This remaining growth makes surgical decisions for younger patients with osteosarcoma or Ewing's sarcoma more challenging [32]. Pelvic resection in the skeletally immature should not involve significant leg length discrepancies unless their resection is in a patient under the age of 12 years or involves interruption of the acetabular and or proximal femoral epiphysis (Fig. 17.5). Loss of height or length with iliac or pelvic resections is difficult to predict and measure and should be carefully measured radiographically along with length assessment of the lower extremities at regular intervals.

17.9 Physeal Biology and Predicting Pelvic Growth Issues

An epiphysis occurs in a weight bearing long bone, while an apophysis represents a growth center associated with an insertion point for a major muscle group (i.e., gluteus).

"Pressure" epiphyses, or normal growth plate "physes," such as the proximal femur or acetabular physes and iliac and trochanteric apophyses account for both hip development and longitudinal growth in the pelvis, acetabulum, and proximal femur. For example, the femoral neck can be significantly narrowed as a result of the placement of an intramedullary rod in the proximal femur in a child under the age of 12 years. Traction epiphyses or apophyses, such as the greater trochanter, or iliac apophysis does not involve the joint but does serve as a major muscle insertion point for the gluteal and abdominal muscles [33, 34]. The acetabular physis is a "triradiate" growth plate that produces acetabular/pelvic growth in three planes until it reaches maturity at approximately age 12 years (Fig. 17.5) [35–37]. Other apophyseal and epiphyseal maturation points serve as an important sign of growth in immature patients.

The growth plate is composed of chondrocytes in various stages of differentiation termination with endochondral ossification [36, 38]. The growth plate is composed of three zones: the resting, proliferative, and hypertrophic zones. In the resting or reserve zone, chondrocytes rarely divide, and nutrients are stored. In the proliferative zone, chondrocytes divide rapidly and arrange themselves vertically in columns. In the hypertrophic zone, chondrocytes enlarge and proliferate. As a result of the cellular division, the proliferative zone is at greater risk for being affected by neoadjuvant chemotherapy and radiation therapy physeal growth. Regulation occurs via a parathyroid hormone-related protein (PTHrP) feedback loop [39, 40]. This feedback loop affects the expression of Indian hedgehog (Ihh) which is responsible for chondrocyte proliferation and maturation and the rate at which this process takes place [39]. If disruption of the growth plate circulation or bone bridge formation occurs via trauma or surgical resection, angular growth deformities and/or limb-length discrepancies can result [40].

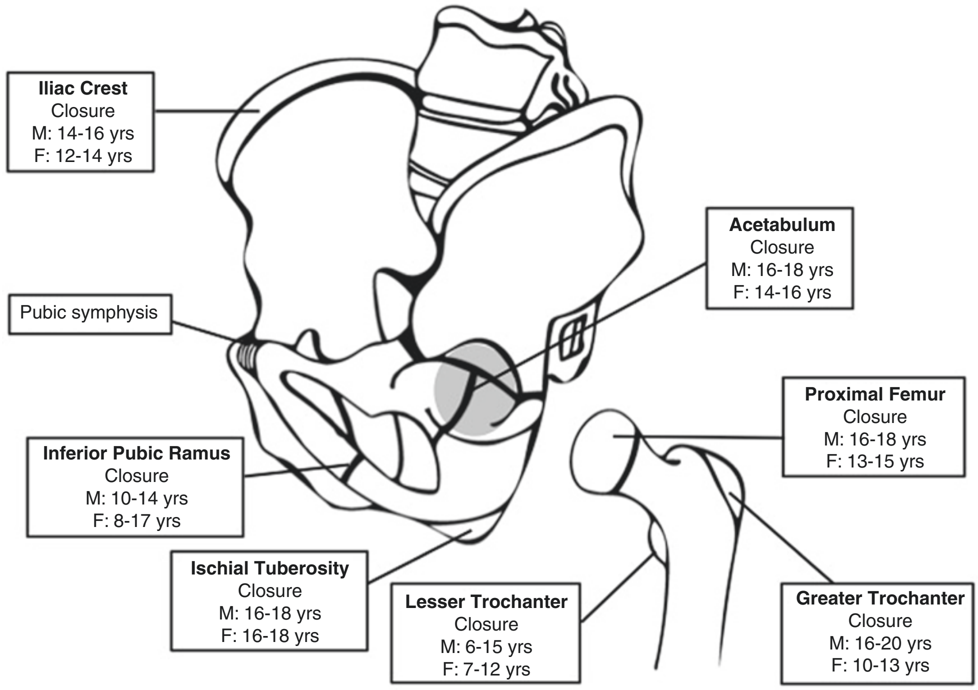

Fig. 17.5 Pelvic maturation milestones: pelvic growth plates and apophyses in the skeletally immature

17.10 Managing Leg Length Discrepancies in Children

The most important factor in assessing patients with a leg length discrepancy after surgery is the consistent tracking and documentation of the patients' bilateral extremity growth at regular 6-month intervals. It is critical to begin that "tracking" in the first 2 postoperative years such that postoperative epiphyseodeses are not delayed for 1–2 years. Assessing and predicting pelvic and acetabular growth is challenging because of the acetabular triradiate and more complex bony anatomy. Significant apophyseal growth occurs at the iliac crest (e.g., Risser sign) and traditionally has been used as an easy and common skeletal maturity assessment for pediatric orthopedic surgeons [34, 41]. Growth milestones for the acetabulum have not been well studied or documented in children, but the acetabular physis is thought to naturally reach maturity and "close" at 12–14 years of age [41–47]. Routine scanogram assessment of leg length should include assessment of the ipsilateral iliac and acetabular sciatic notch.

The management of leg length discrepancies can be simplified for small discrepancies that are less than 2 cm through the use of a shoe lift and nonoperative management. These shoe lifts can be made in variable sizes and built into a patient's shoes. They do not require any invasive procedures but have cosmetic limitations on how large a discrepancy can be corrected without patient complaints. Surgical methods can be utilized to partially correct anticipated leg length discrepancy concerns. At the time of pelvic resection, a reconstruction can be carried out in a way that effectively lengthens the limb by placing a pelvic implant or allograft that is slightly (1–2 cm) larger than preoperative length for the correction of future leg length discrepancies. This method is limited to corrections of 10–15 mm and requires precise pelvic reconstructive metrics that include the proximal femur and acetabulum and ilium.

Arresting the physes of the contralateral "normal" leg is a popular and effective option for addressing leg length discrepancy after resection and could be considered for a leg length discrepancy after pelvic resection with arrest of the contralateral distal femoral physis. This method should be carried out within the first year postoperatively in order to minimize the magnitude of the discrepancy. Depending on the number of physes arrested and the timing of epiphysiodeses, a discrepancy of 2–3 cm could be addressed with an epiphysiodesis of the contralateral distal femur but at the expense of the patient's ultimate overall height at skeletal maturity. This procedure usually occurs 1 year after limb salvage in a skeletally immature patient. An alternative leg length discrepancy can be corrected by skeletal shortening via femoral osteotomy and fixation. The longer contralateral "normal" extremity can be shortened surgically by 2–3 cm if necessary in a procedure that typically occurs 2–3 years after limb salvage and is best achieved as femoral shortening fixed with a femoral rod or plate [48–50]. Femoral osteotomy for the correction of leg length discrepancy usually heals at 6 weeks and has a low complication risk.

In cases for whom the discrepancy is projected to be greater than 2 cm and shortening of the contralateral limb is refused, distraction osteogenesis could be considered as an alternative to lengthen the short extremity. Bone lengthening is accomplished with an osteotomy and gradual lengthening through the use of an Ilizarov-style external fixator, which is difficult to implement after chemotherapy and requires 6–12 months of postoperative rehab. This procedure may be limited by the amount of remaining viable bone that is suitable for distraction osteogenesis and has limited applications in the pelvis and in sarcoma patients. The entire course of lengthening requires multiple operative procedures with significant risk of pin sepsis and in not an attractive alternative in patients who have just completed chemotherapy and/or radiation treatment for a sarcoma [51].

17.11 Future Directions

Future directions for the treatment of pediatric sarcoma patients include three promising areas for advancement: (1) improved assessment and documentation of surgical margins by intraoperative technology; (2) the development of more sophisticated modular, 3D-printed oncologic pelvic biologic implants; and (3) improved methods to preserve or reproduce skeletal growth after sarcoma treatment.

In the near future, improvements in the intraoperative assessment of surgical tumor margins will involve the ability to assess the adequacy of surgical margins with intraoperative transducers and biologic tumor imaging. Immunofluorescence imaging techniques or other intraoperative real-time assessment tools of critical tumor margins will facilitate that process. The development of quantitative and "co-registered" (overlapping) MRI and PET should also allow for the improved preoperative assessment of tumor viability and the improved intraoperative assessment of tumor margins. Current studies are investigating the role of intraoperative navigation systems in the planning and documentation of adequate surgical margins [22].

New oncologic implant 3D printing manufacturing processes should allow for the improved attachment of the gluteal and hip muscles and other tendon attachments at the greater trochanter, lesser trochanter, and hip capsule. As manufacturing technology continues to advance, the options for improved pediatric pelvic implants should allow for improved fixation, osseous healing, and physical function. Ideally, these new implants will allow for modularity, possible noninvasive lengthening, and improved function. An improved viable biologic alternative for pelvic iliac allografts and physeal acetabular cartilage may provide dramatic improvements for pediatric patients with viable pelvic grafts that will achieve bony healing in 3 months instead of the current 6–12 months. Improved pelvic grafts might include a collagen matrix incorporating osteoblastic stem cells to be used in conjunction with demineralized 3D-printed allografts to allow for earlier bony union of hemipelvic reconstructions. The current 12–24-month delay for bony healing of pelvic allografts is a mandate for better options and progress with both biologic and molecular mechanisms for cartilage and bone transplants in the pelvis and extremities.

Limb salvage for sarcoma patients gained popularity in the 1970s and 1980s with the success of chemotherapy regimens. Adult surgical resections have shown significant improvements in the last two decades in the function and survival of extremity oncologic implants. The focus is now on developing better reconstruction options for children with a better capacity for growth and improved methods for antibacterial therapies. Other challenges in pediatric oncologic limb salvage currently involve significant recent improvements in perioperative imaging and expectation for improved pediatric pelvic implants and more successful methods for skeletal growth. With advances in technology and biologic research, these improvements will likely be accomplished in the next 5–10 years and have significant consequences for the skeletally immature patients who have the greatest risks and rewards after tumor resection.

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18 Perioperative Management

Zhiye Du and Wei Guo

18.1 Preoperative Preparation

18.1.1 Routine Preparation

The operation time of patients with pelvic malignancies is long and commonly needs 4–6 h or longer, and the amount of blood transfusion often exceeds 1600 ml. It's of significance for patients to receive a full assessment of the general situation including the examinations of metabolic disease, cardiovascular diseases, and respiratory disease. Radiotherapy or other conservative treatment should be adopted for patients with poor surgical tolerance and high risk of receiving surgical intervention. After the improvement of general status, surgical intervention could be considered. Surgery without perfect preoperative preparation often causes high level of intra- and postoperative risk, recurrence of tumor, or even the death of patients.

Early stage of infection is commonly caused by bacteria or fungus from the intestinal system or urogenital system, and these infection factors must be dispelled preoperatively. The most significant preoperative preparation is bowel preparation including the use of enema and laxative. Deep infection of wound is inevitable once there are intestinal damages during operation without a full preoperative bowel preparation. Commonly enema and laxative are adopted. Patients take fluids and laxative 2 days and 24 h preoperatively, respectively, and accept cleaning enema the night before surgery. If patients have high probability to receive intestinal surgery during operation, the preoperative bowel preparation should be more regular. Total parenteral nutrition can be applied for patients several days preoperatively for the purpose of keeping intestinal clean and saving strength. Huge retroperitoneal tumors often invade retroperitoneal viscera such as the ureter, colon, and rectum. For most of these cases, urethra shunt, nephrectomy, and colectomy should be adopted before surgery. If the ureter is invaded by tumor and difficult to be separated, ureteral catheterization should be adopted for the purpose of avoiding accidental injury of the ureter. If there is a probability to perform colectomy combined with colostomy or urethra shunt during surgery, surgeons of general surgery and urology should be involved in the surgical planning together. Although not all cases need these treatment measures during surgery, teamwork is a key factor and prerequisite for the success of pelvic tumor surgery.

Surgery for patients with pelvic lesions commonly encounters long surgery time and needs a large amount of blood transfusion. The surgery time and volume of operative blood loss for patients with pelvic lesions commonly varied depending on the different experience of practiced surgeon, locations of tumors, and pathological types. Autologous blood transfusion can only be adopted for individual cases, while for most of cases, sufficient blood supply should be prepared for intraoperative and postoperative use. When anticipated blood loss volume exceeded 2000 ml, fresh frozen plasma should be prepared to avoid coagulation dysfunction (anticipated blood loss volume Table 18.1).

Table 18.1 Anticipated blood loss volume of surgery for pelvic tumor

Type of resection	Intraoperative blood loss (ml)	Postoperative blood loss (ml)
Traditional hindquarter amputation	800–3500	300–600
Internal hemipelvectomy	1500–5000	300–800
Region I resection	400–2000	200–300
Region II resection		
Prosthetic replacement	1500–3000	400–600
Arthrodesis	2000–4000	300–600
Region III resection	400–2000	100–300
Sacrectomy	1000–3000	400–800
Total sacrectomy	3000–6000	400–1000

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18.1.2 Technique of Vascular Occlusion

The pelvic ring of the human body is a complicated and irregular area full of crucial vessels and highly vascularized. Surgery for patients with tumors located in this area leaves surgeons a chief difficulty: a large amount of intraoperative blood loss. Because of the irregular and complicated anatomical structure around the pelvis and sacrum, surgery for huge tumors located in the pelvis and sacrum commonly causes a large amount of intraoperative blood loss which may even influence the implementation of the operation plan. What's more, a large amount of blood loss and blood transfusion can also influence internal environment of the human body which raises the related risk of postoperative complications. Thus, reducing the intraoperative blood loss is the key factor for the success of surgery.

18.1.2.1 Intraoperative Ligation of Ipsilateral Internal Iliac Artery and Temporal Occlusion of Abdominal Aorta or Ipsilateral Common Iliac Artery (Fig. 18.1)

Before the application of abdominal artery balloon occlusion, intraoperative ligation of ipsilateral internal iliac artery and temporal occlusion of abdominal aorta or ipsilateral common iliac artery are often adopted for complicated surgeries of sacral and pelvic tumors. The patients are placed in a lateral position on the contralateral side and the barley incision is adopted. Then the musculus obliquus externus abdominis, musculus obliquus internus abdominis, and musculus transversus abdominis are incised layer by layer to reveal ipsilateral common iliac vessels and ipsilateral iliac arteriovenous. Confirm and ligate the ipsilateral internal iliac artery, and ligate the contralateral internal iliac artery if necessary. Detach upstream to reveal abdominal aorta, and place a nail file slipping into a rubber hose at 1 cm above the bifurcation of the common iliac artery for the purpose of preparing to occlude the abdominal aorta. This method of controlling intraoperative bleeding is still adopted for those patients with high risks of receiving aortic balloon occlusion such as advanced-aged patients with hypertension or diabetes.

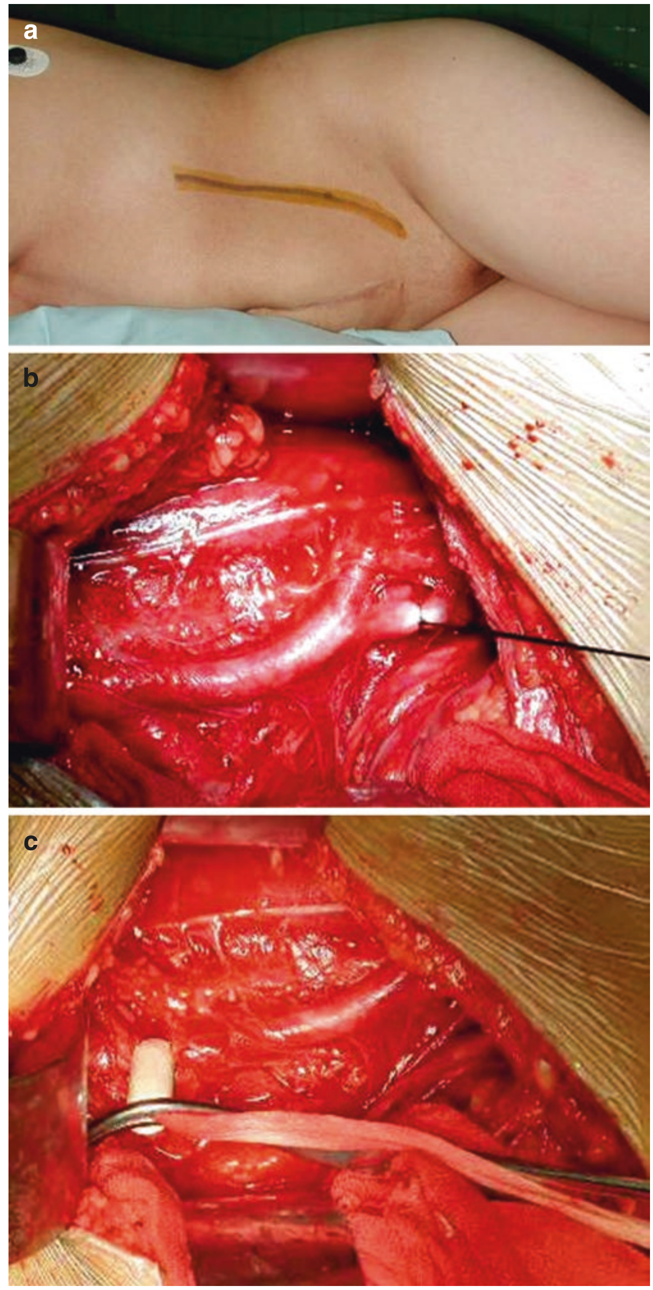

Fig. 18.1 (**a**) The patients are placed in a lateral position on the contralateral side and the barley incision is adopted. (**b**) Intraoperative ligation of ipsilateral internal iliac artery. (**c**) Temporal occlusion of abdominal aorta

18.1.2.2 Embolism Under Digital Subtraction Angiography (DSA)

With the ability of showing the blood supply of tumors definitely and occluding the blood supply by the means of embolism, DSA has gradually become a significant auxiliary means for pelvic surgery. Preoperative angiography and selective endovascular embolization under DSA as a common and important part of preoperative preparation of surgery for pelvic and sacral tumors can definitely show the blood supply within the extent of tumors and reduce the intraoperative blood loss.

The method of preoperative embolism for internal iliac artery is as follows: the catheter is inserted retrograde and craniad using the Seldinger technique via the percutaneous femoral arterial catheterization and then inserted into unilateral or bilateral internal iliac artery to investigate the location, character, extent, and blood supply of tumors after abdominal aortography. Embolize unilateral or bilateral internal iliac artery (commonly we choose the more severe side) and other target vessels using gelfoam particles or

Fig. 18.2 (**a**) The blood supply mainly comes from bilateral internal iliac arteries. (**b**) The blood supply decreases obviously after embolization of bilateral internal iliac arteries

stainless steel coils, and finally determine the embolization effect by abdominal aortography. Lateral circumflex femoral artery should be embolized by gelfoam particles for those cases tumors locate in front of acetabulum or ilium and semi-pelvic prosthesis replacement or hip replacement is considered. Retrograde the catheter back to the level of aorta abdominal or common iliac artery, and check the branches from the fourth and fifth lumbar arteries to the pelvis, and then embolize them selectively for those cases tumors locate in the rear of ilium. Strengthen packing of the puncture site after finishing the embolization, and carry out the surgery of the pelvis or sacrum within 24 h.

Commonly bilateral internal artery occlusion is applied before complicated pelvic surgery (Fig. 18.2). There are three disadvantages with this traditional method: the first is that patients may suffer from discomfort of lower abdomen such as acute abdominal pain, the second is that the hemostatic effect commonly is not that satisfying, and the third is that patients often suffer poor wound healing. All the phenomenons above are based on the anatomical structure below.

There are two groups of vascular networks within iliac blood vessels. One is vessel anastomosis between bilateral internal iliac arteries. This vascular network full of abundant anastomosis traffic branches containing the component from other vessels of enterocoelia and pelvic cavity is responsible for the blood supply of area around the pelvis especially gluteus maximus and lateral pelvic wall. The superior gluteal artery originates from the internal iliac artery. That's the reason why patients often suffer from discomfort of the lower abdomen and poor wound healing after bilateral internal iliac artery occlusion. Full occlusion of unilateral or even bilateral internal iliac artery is confirmed riskless for the reason that this vascular network contains numerous origins and anastomosis. Most of surgeons focus on the character of these vascular anastomosis branches and regard bilateral internal iliac artery occlusion as a routine method of iliac blood vessel embolism. The other important but undervalued vascular network is the vessel anastomosis between ipsilateral internal iliac artery and external iliac artery. This vessel anastomosis is mainly comprised of iliolumbar artery from the internal iliac artery and deep iliac circumflex artery from the external iliac artery and also has the component from lateral femoral circumflex artery and some branches from lumbar artery. This vessel anastomosis is responsible for the blood supply of pelvic medial wall and muscles around where the area is often invaded by tumors and operated on. Because of the existence of this vessel anastomosis, the hemostatic effect commonly is not that satisfying when bilateral internal iliac arteries are occluded. Thus, this vessel anastomosis is of great significance to the blood supply of pelvis.

18.1.2.3 Aortic Balloon Occlusion (ABO)

Blood volume loss during sacral tumor resection is mainly influenced by the location of the tumor, volume of the tumor, and tumor blood supply. Large pelvic tumors located anterior to the ilium or sacroiliac joint covering the surface of iliac vessels often hinder surgeons from ligating internal iliac artery or occluding the common iliac artery temporarily. In these situations, the ABO is necessary preoperatively for the surgical treatment of pelvic tumors.

Because of the existence of the second vessel anastomosis elaborated above, the hemostatic effect commonly is not that satisfying when unilateral or even bilateral internal iliac arteries are embolized. For those complicated cases with large sacral or pelvic tumors, preoperative aortic balloon occlusion is necessary. Commonly patients receive embolism the day before the surgery and receive preoperative aortic balloon implantation just before the surgery the next day. Sometimes embolism and surgery are planned to be implemented on the same day, and the aortic balloon is implanted just after the embolization. We should confirm repeatedly if obvious abnormity of segment between femoral artery and inferior abdominal aorta such as aortic aneurysm and aortic dissection exists. Severe artery diseases such as aortic aneurysm and aortic dissection are taken as contraindications for applying of ABO, while aortic plaque is acceptable for this operation.

The method of preoperative balloon dilation catheter implantation is as follows: the catheter is inserted retrograde and craniad using the Seldinger technique via the percutaneous femoral arterial catheterization and then inserted into the abdominal aorta. Confirm that there is no obvious abnormity of aorta that exists, and determine the location of bilateral renal artery and the bifurcation from the abdominal aorta to the common iliac artery. Commonly the bilateral renal artery is located on the level of L1, and the bifurcation is located on the level of L4–5. The balloon should be implanted between these two anatomical positions and commonly is located on the level of L3 (Fig. 18.3). Never prolong a single occlusion interval more than 90 min. When a longer occlusion period was needed, the balloon was deflated for 10–15 min after 90 min and then was reinflated. Before the operation, bilateral dorsalis pedis arteries should be labeled for the purpose of checking if femoral artery thrombus exists after the extraction of balloon and artery sheath.

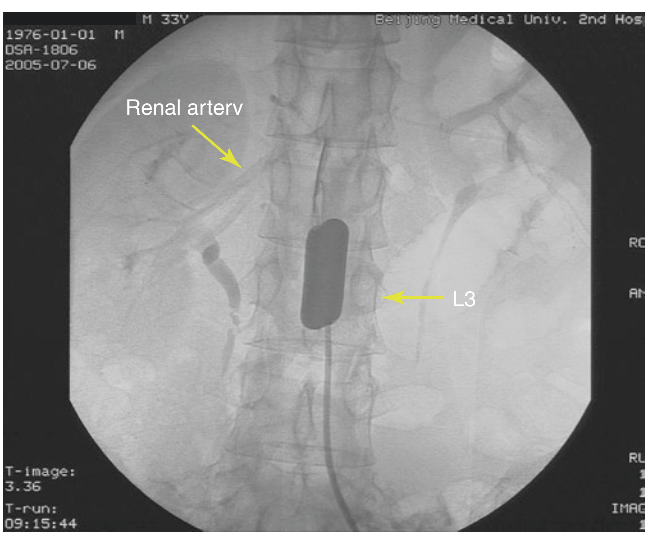

Fig. 18.3 Aortic balloon occlusion at the level of L3

Different levels of occlusion for the abdominal aorta bring different influences on systematic hemodynamic and visceral ischemia reperfusion injury. The low-level abdominal aorta occlusion applied for the surgery of sacral and pelvic tumors is a relative security. Implantation of the balloon at the level of the distal abdominal aorta and upstream of bifurcation of bilateral common iliac arteries doesn't occlude the blood supply for organs sensitive to warm ischemia such as the liver, kidney, and medulla spinalis. The feeding arteries of some lower abdominal organs sensitive to warm ischemia such as testis and ovary derive from abdominal aorta slightly below the renal arteries. Thus, the testicular arteries or ovarian arteries are excluded from the occluded list, and the blood supply for testis or ovary is unaffected by abdominal occlusion.

When patients return back to the ward or intensive care unit (ICU) after surgery, the volume of drainage should be recorded every single hour or even every half an hour to evaluate if active bleeding still exists. Active bleeding should be highly suspected if the drainage volume increases every single hour, the color of drainage presents bright red, heart rate increases even combined with decrease of blood pressure, and the hemoglobin decreases continuously. In these situations, the balloon and the arterial sheath should not be pulled out for the reason that the balloon can be reinflated to control the postoperative active bleeding and the arterial sheath can be used to implement postoperative embolization. Active bleeding is considered controlled if the postoperative drainage volume decreases every single hour gradually and the heart rate, blood pressure, and hemoglobin stay stabilized. The balloon and arterial sheath can be pulled out in this situation on the night of the operation day.

Do palpate the bilateral dorsalis pedis arteries after the extraction of the balloon and the artery sheath. Sometimes the dorsalis pedis arteries pulse weakly because of the femoral artery spasm. There is no evidence of lower limb ischemia if the oxygen saturation numerical value and waves of the second toe can be detected by pulse oximeter and if the lower limb has no dysfunction of motor and sensory. It is highly suspected of femoral artery thrombus if the ipsilateral dorsalis pedis artery is impalpable combined with no oxygen saturation detected and the lower limb gradually presents the symptoms of 5P syndrome: pain, paresthesia, paralysis, pulselessness, and pallor. In this situation, a lower limb arterial ultrasonic Doppler data has to be done, and an emergency operation of femoral arterial embolectomy should be carried out if the femoral arterial thrombus is detected by the lower limb arterial ultrasonic Doppler. Patients should receive parcel oppressed stanch at the puncture site for 8 h, and the ipsilateral lower limb should be kept immobilized for 24 h.

18.2 Intraoperative Monitoring

The operation time for pelvic or sacral tumors always lasts long, and patients should be placed in a comfortable surgical position to avoid neural impairment. Body regions like the head, neck, and joints should be protected, and a cushion should be placed under the axilla when patients are placed in a lateral position.

Before surgery, multiple venous access should be established to facilitate intraoperative blood transfusion and fluid infusion. Central venous and radial artery catheter should be applied to monitor intraoperative circulatory system well-timed. Both surgeons and anesthetist should pay close attention to the volume of intraoperative blood loss and perform blood transfusion without delay because the real volume of intraoperative blood loss is often underestimated. Timely blood transfusion is beneficial in avoiding coagulation dysfunction and circulatory failure.

Intraoperative controlled hypotension is reported to decrease the volume of blood loss by 50% and be more effective than intraoperative hemodilution. Nitrates such as sodium nitroprusside and nitroglycerin are the most commonly used antihypertensive drugs. Another method to control intraoperative blood loss is hemodilution including preoperative hemodilution (isovolemic hemodilution) and hypervolemic hemodilution. Isovolemic hemodilution means letting blood from arteries or veins and transfusing equivoluminal colloid solution and/or crystalloid solution at the same time after the anesthetic with the purpose of reducing hematocrit (Hct) rather than blood volume. The blood drew out is transfused back after the massive hemorrhage is controlled. Hypervolemic hemodilution means alleviating the central venous pressure (CVP) to the high limit of normal value (10–12 cm H_2O) by transfusing crystalloid solution and colloid solution (1:1) after the anesthetic to expand the volume of endovascular and extracellular fluid. The expansion of endovascular and extracellular fluid leads to hemodilution (Hct $\geq$ 30%) which decreases the loss of blood cell components and strengthens the ability of defending against hemorrhagic shock when massive hemorrhage is encountered.

Intraoperative monitoring of the abdominal aortic balloon catheter: commonly we use arterial piezometer for dynamic monitoring of the whole course. An ambulatory blood pressure monitoring device is connected to the catheter after successful puncturing. We can indirectly keep track of the situation and parameters of lower limb arteries by comparing the parameters to that of radial arteries. The arterial waveform breaking to a straight line and arterial number reducing to a low fixed numerical value mean the blood supply for the main arteries of lower limbs is excluded and the operation can be performed under the occlusion. Surgeons can master the effect of occlusion by intermittent observation of the numerical value. Intermittent trace of heparin saline pumped through the catheter can effectively avoid the formation of femoral thrombosis.

18.3 Postoperative Management

The removal of huge tumors by surgery of sacral or pelvic tumors always leaves huge dead spaces, and the management for the dead spaces is of significant. The dead spaces are packed with blood clot in the initial phase after the surgery, and secondary infection is common due to hematoma formation and incomplete drainage. Infection is a disaster for surgery with artificial medical implantations. Commonly we use unrevealed gauzes to oppress the dead spaces outside of the wounds and lace the patients with elastic corset. Patients with sacral tumors are placed to a horizontal position to oppress the dead spaces by weight. After the active bleeding is controlled, small quantities of negative pressure drainage should be applied if the dead spaces exist or definite effusions are found sealed in the wounds.

The third-generation cephalosporin is commonly applied until the drainage tube is pulled out without signs of infection. The drainage tube should be retained until the drainage volume is less than 50 ml/24 h. Bacterial culture of the terminal of the drainage tube should be routinely performed when the drainage tube is pulled out for the purpose of selecting sensitive antibiotic once some bacterial is detected. Blood tests such as blood routine, ESR, and CRP should be tested every 2 or 3 days, and the trend of WBC, NEU%, ESR, and CRP should be observed closely. The antibiotic should be upgraded if the numerical values above keep a high level combined with high temperature.

Patients who received hindquarter amputation are encouraged to be ambulant as soon as possible, and commonly within 1 week is recommended. Young patients are able to walk with the assistance of crutches, while aged patients commonly walk with mobility aid after they can stand steady supported by others. It is not necessary for most of aged patients to be fitted with an artificial limb, while young patients are encouraged to use it. Postoperative management for patients who received region I/III resection is roughly the same as that of hindquarter amputation because there is no pelvic reconstruction for these patients. Reconstruction for pelvic integrity and function of hip joint is always required for patients who received region II resection, and optional methods include saddle prosthesis, modular hemipelvic prosthesis, and arthrodesis. Periacetabular soft tissue reconstruction is essential for patients who received modular hemipelvic prosthesis, and it is necessary for these patients to lie in bed for at least 6 weeks to increase the stability of hip joint. Patients who are treated with bone graft should lie in bed for at least 8 weeks. Patients are able to practice standing after gradual stabilization of hip joint and gradually increase the bearing weight on the affected extremity. Periacetabular soft tissue scarring 3 months after surgery may enhance the stability and prevent dislocation of hip joint.

19 Complication Prevention and Management

Dasen Li and Wei Guo

Detailed resection margins, including bone and soft tissue margins, and strategies aimed at reducing blood loss, as well as bone and joint reconstruction methods, are key points that should be considered during preoperative planning. However, pelvic ring tumor surgeries are often accompanied by a high rate of operative complications, such as hemorrhagic shock, deep vein thrombosis (DVT), pelvic viscera injury, nerve injury, and blood vessel injury. Functional reconstruction remains difficult in pelvic surgery and is related to some postoperative complications of this type of surgery, such as artificial hip dislocation, loosening of the hemipelvic endoprosthesis, hemipelvic endoprosthesis breakage, and postoperative neuralgia. Strategies aimed at preventing and managing these possible complications will be discussed in this chapter. Wound complications, including surgical site infection (SSI) and wound dehiscence, will be discussed in the chapter entitled "Complications After Sacrectomy and Pelvic Tumor Surgery."

19.1 Intraoperative and Postoperative Bleeding and Hemorrhagic Shock

19.1.1 Causes of Hemorrhage During Resection of Pelvic Ring Tumor

Resection of a pelvic tumor is a time-consuming procedure because of the complexity of the regional anatomy. Abundant blood supply, pathological diagnoses, tumor volume, the anatomic location of the mass, and resection margin are factors that affect blood loss.

The internal iliac artery supplies most of the blood to the pelvis, and its various branches arise in a variable manner. The parietal branches of the internal iliac artery include the iliolumbar, lateral sacral, obturator, superior and inferior gluteal, and internal pudendal arteries. The visceral branches include the umbilical artery, the superior and inferior vesical arteries, the uterine artery (or the artery of the ductus deferens), and the vaginal and middle rectal arteries. All of these arteries and their corresponding veins form a network with abundant blood flow. Moreover, these vessels can be stimulated by malignant tumors or local aggressive tumors, forming a vascular plexus with a large diameter. In the case of tumors such as giant cell tumor of the bone and alveolar soft part sarcoma, these newly formed vessel plexuses may be numerous and can lead to massive blood loss during dissection.

It is not easy to control the bleeding that occurs during the intralesional resection of a tumor. In some of our cases of palliative external hemipelvectomy, the blood loss was greater than 8000 mL during tumor resection. The achievement of a wide or marginal resection margin may lead to a reduction in blood loss.

19.1.2 Approaches to Reduce Blood Loss

Temporary occlusion of the common iliac artery or abdominal aorta using atraumatic forceps or a string is an effective way to control bleeding in most occasions. This procedure can be performed before tumor resection. When necessary, the ipsilateral internal iliac artery can be ligated. To avoid venous congestion and extensive oozing of the blood, the internal iliac vein should not be ligated.

If the common iliac artery or abdominal aorta cannot be exposed because of the large size of the tumor mass or severe adhesion caused by former operations, the use of an intra-abdominal aortic balloon is a good option [1]. This method should also be considered when the sacrum is affected by huge pelvic tumors or when palliative external hemipelvectomy is planned.

For hypervascular tumors, such as giant cell tumor of the bone or metastatic lesions of renal cancer, hepatic cancer, or thyroid cancer, preoperative angiography and

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interventional embolization can reduce blood loss effectively. A preoperative enhanced CT scan of the pelvic region can provide an accurate assessment of tumor blood supply. The interventionalist's role in managing intraoperative hemorrhage has been promulgated in the surgery, tumor, and radiology literature [2, 3]. Successful treatment by embolization will prevent a substantial amount of blood loss and provide better exposure and, thus, a better resection margin. When considering embolization, it is important to first document adequate collateral flow to the pelvic viscera.

The posterior peritoneum has a very limited ability to confine hematomas after pelvic tumor resection. Adequate hemostasis and adequate drainage may prevent the formation of large hematomas. To manage blood oozing around the bladder, posterior urethra, and prostate in males, simple suture ligatures or suturing for hemostasis is required. In rare cases, postoperative active bleeding can be managed successfully by embolization.

19.2 Deep Vein Thrombosis (DVT)

19.2.1 Causes of the High Incidence of Perioperative DVT in Pelvic Tumors

Anticoagulant and fibrinolytic systems can lead to thrombosis, as observed in conventional hip replacements. The incidence of perioperative DVT in pelvic tumors is comparable to, or even higher than, that of total hip replacement. Tumor masses with a very large size may compress the iliac veins directly. In fact, some patients with pelvic tumors even have DVT of the lower limbs at diagnosis. Malignant tumors and longer immobilization times are also risk factors for this condition. In the perioperative period, especially during tumor dissection, blood clots that are present in veins can break loose, travel through the bloodstream, and lodge in the lungs, thus blocking blood flow (pulmonary embolism). Direct venous damage during operation, including blood vessel stretching, branch ligation of the iliac vein, and repair of veins, may induce DVT.

19.2.2 Prevention and Treatment of DVT

Detailed history collection and physical examination can help identify the risk factors of DVT (smoking, taking birth control pills, being overweight or obese, history of inflammatory bowel disease, and a personal and family history of DVT or pulmonary embolism) preoperatively [4]. Once DVT is identified preoperatively, a filter should be inserted into the vena cava, which prevents the lodging of clots that break loose in the lungs [5].

During operation, the iliac vein should be gently dissected and protected. On some occasions, clots in the iliac vein system can be removed carefully.

Massage the gastrocnemius and soleus postoperatively. Encourage the patient to perform active ankle joint activities periodically, when possible, and to take deep breaths and cough, to increase diaphragm movement. An intermittent pneumatic compression pump should be used, unless DVT has occurred.

Compression stockings can restrict the veins of the lower limb and prevent venostasis. The contraindications of these stockings include serious leg edema, pulmonary edema, severe peripheral arterial sclerosis occlusion disorder, and severe leg deformity or dermatitis.

The anticoagulant prophylaxis of DVT is different from that of a conventional hip surgery. To date, there are no clear guidelines regarding this issue. Risk factors, such as massive blood loss, excessive consumption of clotting substrates, and extensive wound errhysis, must be carefully considered before the administration of anticoagulants. Anticoagulant therapy leads to an increased risk of postoperative wound bleeding and local hematoma formation. This can cause fatal hemorrhagic shock and/or wound complications, such as delayed healing, necrosis, surgical site infection, etc. In selected cases, the administration of an individualized dose of an anticoagulant could be considered 12 h postoperatively. Close attention must be paid to vital signs, abdominal tension, and the characteristics and amount of drainage during the course of anticoagulation.

For patients with perioperative DVT, an inferior vena cava filter can effectively prevent the occurrence of fatal pulmonary embolism. However, this procedure is associated with several complications, which include early and long-term complications. Hemorrhage and hematoma at the puncture site, pulmonary embolism, and filter release failure are common early complications. Long-term complications include filter offset, wandering, angulation, and perforation; ascending colon perforation; thrombosis in the filter; etc. Therefore, inferior vena cava filter implantation requires strict indications. Lower-extremity DVT must be diagnosed, and pulmonary embolism should be detected. Preoperative examination includes vascular ultrasound Doppler, CT angiography, and vein angiographic examination, with vein angiography having the highest accuracy. However, superficial vein puncture and anterograde contrast are often difficult to achieve. Once a diagnosis of DVT is established, the vascular surgeon determines whether an inferior vena cava filter should be placed. A permanent inferior vena cava filter should be placed in cases of a floating thrombus in the iliac, femoral, and popliteal veins or of large iliac–femoral venous thrombosis. The placement of a permanent inferior vena cava filter requires prolonged anticoagulation therapy and outpatient follow-up. A temporary inferior vena cava filter might be considered in young patients.

19.3 Complications Related to Pelvic Viscera

A huge pelvic tumor could grow into the pelvic cavity. The iliac muscle would confine the tumor, and the pelvic viscera would be pushed away in most cases. However, in some cases, the pelvic organs, including the ureter, bladder, prostate, colon, and rectum, as well as the uterus and vagina (in female patients), may be affected by the tumor. To obtain a satisfactory tumor resection margin, a part or all of the invaded organs have to be removed with the tumor. Thus, a multidisciplinary team is needed. Although modern endoscopy and imaging techniques, such as high-resolution CT or MRI, can provide extremely valuable information regarding pelvic organ involvement during preoperative planning, the real situation of pelvic organ invasion can only be determined by surgical exploration.

19.3.1 Bladder Injury

The causes of bladder injury include direct pelvic tumor invasion, as well as the formation of scars and adhesions caused by operation or radiotherapy. The general principles of management of this condition include placement of indwelling catheters, adequate drainage, and bladder repair, as well as adequate drainage around the repaired bladder and/or other urinary extravasation area.

An accidentally torn or cut bladder can be repaired with a 3-0 or 2-0 absorbable stitching line in two layers. If such damage area is situated around the trigone, ureteral stents should be placed at the same time.

If the bladder injury is mild and not found during operation, its symptoms are light, and postoperative cystography finds only a small amount of urinary extravasation, nonoperative treatment, such as continuous urinary catheterization for more than 14 days, could be adopted. Usually, the injured bladder wall can heal. If bladder urine extravasation is serious, open surgery or laparoscopic surgery should be performed as soon as possible. In cases of bladder neck tearing, this injury must be repaired with a 3-0 absorbable thread accurately, to avoid postoperative urinary incontinence. When the injury site is difficult to find, a cystoscope could be used. When necessary, a methylene blue solution should be injected through the urinary catheter, to indicate the site of bladder leakage. After exploration and bladder repair, sufficient drainage around the bladder is mandatory. Systemic antibiotics should be used and suprapubic cystostomy could be considered.

Once a bladder–vagina fistula has formed, repair measures depend on the location and size of the fistula, the condition of the surrounding tissue, and the experience of the urologists and gynecologists involved in the procedure. If the fistula hole is located at a low position, the hole is small, and there is no (or just slight) inflammatory edema of the surrounding tissue, the bladder–vagina fistula can be repaired through the vagina. When the fistula hole is located at a high position and the hole is big, open surgery or laparoscopic surgery should be performed. Special attention should be paid to the relation between the fistula hole and the ureter openings. Normal bladder mucosa and muscularis and the normal tissues surrounding the fistula hole should be separated, and all inflammatory tissue around the hole should be removed before repairing the injury layer by layer using absorbable sutures. Subsequently, the absence of leakage is ensured using a bladder filling experiment. Systemic antibiotics, avoidance of activities that increase abdominal pressure, and prohibition of sexual activity for half a year (together with the administration of estrogen agents, which will promote mucosa healing) should be implemented. In fact, although the probability of having a bladder–vagina fistula is small, once it appears, it is difficult to repair because of inflammatory edema and other adverse factors [6].

19.3.2 Ureteral Injury

Common causes of ureteral injury include ureter displacement, tumor infiltration, local adhesions caused by previous surgery, local tissue inflammation and fibrosis after radiotherapy, the use of clamps and suture blindly during intraoperative bleeding, ureteral ischemic necrosis after large segment dissection, direct damage caused by electrohemostasis, and ureteral repetitive malformation. On rare occasions, the ureteral nerve, which is responsible for ureteral peristalsis, is injured during surgical dissection. Impairment of ureteral peristalsis may lead to expansion of the tube cavity, increase in internal pressure, and ureteral ischemic leak. The types of ureteral injuries include ligation, crush injury, transection (partial or complete), and ischemic injury. It is advisable to place ureteral catheters preoperatively when a difficult operation is anticipated. This procedure will facilitate the identification of the ureter during operation.

Ureteral injuries found during operation can be repaired immediately, because the tissue has no edema or adhesion at this time; thus, repair surgery results in good healing. Visual inspection is the reliable means of assessing ureteral integrity during operation. When necessary, methylene blue can be injected intravenously. When recognition of the problem is delayed, retrograde pyelography is the most sensitive tool for identifying ureteral extravasation or obstruction from ligation. Ureteral injury found within 72 h postoperatively should also be repaired immediately, whereas delayed repair is recommended for injuries found 72 h or even several weeks later, because in these cases the urinary extravasation has caused local tissue congestion, edema, and inflammation. Immediate proximal urinary diversion, such as percutaneous nephrostomy, should be performed in most cases. It is also reported that ureteral injury can be repaired within 2–3 weeks postoperatively [7].

If the ureteral injury is a contusion or the cleft is very small, and the integrity of the ureter is not damaged, retrograde ureteral catheterization via cystoscope is preferred. If the ureter is ligated, remove the ligature. If the ureter is completely transected or almost transected, perform ureteroureterostomy using 4-0 or 5-0 absorbable sutures. In all of these procedures, the retroperitoneal area must be adequately drained, and the average retention time of a double-J tube is 4 weeks. In cases of resection of a large segment of the ureter, ureterocystostomy, anastomosis of the ureter, and bladder flap or reconstruction of the ureter using the ileum should be considered. If long-time obstruction of the ureter resulted in complete function loss of the ipsilateral kidney, and the contralateral kidney has normal function, the ipsilateral kidney can be removed.

19.3.3 Uterine, Ovarian, and Vaginal Injury

When the vagina is affected by the tumor, preoperative vaginal preparation must be performed according to the requirements of gynecologists. If part of the vaginal wall is resected, the necessity for reconstruction and the individual reconstruction plan depends on the patient's age, menstruation situation, etc.

19.3.4 Resection of Parietal Peritoneum

If the pelvic tumor adheres to the parietal peritoneum, the latter could be resected with the tumor. The peritoneal cavity should be closed.

19.4 Nerve Injury

If the obturator nerve is resected with the pelvic tumor, postoperative function loss is not obvious. Conversely, if the femoral nerve is resected with the tumor, postoperative quadriceps dysfunction is obvious.

When the greater sciatic foramen is affected by the pelvic tumor, peroneal and/or tibial nerve dysfunction is common, because the sciatic nerve and/or its components may be injured or transected during operation. It is important to note the relationship between the sciatic nerve and the piriformis muscle. In about two-thirds of the patients, the sciatic nerve runs through the infra piriformis foramen as one trunk. In a small proportion of patients, the sciatic nerve is divided into two bundles in the pelvis; one bundle finally becomes the tibial nerve and runs through the infra piriformis foramen, while the other bundle becomes the common peroneal nerve and runs through the piriformis itself. Other anatomic variations also exist. Overlooking these variations can lead to unexpected sciatic nerve injury.

19.5 Injury of Blood Vessels

In most cases, external hemipelvectomy is the first choice when the external iliac blood vessel bundle and the superficial femoral blood vessel bundle are affected by the tumor. However, internal hemipelvectomy could also be considered in carefully selected cases. The blood vessels involved would be resected with the tumor en bloc, followed by blood vessel reconstruction by vascular surgeons. Thrombosis may occur after autologous blood vessel transplantation or artificial blood vessel replacement. Arterial thrombosis may lead to limb necrosis, and it must be intervened immediately. Venous thrombosis is common; in most cases, it can be compensated by superficial vein reflux.

19.6 Complications Related with Modular Pelvic Prosthesis

19.6.1 Early-Stage Hip Dislocation After Hemipelvic Endoprosthesis

The resection of a pelvic tumor involving the acetabulum and the performance of reconstruction using a modular hemipelvic endoprosthesis are challenging. After radical tumor resection, there is extensive bony loss, including that of the acetabulum and the surrounding ipsilateral pelvic bone. The lack of important bony anatomical landmarks severely hampers the accurate positioning of the artificial acetabulum. The hip joint capsule and the gluteus, iliopsoas, and other muscles that surround the hip are often resected widely together with the tumor, which will inevitably lead to a decrease in muscle strength around the reconstructed hip joint. In some cases, the gluteal nerves are injured or transected, and even if most muscles are preserved, including adequate gluteal muscles, muscle strength around the hip could still be insufficient. Improper positioning of the artificial acetabulum and insufficient muscle strength are causes of hip dislocation after hemipelvic endoprosthesis reconstruction.

Early-stage dislocation is defined as a dislocation that occurs within 6 months postoperatively. Sudden hip pain after a postural change is a common complaint in this setting. External rotation and shortening deformities of the ipsilateral leg can be identified by physical examination. Hip dislocation can be confirmed by pelvic X-ray examination. Closed reduction under subarachnoid anesthesia or general anesthesia should be performed once hip dislocation is confirmed. To avoid the sciatic nerve injury and the loosening of the whole artificial pelvic endoprosthesis that are caused by violent reduction, the dislocated hip can only be gently reduced under X-ray fluoroscopy, with several repetitions. If this procedure is unsuccessful, open reduction should be performed. Place the patient in a lateral decubitus position, and expose the polyethylene acetabular cup through a 4 cm incision. Clean the soft tissue embedded in the acetabulum, and reduce the dislocated artificial hip joint accurately.

Intraoperative preventive measures include accurate positioning of the artificial acetabulum, selection of an artificial hip joint with anti-dislocation mechanisms, suturing the residual hip capsule to the artificial acetabulum with an artificial tendon, and restoring the muscle tension around the hip joint.

Placement of the affected lower limb in a rotationally neutral and 15–25° abduction position is the key principle of the postoperative preventive measures for the management of this condition. Body position changes must be directed and trained by professional nurses. Rehabilitation exercises, such as the isometric contraction exercise of quadriceps, starts on the first postoperative day. Patients are restricted to bed rest (confined to their beds) for at least 6 weeks, to allow the soft tissue to heal. Subsequently, the patients may begin to practice standing and walking with a walking aid.

One hundred and sixty-nine patients had pelvic tumor resection and hemipelvic endoprosthesis reconstruction between July 2003 and December 2011 [8]. Fourteen patients suffered from early-stage dislocation. The median time between dislocation and operation was 16 days (range, 0–94 days). Successful closed reduction was accomplished in five patients, whereas the remaining nine patients underwent an open reduction. No hemipelvic prosthesis loosening or sciatic nerve injury was observed during reduction. A second hip dislocation happened in two cases after closed reduction and unilateral hip spica braces were used. No artificial acetabulum had an acetabulum abduction angle <30° in our study. Five patients in the dislocated hip group and 50 patients in the non-dislocation group had an acetabulum abduction angle >55° (Fisher's exact test, $P = 0.773$) (Fig. 19.1).

19.6.2 Loosening of the Hemipelvic Endoprosthesis

In cases in which the pelvic region I can be kept after tumor resection, the modular hemipelvic endoprosthesis can be firmly fixed immediately to the residual ilium and sacrum using screws. The long-term stability of the reconstruction had been questioned because of the stress fatigue of the metal components. However, among the 100 patients who had modular hemipelvic endoprostheses after pelvic tumor resection between June 2001 and March 2010, endoprosthesis loosening occurred in only two patients [8]. Scar formation and bone reconstruction under stress may play an important role in the long-term stability of the endoprosthesis. In fact, the implant had to be removed in six patients because of deep infection; all six endoprostheses had been fixed to the residual pelvis tightly, and the implant-removal process was strenuous.

When the pelvic region I is affected by and resected with the periacetabular tumor, a modular hemipelvic endoprosthesis has to be fixed to the lumbar vertebrae and sacrum (pedicle–hemipelvic endoprosthesis, Fig. 19.2) [9]. The shear stress between the endoprosthesis and the bone surface often leads to reconstruction failure. After numerous improvements, this problem has now been resolved by the new type of lumbar vertebrae–sacrum–hemipelvic endoprosthesis system designed by Dr. Wei Guo (Fig. 19.3).

19.6.3 Pelvic–Hemipelvic Endoprosthesis Breakage

Among the 100 patients mentioned above, three patients had pubic bone plate fracture, and two patients had iliac screw fracture. The broken pubic bone plates had to be removed because of constant pain. The remaining three patients had no obvious symptoms and were followed up. Although stress experiments and finite element analysis confirmed that the pubic bone plate can help strengthen early stability, it is no longer used in our current implant system because of its high frequency of breakage (Fig. 19.4).

19.7 Postoperative Neuralgia

When the pelvic region II + I + IV is resected, a complicated reconstruction that includes a vertebral pedicle screw–rod system and a modular pelvic endoprosthesis (pedicle–hemipelvic endoprostheses) can be performed. This implant complex may compress or irritate L4 and L5 nerve roots and other components of the sciatic and femoral nerves and cause postoperative neuralgia. Preventive measures include avoiding the entrapment or traction of the

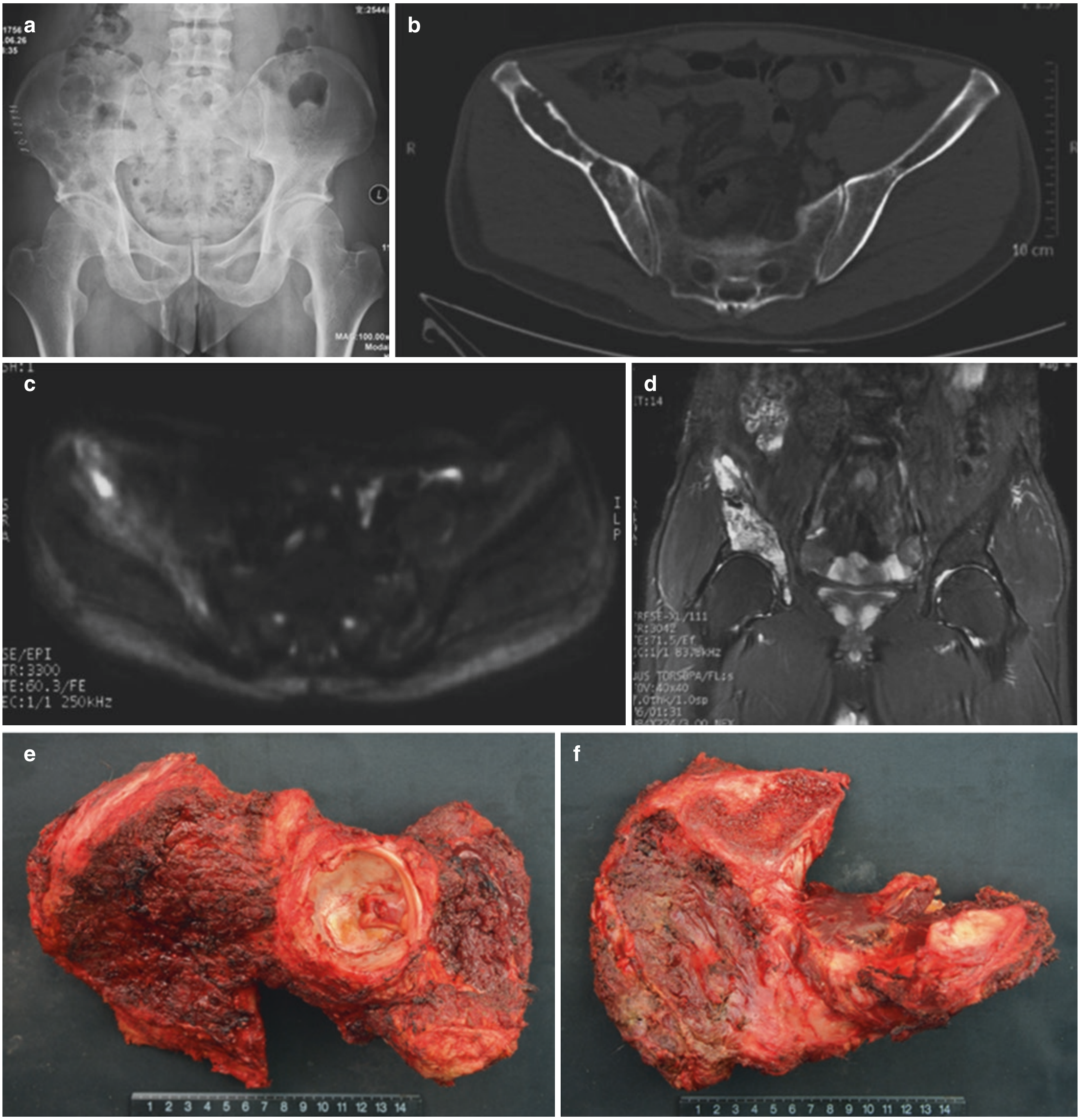

Fig. 19.1 A 49-year-old male with a pelvic chondrosarcoma. (**a**–**d**) A large pelvic lesion can be seen. Wide resection of the tumor and hemipelvic endoprosthesis reconstruction were performed. (**e**, **f**) En bloc resection of the tumor and peripheral muscles. (**g**) Hip dislocation occurred 2 days after operation. (**h**) Closed reduction was successful

above nerves during reconstruction and avoiding direct contact between the nerve and the metal components. In some cases, the implant system cannot be fixed rigidly, because of the large size of the bone defect, and a small displacement of the implant during changes in body posture may lead to neuralgia [10]. The new type of lumbar vertebrae–sacrum–hemipelvic endoprosthesis system designed by Dr. Wei Guo can be fixed rigidly to the residual pelvic ring and lumbar vertebrae and achieve favorable functional outcomes. However, from the point of view of surgical skills, this procedure remains challenging, because the screws that are driven into the sacrum may compress the sacral nerves or even the dural sac, resulting in persistent and fierce pain.

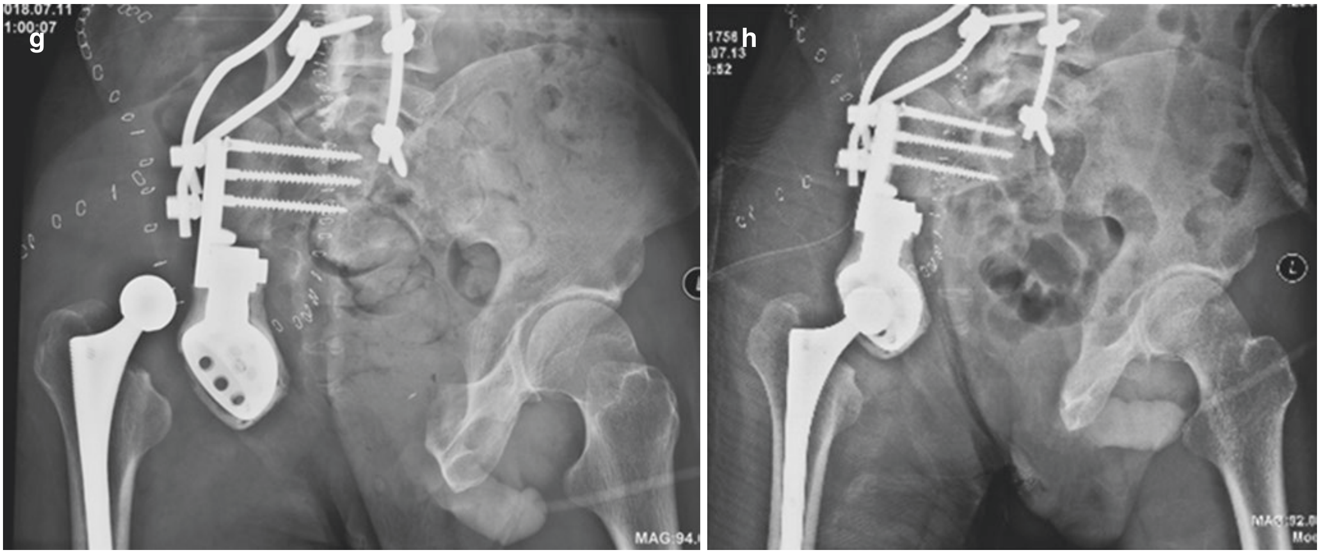

Fig. 19.1 (continued)

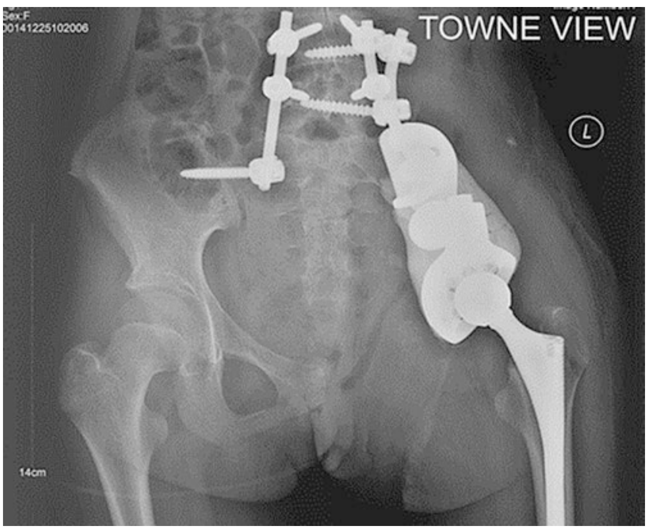

Fig. 19.2 Pedicle–hemipelvic endoprosthesis

Fig. 19.3 Lumbar vertebrae–sacrum–hemipelvic endoprosthesis

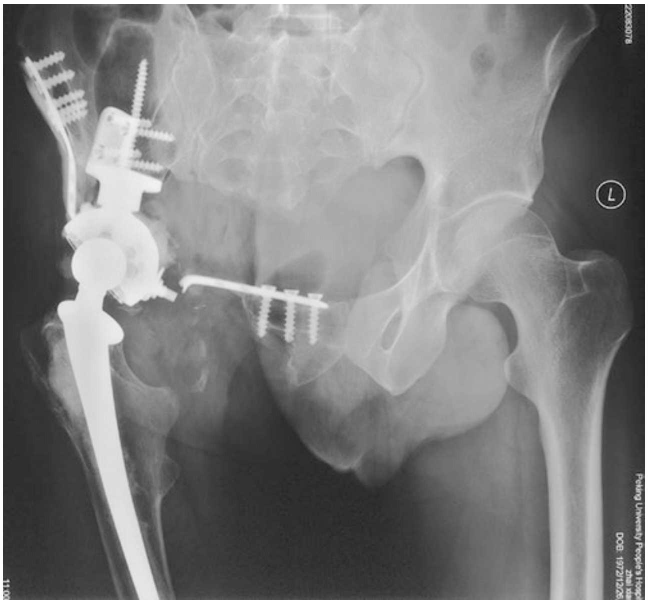

Fig. 19.4 Modular pelvic endoprosthesis and broken pubic bone plate

Acknowledgment *Source of All the Clinical Images*: All the clinical images were from the database of our center.

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Part V

Sacral Tumors: The Fundamentals

20 General Considerations

Yifei Wang and Wei Guo

Sacral tumors are rare. To diagnose a sacral tumor is challenging because of the lack of specific clinical symptoms, so sacral tumors are often diagnosed in advanced stages with extensively involving the sacral nerves, iliac vessels, and other surrounding structures. The management could be quite challenging for orthopedic surgeons because of the complicated anatomy. For most cases, only aggressive procedure with adequate surgical margin (en bloc resection) could guarantee satisfied local control, but in the meantime, sacrectomy also brings several problems such as bowel, bladder, and sexual dysfunction; wound infection; and major blood loss during the surgery or postoperatively which might jeopardize the safety of the surgery, the postoperational function [1], and prognosis of the patients.

Primary benign and malignant tumors of the sacrum are 2–4% of all primary bone neoplasms and 1–7% of all primary spinal tumors [2]. Sacral tumors can be classified into four main categories: congenital, metastatic, primary osseous, and primary neurogenic. Congenital lesions of the sacrum include dermoid cysts, anterior and intrasacral meningoceles, perineural cysts, teratomas, hamartomas, and chordomas. Of these neoplasms, chordomas are the most common primary sacral tumors that account for 40% of all primary sacral neoplasms in the USA [3]. According to a research from Peking University People's Hospital, chordomas account for 24.4% of all primary sacral tumor cases; they are also the most common primary sacral neoplasms in the Chinese population [4]. Chondrosarcoma is the second most frequent primary malignant bone tumor of the sacrum in the USA, though its sacral location is less than 7% of all cases [5]. Giant cell tumor (GCT) is the most common benign sacral neoplasm and accounts for 8–18% of all primary sacral tumors [3]. Neurogenic tumors (benign or malignant) are also frequently seen in the sacral tumor cases which represent up to 16.6% of all primary cases [4]. Rarer tumor types such as multiple myeloma, teratoma, Ewing sarcoma, osteosarcoma, lymphoma, hemangioma, and angiosarcoma could also be seen in the sacrum.

More than half of all sacral tumors are metastatic tumors. These lesions are most often disseminated from a solid organ. Lung, breast, prostate, thyroid, renal, and rectal cancers are the most common origins of sacral metastases [6]. Aggressive rectal carcinomas can directly invade the sacrum, increasing the complexity of surgical resection.

As it is mentioned above, the surgical treatment of these tumors is challenging because of the complex regional anatomy and the advanced stage of cancer at the time of diagnosis. Surgeons must not only be familiar with local anatomy from a neurologic, colorectal, urologic, gynecological, orthopedic, and plastic standpoint but also sometimes have to face the dilemma between functional preservation and cure of the tumor. The operating strategy requires precise preoperative planning to locate the exact extent of tissue involvement to decide the level of the osteotomy and the muscle, the nerve, and vessels that will require resection; to plan reconstruction method; and to determine if adjuvant treatments as preoperative embolization and radiation (preoperative or intraoperative) are needed. However, this chapter focuses only on the general information of sacral tumors, as the anatomy, surgical approaches to the sacrum, and some other information are discussed more extensively elsewhere in this textbook.

20.1 Clinical Presentation

Sacral tumors are very rare and usually grow insidiously with nonspecific symptoms such as lower back, buttock, sacrococcygeal, or referred leg pain [7]. Routine X-ray, CT, and MRI studies often fail to detect the sacrum neoplasm especially that of the lower sacrum. Unfortunately, many patients are misdiagnosed with lumbar disc disease for which they receive subsequent treatment. Norstrom et al. reported the mean delay from symptom onset to diagnosis is 2 years

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[8]. This delay often allows the tumors to grow larger and cause neurological dysfunction and/or mechanical instability.

The first presentation may be a painless sacral mass; sometimes the giant mass could even be palpable on abdominal examination. Some patients may only present with minor neurologic symptoms, with or without pain. The large presacral mass often causes constipation because of rectal compression, as well as impedes bladder function.

The pain caused by sacral tumors is the most common symptom. The location of pain usually indicates the origin of the tumors. For example, chordomas from lower sacrum could cause continuous rectal-anal pain [9]. Invasion of bone such as the sacroiliac joints can cause pain when sitting or walking and could be alleviated by lying supine. This usually suggests the instability of the joint and need to be reconstructed in the operation.

When the tumor invades the neurological structures, patients may present with radiated pain, numbness, paresthesias, or muscle weakness. On neurologic examination, the patients might have decreased reflexes, anal sphincter dysfunction, and lower extremity weakness. Bowel and bladder dysfunction can also occur in such scenarios. Some patients could present with cauda equina syndrome when the cauda equina is compressed by tumor, and these cases often need immediate intervention. General signs of cancer such as weight loss, anemia, or weakness are signs of metastatic cancer rather than primary sacral tumors.

20.2 General Information of Common Primary Sacral Tumor Types

20.2.1 Giant Cell Tumor

Giant cell tumors are the most common benign primary tumors of the sacrum and usually occur with a peak incidence in the third decade of life, with a female predilection [3]. GCTs are typically eccentric, expansile, osteolytic lesions without sclerotic margin or calcification. The lesions usually arise in the upper sacrum and often invade sacroiliac joints and intervertebral disks [10, 11], which is rare for other benign sacral tumors. Giant cell tumors (GCTs) are locally invasive and highly vascularized [12]; curettage surgery with no intraoperative hemorrhage control usually leads to local failure and major blood loss during the surgery. Because of the high recurrence rate, some scholars suggest the optimal treatment is wide resection [12]. However, total resection often associates with sacral nerve dysfunction and higher morbidity. So we preferred conservative surgery such as intralesional curettage and/or partial excision with effective intraoperative bleeding control (embolization and/or aortic balloon) in patients with sacral GCTs. During 2000 to 2013, 135 cases underwent such conservative surgery in our department with a mean blood loss of 3223 ml, and only 25 (25/135, 18.9%) cases had local recurrence [4].

There is some adjuvant therapy for GCTs such as argon beam coagulation, cryotherapy, serial embolization, and application of bisphosphonates, interferon alpha-2b, and denosumab. FDA had approved denosumab for the treatment of GCTs. Denosumab is a monoclonal antibody that targets receptor of activator nuclear factor kappa-B ligand (RANKL), thereby downregulating osteoclast activity. Denosumab has been shown to induce significant radiographic responses and to alleviate pain in extremity and spinal GCTs [13]. Chawla et al. [14] showed that denosumab had a 96% response rate in surgically unresectable GCTs in a 63-sacral GCT patient cohort. Thomas et al. treated 37 GCT cases with denosumab; 24 of them had recurrent disease. A positive response was seen in 86% of the patients [15]. Rutkowski et al. reported a 222 case series with denosumab; 86% of all patients experienced surgical downstaging. Only 15% of all the patients who underwent surgery had a local recurrence [16]. The current evidence shows that denosumab can control GCTs and sometimes shrink the tumor which facilitates subsequent surgery. However, there is no defined duration for the use of denosumab preoperatively as “overcalcification” of the tumor may make curettage during the surgery difficult. Moreover, we have no consensus on the endpoint for the use of this medicine as stand-alone treatment.

In conclusion, together with preoperative arterial embolization and/or aortic balloon, complete resection with perioperative denosumab might be the optimal treatment. However, because en bloc resection of these tumors is often very morbid, conservative resection could be a good choice. Denosumab, serial embolization [17], or radiotherapy might be considered for the patients who have an unresectable tumor or when it is too risky for surgery. Nevertheless, the role of radiotherapy remains unclear, as it has been implicated in the sarcomatous transformation [18].

20.2.2 Chordoma

Chordomas are slow-growing, low-grade malignant tumors that arise from vestigial notochordal remnants with a predilection for the ends of the spine. Chordomas often occur in the sacrococcygeal region and involve the fourth and fifth sacral segments [3, 19]. Chordomas are the most common primary malignant sacral tumor, with a peak incidence in the sixth decade and a male predominance [3, 18]. Chordomas are relatively resistant to conventional radiotherapy [20, 21] and chemotherapy [22], so surgical excision is the mainstay of treatment. It is reported that 5–40% of patients had local recurrence or distant metastases [22, 23]. The overall 5- and

10-year survival rates after sacrectomy are 45–77% and 28–50%, respectively [22–25].

The imaging of a chordoma often shows a large, lytic, destructive midline lesion with or without peripheral amorphous calcification centered in the vertebral body with adjacent presacral and/or sacral canal mass. Chordoma could extend across the sacroiliac joint and the intervertebral disk [3] and invade the surrounding muscles along with the piriformis and gluteus maximus muscle.

Stener and Gunterberg [26] first reported the idea of wide en bloc surgical resection for the treatment of sacral tumors. Since then, en bloc resection has been a goal in the surgical management of sacral chordomas. Fuchs et al. showed a significant difference in local control rate between patients who underwent wide resection and those who had a subtotal resection of sacral chordomas [27]; the time from surgery to local recurrence was 2.27 years and 8 months, respectively. Several other research also support that aggressive surgical resection could bring optimal local control in chordomas of the sacrum [27]. Moreover, some researcher advocate that total resection combined with advanced radiotherapy could substantially improve the local control rate of chordomas of the sacrum in recent years [22, 27].

20.2.3 Neurogenic Tumors

Benign sacral neurogenic tumors include peripheral schwannoma and neurofibroma. Malignant peripheral neurogenic tumors include malignant schwannoma (malignant peripheral nerve sheath tumor) and neurofibrosarcoma [28]. Neurogenic tumors arising from the sacrum are rare, with only about 7% of intraspinal neurogenic tumors involving the sacrum. The tumors often originate in the spinal canal or in close relation to the sacral nerve roots or their coverings, and grow out of the spinal canal through the neural foramina from the sacral canal and have a dumbbell shape. Inward growth of the tumors is generally limited due to the defined space of the sacral canal. However, for outward-growing tumors, a huge mass is often seen anterior to the sacrum. Initially, clinical symptoms are not evident in patients with sacral neurogenic tumors, especially for benign tumors, and symptoms such as lower back pain and sciatica occur only when the tumors become very large. Many patients visit the hospital because of an abdominal palpable painless mass or because a mass is discovered in the lower abdomen during a physical examination. The tumors often occur in females between 20 and 50 years old [28, 29].

Radiograph examination usually shows enlargement of the sacral neural foramen in benign cases, although the feature is not obvious in malignant neurogenic tumors. Most of the benign neurogenic tumors are shown as homogeneous lesions on an MRI, with about 6% showing a cystic degeneration change; however, most malignant neurogenic tumors show a heterogeneous signal change in the MRI, with about 75% showing cystic change. Therefore, an inhomogeneous signal and cystic change in huge neurogenic tumors indicates the possibility of malignancy [30, 31].

In a 790 consecutive primary sacral tumor case series from Peking University [4], there were 150 neurogenic tumors, with 131 benign neurogenic tumors (83 neurofibromas and 48 schwannomas) and 19 malignant schwannomas, which accounted for 19% of all primary sacral tumors. Among 131 benign neurogenic tumors, there were 62 males and 69 females with an average age of 42.3 years (17–67 years). All cases experienced marginal excision, and postoperative recurrence occurred in 17 (12.9%). According to our experience, surgical approach depends on the location and size of the tumors. Intraspinal tumors should be excised from a posterior approach. For giant neurogenic tumors that arise from the sacrum and involve the sacral canal, excision should be done by a combined anterior-posterior approach. Giant presacral neurogenic tumors located below the S1 level could be removed by a posterior approach. The anterior surgical approach should be applied for giant presacral neurogenic tumors that are located above S1 and do not involve the spinal canal [28].

20.2.4 High-Grade Osseous Tumors

High-grade osseous tumors including chondrosarcoma, osteosarcoma, and Ewing sarcoma often are very aggressive. Because these tumors respond poorly to chemotherapy and irradiation as for the advanced stage and axial location, they require a wide excision in the absence of systemic disease [27, 32]. Most osteosarcomas are primary, but sometimes they are the consequence of a malignant transformation of a giant cell tumor or of Paget disease. Sacral osteosarcoma is very rare, with only 2% of osteosarcomas involving the sacrum [31]. Peak incidence occurs in the third to fourth decade [33]. Osteosarcoma typically shows an aggressive, osteolytic, permeative pattern of bone destruction with cortical breakthrough and soft tissue mass [33]. Matrix mineralization of the soft tissue mass is more easily detected on CT.

Chondrosarcoma tends to be less aggressive except for the dedifferentiation type. Less than 7% of all chondrosarcomas arise in the sacrum [5]. Patients most commonly present in the fourth to sixth decades [31]. The typical imaging characteristic of chondrosarcoma is an osteolytic lesion with soft tissue mass and characteristic “rings and arcs” chondroid matrix mineralization. Unmineralized chondroid matrix often shows intermediate signal on T1-weighted and high signal on T2-weighted images. Mineralized region displays low signal intensity on all MRI sequences. Sometimes it is hard to distinguish between chondrosarcoma and chondroid chordoma because both of the tumors had chondroid matrix mineralization, though chordomas often affect the fourth and fifth sacral segments and centered in the vertebral body, while chondrosarcomas tend to originate from the upper sacrum in an eccentric pattern.

Ewing sarcoma is a small, round blue cell malignancy usually seen in the second decade of life with a male predominance [34]. More than half of spinal Ewing sarcomas occur in the sacrum, and more than two thirds occur in the sacral ala [34]. On radiographs, the tumors may show permeative osteolysis. CT often reveals a permeative pattern of bone destruction. Sometimes, lesions may show a mixed pattern of osteolysis and sclerosis, but there is no matrix mineralization in the soft tissue mass [34]. Soft tissue mass and spinal canal involvement are frequently seen and best defined on MRI. Ewing sarcoma has one of the highest mortality rates among all malignant bone tumors. It is regarded as a surgical condition only when encountered in the sacrum because of its propensity to metastasize early and because of its favorable response to both irradiation and chemotherapy [32].

From 2000 to 2013, 26 osteosarcomas, 49 chondrosarcomas, and 28 Ewing sarcomas/PNETs out of all 790 primary sacral tumors underwent surgeries in our department [4]. Ewing sarcomas/PNETs accounted for 3.5% of all primary sacral tumors. Twenty-one cases accepted neoadjuvant chemotherapy, while 27 cases received postoperative chemotherapy and radiotherapy. Fourteen cases underwent en bloc resection or total sacrectomy, while 14 cases underwent piecemeal resection. Fifteen cases (53.6%) were noted postoperative recurrence. Three-year overall survival rate was 39.1%, and the 5-year overall survival rate was 19.6%. A total of 49 sacral chondrosarcomas accounted for 6.2% of all cases. There were 26 males and 23 females with an average age of 42.5 years (17–69 years). Among these 49 patients, 29 cases underwent en bloc resection or total sacrectomy, while piecemeal resection was performed on the others. Twenty-two cases (44.9%) were noted postoperative recurrence. The overall survival rate at 2 years and 5 years was 58.7% and 47.0%, respectively. The disease-free survival rate at 2 years and 5 years was 42.3% and 31.8%, respectively. A total of 26 sacral osteosarcomas were enrolled [35], which accounted for 3.3% of the whole series. There were 15 males and 11 females with a median age of 28 years (range, 12–68 years). Adequate and inadequate surgical margins were obtained in 16 and 10 cases, respectively. Distal metastasis occurred in 13 patients (50%), and local recurrence occurred in 10 patients (38.5%, including six patients with additional distal metastasis). The 1-year and 5-year survival rates were 92.3% and 38.7%, respectively. The result of this research reveals that adequate margins can significantly improve the recurrence rate and event-free survival rate compared to inadequate margins. There are very limited reports about the prognosis after integrated therapy such as chemotherapy and radiation of high-grade osseous sacrum tumors because of the rarity. So it is hard to draw a sound conclusion of the optimal treatment strategy for these cases yet. Current evidence support that surgical excision with adequate margin is still the golden standard for high-grade primary sacral tumors. The prognosis remains dismal, and more collaborative clinical trials are needed to improve the survival.

20.3 Management Consideration

20.3.1 Biopsy

With the age, gender, symptom, location, and imaging characteristics, experienced doctors should make a preliminary diagnosis. However, the preoperative biopsy is still critical especially for the patients whose pathologic diagnosis would influence the decision to operate or the type of surgery. Although open biopsies and transrectal biopsies were common in the past years, almost all biopsies are now performed percutaneously with the assistance of image guidance. The biopsy tract should be included within the boundaries of the subsequent surgery. As for the spine tumors, the accuracy of percutaneous vertebral biopsy varies from 66% to 96% [36–38]. The highest diagnostic precision is generally achieved in metastatic cases, with a diagnostic accuracy rate of 79–96% [36–38]. Accuracy and diagnostic value are lower in cases of primary bone tumors, estimated at 60–80% [36–38]. Taking into account that the sacrococcygeal region is easily accessible for surgical sampling, an open biopsy would be considered if the percutaneous biopsy failed to obtain an adequate sample.

20.3.2 Primary Sacral Tumors

The goal of the treatment of primary sacral tumors is to be curative. Surgical excision is still the optimal way to achieve local control. The surgical intervention will be discussed profoundly in other chapters. Because of the advanced disease and critical anatomy location, surgery with adequate margin is not always feasible for all cases. Conventional therapeutic methods, such as radiotherapy and chemotherapy, could be used as the neoadjuvant or adjuvant treatment in certain histologic types or even the only treatment for the unresectable tumors.

20.3.3 Radiotherapy

Radiotherapy is a feasible adjuvant option especially for subtotally resected tumors, local recurrences, and unresectable tumors. Carbon ion radiotherapy and proton/photon therapy were shown to have better results compared to conventional radiotherapy because of increased effective doses and the lower complication rate [39]. Reiko et al. reported [40] patients with unresectable sacral chordomas received carbon ion radiotherapy with the dose of 52.8–73.6 Gray equivalents. The 5-year overall survival rate was 86%, and the 5-year local control rate was 89%. Thomas et al. [41] reported a total of 50 patients (29 chordomas, 14 chondrosarcomas, 7 others) underwent gross total [29] or subtotal [14] resection or biopsy [15]. With 48-month median follow-up, 5-year local control rate, recurrence-free survival, and overall survival are 78%, 63%, and 87%, respectively. Moreover, modern intensity-modulated radiation therapy and stereotactic radiosurgery (SRS) also allow high-dose hypofractionation and minimized complications [42, 43]. Radiotherapy is a crucial treatment for sacral sarcomas. Osteosarcomas are considered to be radiation resistant, but advanced techniques can also improve the local control rate for the cases undergoing piecemeal or subtotal resection [44]. For sacral Ewing sarcomas, radiotherapy might be the best way for local control when en bloc resection is not feasible or patient could not tolerate the postoperative nerve dysfunction [45].

20.3.4 Chemotherapy

Most sacral tumors are benign lesions or low-grade malignancies. Thus, chemotherapy is not necessary for such cases. However, for the chemo-resistant tumors such as chordomas and GCTs, the targeted therapeutic agents are used in recent years. Casali et al. [46] first published the result of imatinib therapy in chordoma patients. A multicenter phase II study also supports the positive effect of imatinib on progression-free survival. Hof et al. [47] used cetuximab and gefitinib with a good response in a chordoma patient with local recurrence and lung metastases. Chawla et al. [14] showed that denosumab had a 96% response rate in surgically unresectable GCTs in a 63-sacral GCT patient cohort. Thomas et al. treated 37 GCT cases with denosumab; 24 of them had recurrent disease. A positive response was seen in 86% of the patients [15]. Rutkowski et al. reported a 222 case series with denosumab; 86% of the patients experienced surgical downstaging. Only 15% of all the patients who underwent surgery had a local recurrence [16].

Similar to the treatment strategy for sarcomas of other sites, chemotherapy is crucial for high-grade primary malignant sacral tumors (Ewing and osteosarcoma). Recurrence-free and overall survival is increased significantly with combined adjuvant chemotherapy in the osteosarcomas and Ewing sarcomas [48–50]. Hoffman et al. reported the histologic response to chemotherapy was analyzed in the surgical specimen and had a significant influence on survival [51]. Unfortunately, there is no optimal chemotherapy protocol for chondrosarcomas yet. Italiano et al. reported 180 patients treated with chemotherapy in 15 institutions in Europe and the USA; the response rate is 31% for mesenchymal chondrosarcoma, 20.5% for dedifferentiated chondrosarcoma, only 11.5% for conventional chondrosarcoma, and 0% for clear cell chondrosarcoma [52]. But with the development of immunology and in-depth study of the mechanism of cancer initiation, there are high hopes for new therapeutic agents.

20.4 Metastatic Tumors

Metastatic tumors are the most common malignancies in the sacrum. Breast, lung, renal, thyroid, and prostate cancers are the predominant primary origins; less common primary lesions include lymphoma, melanoma, and tumors of unknown origin [53, 54]. Hematogenous metastasis is the main way of spreading cancer, although direct invasion from pelvic tumors is also commonly seen [55, 56].

The treatment decision of sacral metastasis is based on individualized evaluation such as the health status of the patient, the location of the lesion, the biology, and the response to chemotherapy of the tumors. The mainstay of management for sacral metastatic tumors is palliative therapy. Radiotherapy is the major treatment for the cases without spinal instability. Good pain relief and neurological improvement are attainable after radiation [57, 58]. Minimal invasive procedures such as sacroplasties could also provide immediate pain relief and improvement with ambulation [59]. Only when the bone destruction jeopardizes the spinal stability and conservative therapy fails to improve neurological status, an aggressive surgery including tumor resection and lumbosacral reconstruction should be considered [60, 61], as radiosurgery has demonstrated promising results with local disease control [62].

20.5 Summary

Sacral tumors are rare in incidence. The most common tumors are metastatic neoplasms. Chordomas, giant cell tumors, neurogenic tumors, and chondrosarcomas are the most frequently seen primary pathologic types. The rarity and insidiously growing pattern lead to a delay in diagnosis and intervention. The surgical treatment of sacral tumors could be very challenging because of the advanced disease and complicated anatomy. The multidisciplinary team including neurologic, colorectal, urologic, gynecological, and plastic surgeons, as well as sophisticated ICU doctors and anesthetists, are often needed. Surgical resection with adequate margin (en bloc resection) is still vital for local control, but only quite limited centers have enough such experiences. Radiotherapy and chemotherapy are proved to be effective for certain tumors. However, the prognosis of sacral sarcomas and metastatic cancer is still unencouraging. In the later chapters, we will discuss the topics including the anatomy of the sacrum, diagnostic imaging method, pathology, surgical treatment strategy, and its results of the sacral tumors. Some special experience from Peking University such as hemorrhage control, neurologic complications and wound care, and rehabilitation protocols are also to be introduced.

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21 Surgical Anatomy of the Sacrum

Huayi Qu and Wei Guo

The sacrum is a triangular bony structure which is fused together with five sacral vertebrae. The sacrum articulates with the ilium, the fifth vertebra, and the coccyx, forming the posterior wall of the bony pelvis. It was oblique to the spine, with the anterior surface being concave, which increases pelvic capacity to contain the pelvic organ. The lateral surfaces of the sacrum form the joint surfaces, which are articulated with the posterior inner parts of the ileum. It has a base, three surfaces, and an apex.

Base. The base of the sacrum, which has a similar shape to that of the lower surface of the fifth lumbar vertebral body, is oval in shape. It is the upper surface of the first sacrum. The anterior edge is termed the sacral promontory. The pedicles of the sacra are short and divergent posterolaterally to join with the laminae. The laminae incline backwards and medially and then meet in the midline and form the spinous tubercle. The upper part of the sacrum articulates with the fifth lumbar vertebra by the intervertebral disc and the facet joints, which form by the superior articular processes of the sacrum and the inferior articular processes of the fifth lumbar vertebra. The transverse process is a broad bony structure, sloping and projecting laterally away from the sacral body and the pedicle. It consists of transverse process and costal element, forming the superior part of the sacral ala.

The surfaces of the sacrum. The ventral surface of the sacrum is concave and the dorsal surface is convex. Four pairs of sacral foramina communicate through intervertebral foramina with the sacral canal, transmitting ventral rami of the upper four sacral spinal nerves. The large area between the right and left foramina formed by flat pelvic aspects of the sacral bodies shows their fusion by four transverse ridges. Bars between foramina are costal elements fused to the vertebrae. Lateral to the foramina, the costal elements unite together and posteriorly with transverse processes to form the ala of the sacrum. The pelvic surface gives attachment to the piriformes. The first three sacral ventral rami emerge from the pelvic sacral foramina and then pass anteriorly to piriformis to join in the sciatic nerve and the pudendal nerve. The sympathetic trunks descend medially to the sacral foramina; and the median sacral vessels descend in the midline, which lie in direct contact with the ventral surface of the sacrum. The lateral sacral vessels pass through laterally to the sacral foramina. The parietal peritoneum covers the ventral surfaces of the first three sacral bodies; and the rest of the ventral surface of the sacral vertebrae is in contact with the rectum. The superior rectal artery, which serves the distal part of the rectum, bifurcates at the level of the third sacral vertebra.

The dorsal surface of the sacrum was convex and irregular. A raised median sacral crest, with four spinous tubercles, runs down in the middle line. The median sacral crest stops at the sacral hiatus, approximately at the level of the fourth spinous tubercle, where the dorsal surface of the fifth vertebral body is exposed. The posterior surface of the sacrum is constituted by fused laminae. Lateral to the median crest lie four pairs of dorsal sacral foramina, which communicate with the sacral canal through intervertebral foramina. The dorsal rami of sacral nerves transmit through the sacral foramina. The intermediate sacral crests lie medially to the foramina, which are two rows of small tubercles, representing the fused articular processes. The lateral sacral crests, formed by fused transverse processes, lie laterally to the dorsal sacral foramina. The erector spinae attaches to the elongated U-shaped area between the median and lateral sacral crest; the sacral spinal dorsal rami serve these muscles as they emerge from dorsal foramina.

The lateral surface, which is wide above and narrow in its lower part, is a fusion area of transverse processes and the residual costae. It articulates with the ileum with an auricular surface in the inferior and anterior part and a rough and deeply pitted superior and posterior part for the attachment of ligaments between the sacrum and ileum. The auricular surface occupies the upper lateral surface from the first sacral vertebra to the middle of the third of the sacral vertebra.

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Inferior to the auricular surface, the sacrum reduces in breadth and curves medially to the body of the fifth sacral vertebra.

The auricular area of the sacrum is covered by hyaline cartilage. The auricular area has elevations and depressions to increase the contact surface of the joint, so as to increase the stability of the rigid joint. The rough area is superior and posterior to the auricular surface, which has two or three depressions for attachment of the interosseous sacroiliac ligaments. The gluteus maximus and the sacrotuberous and sacrospinous ligaments attach to the lateral side of the sacrum below the auricular surface; and anterior to these structures, the piriformes attach to the pelvic surface and lateral side of the sacrum.

Sacral apex. The apex is the distal end of the fifth sacral vertebral body. The oval surface of the apex articulates with the coccyx.

Sacral canal. The sacral canal is triangular in transverse plane, communicating from the fifth vertebral foramina to the hiatus. It is formed by sacral vertebral foramina. Owing to the sacral inclination, its upper opening is oblique when standing. In the lateral wall lies four intervertebral foramina, which communicate with pelvic and dorsal sacral foramina.

The sacral canal accommodates the sacral dural sac, cauda equina, and filum terminale. Approximately at the end of the second sacrum, the sacral dural sac ceases. The sacral spinal roots and filum terminale pierce the arachnoid and dura maters and pass through the sacral canal and the intervertebral foramina, forming the ventral rami or dorsal rami. The fifth sacral nerves, the coccygeal nerve, and the filum terminale emerge from the sacral hiatus to innervate the coccygeal region.

Sex differences in sacra. Generally, the female sacrum is shorter and wide, producing a wider pelvic cavity. Sacral width, as a percentage of length, yields a sacral index. The ventral concavity is deeper in females, and its deepest point is usually higher than in males; curvature above this point is greater in the female. The dorsal protrusion of the second sacral vertebra is therefore usually less prominent in males. In females the pelvic surface faces downwards more than in males, increasing the pelvic cavity and making the lumbosacral angle more prominent. In the male, the first sacral body occupies a larger proportion of the sacral base, for the fifth lumbar body is usually large and the pelvis is relatively narrow. The transverse diameter exceeds the length of an ala, while the diameter of the sacral base is roughly equal to that of the ala.

Variations. The fifth lumbar or first coccygeal vertebrae may incorporate with the sacrum, which results in five pairs of intervertebral foramina on both the ventral and dorsal surfaces. The incorporation of the fifth lumbar (sacralization of the lumbar spine) is usually at one side, or even only an enlarged transverse process of the fifth lumbar spine articulates with the sacrum at the posterolateral aspect of the ala. Lumbarization of the first sacral vertebra occurs, but relatively rare. The development of laminae and spines may be incomplete, resulting in the deficiency of the posterior aspect of the sacral canal.

Attachments of the first sacral body. The anterior longitudinal ligament attaches to the middle portion of the anterior surface of the sacrum till to the coccyx. The terminal fibres of the posterior longitudinal ligaments attach to the anterior wall of the sacral canal. Upper laminar borders of the sacrum receive the lowest part of the ligamenta flava, which attaches to the bony structure of the posterior wall of spinal canal. The lumbosacral trunk obliquely grooves the smooth area of the superior part of the ala. The lower band of the iliolumbar ligament attaches to the rough area of the lateral side of the ala. The whole ala is covered mostly by the psoas major. Iliacus reaches the anterolateral part of this area.

Sacroiliac joint and the surrounding ligaments. The sacroiliac joint is a rigid weight-bearing joint, formed by ear-shaped auricular surfaces of the sacrum and the ilium and posterior syndesmoses. The auricular surfaces are smooth but with elevations and depressions. The auricular surfaces interlock with each other, allowing the joint a limited mobility. Syndesmoses are quite strong, facilitating the transmission of body weight to the lower limb. Several strong ligaments around the sacroiliac joints improve the stability of it. The anterior sacroiliac ligaments are broad and thin layers of fibrous capsule, which exists only in the superior parts of the joints. The interosseous sacroiliac ligaments, lying between the rough area of the sacrum and ilium posterior to the auricular surface, are the major structures that support body weight of the upper part. The posterior sacroiliac ligaments, which are the superficial parts of the syndesmosis, run upwards and downwards from the lateral crest of sacrum to the posterior parts of ilium and the sacrotuberous ligament (Fig. 21.1).

The ligaments connecting the sacrum and the spine. Sacro-lumbar ligaments are strong triangular ligaments, which connect the fifth transverse process and the iliac crest. The sacrotuberous ligament attaches broadly to the long oblique area of the posterior superior iliac crest, the lower transverse sacral tubercles, and the lateral margins of the lower sacrum and the upper coccyx. Its fibres, which are partly blended with the posterior sacroiliac ligaments, converging and descending laterally, attach to the ischial tuberosity's medial margin. Sacrospinous ligaments are thin and triangular in shape. Lying deeply to the sacrotuberous ligament, it attaches to the lateral margins of the sacrum and coccyx and extends to the ischial spine. The pudendal nerve and internal pudendal vessels, lying anteriorly to the piriformis, leave the pelvis and curve around the dorsal aspect of the ischial spine and then re-enter the internal pelvic wall by the lesser sciatic foramen. From the posterior view, the sacrospinous ligaments lie deeply to the sacrotuberous ligaments. Two ligaments stabilize the sacrum on the pelvis and prevent the tilting of it. The greater and lesser sciatic foramens, which are divided by sacrotuberous and sacrospinous ligaments from the sciatic notch region, are the entrances and exits of muscles, vessels, and nerves (Figs. 21.1 and 21.2).

The sacroccygeal joint. The apex of the sacrum and the base of the coccyx join with each other, forming the sacrococcygeal joint. It is strengthened by sacrococcygeal ligaments anteriorly and posteriorly.

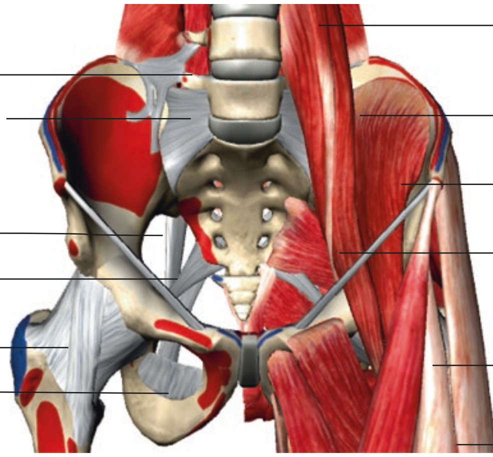

Fig. 21.1 The ligaments and muscles around the pelvis and the sacrum (anterior view)

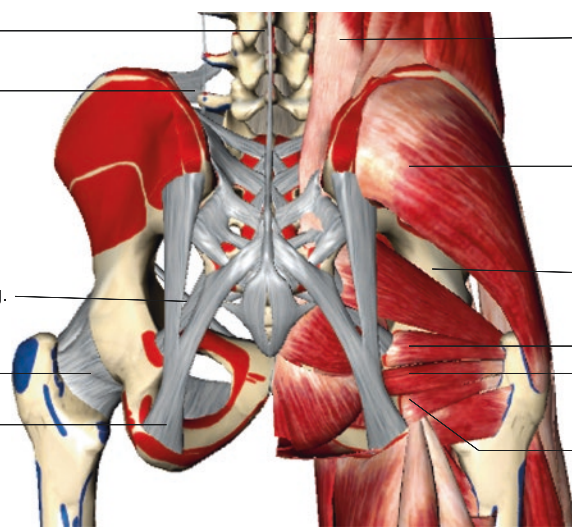

Fig. 21.2 The ligaments and muscles around the pelvis and the sacrum (posterior view)

22 Diagnostic Imaging and Differential Diagnosis

Yi Yang and Wei Guo

Sacral tumors are unique to the skeletal system in that the prevalence of various neoplasms is different from that at other osseous sites. There are also unique imaging-related considerations, related in part to diagnosis, but particularly to the evaluation site for tumor biopsy sampling and resection.

Multiple modalities are available for imaging the sacrum in patients with cancer, including conventional radiography, computed tomography (CT), radionuclide imaging, magnetic resonance imaging (MRI), functional and metabolic imaging, and interventional radiology techniques. After a careful history and physical examination, the next step in the evaluation of the patient with primary or secondary neoplastic involvement of the sacrum is selection of the appropriate imaging modality and its application at the appropriate region of the sacrum.

Sacral tumors are rare. As with nearly any osseous structure, the majority of lesions are metastatic; these will not be discussed further. Primary benign and malignant tumors of the sacrum are rare, accounting for fewer than 7% of all spinal primary tumors [1]. Additionally, there are other processes that can mimic sacral neoplasms clinically and occasionally radiographically, including insufficiency fractures and radiation osteonecrosis. In this section, imaging characteristics of the most common sacral lesions will be presented.

22.1 Benign Sacral Lesions

22.1.1 Giant Cell Tumor

Giant cell tumors are locally aggressive, slow-growing tumors that typically present between the second and fourth decades of life. Giant cell tumors are uncommonly located in the spine. In a multicenter, multinational study of 1277 giant cell tumors, only 2.7% were localized to the spine [2].

Within the spine, the vast majority of GCTs occur in the sacrum. This lesion is the second most common primary tumor of the sacrum, comprising 13% of all sacral tumors, second only to chordoma. It is the most common benign sacral neoplasm (71%) [3].

The majority of GCTs are benign; 5–10% have been reported to be malignant. Many of the malignant lesions are thought to be related to previous radiotherapy. It is somewhat confounding that the degree of aggressiveness depicted on imaging is not strongly predictive of grade or even malignancy. Furthermore, GCTs occasionally metastasize to the lung even though the primary lesion is considered histologically benign.

It typically appears as an area of rarefaction on routine X-ray (Fig. 22.1a). As the tumor expands, it may produce cortical expansion of the sacral vertebra. The GCT is a lytic, expansive lesion that never contains matrix. In the sacrum, it usually develops in an eccentric position [4] but commonly extends to involve both sides of the midline and may cross the sacroiliac joint. There is usually a thin cortical rim, but the soft-tissue mass can break through this rim, and the mass is usually quite large before it is detected. CT is particularly helpful in characterizing the lesion, which may exhibit scalloped borders by CT (Fig. 22.1b). Its soft-tissue attenuation may contain foci of lower attenuation, indicating areas of hemorrhage or necrosis. Because the lesion is hypervascular, it enhances on CT scans. Magnetic resonance imaging often provides much more distinctive images, allowing fairly definitive diagnosis of GCT. On T1-weighted MR image sequences, low to intermediate signal intensity associated with most osseous neoplasms is observed; T2-weighted MR imaging, however, is not as nonspecific, demonstrating low to intermediate signal intensity in a very heterogeneous pattern in 63–96% of cases [1]. This low signal intensity on T2-weighted sequences is a distinguishing feature of the GCT; most other osseous neoplasms demonstrate high signal intensity on T2-weighted sequences. It is thought that the

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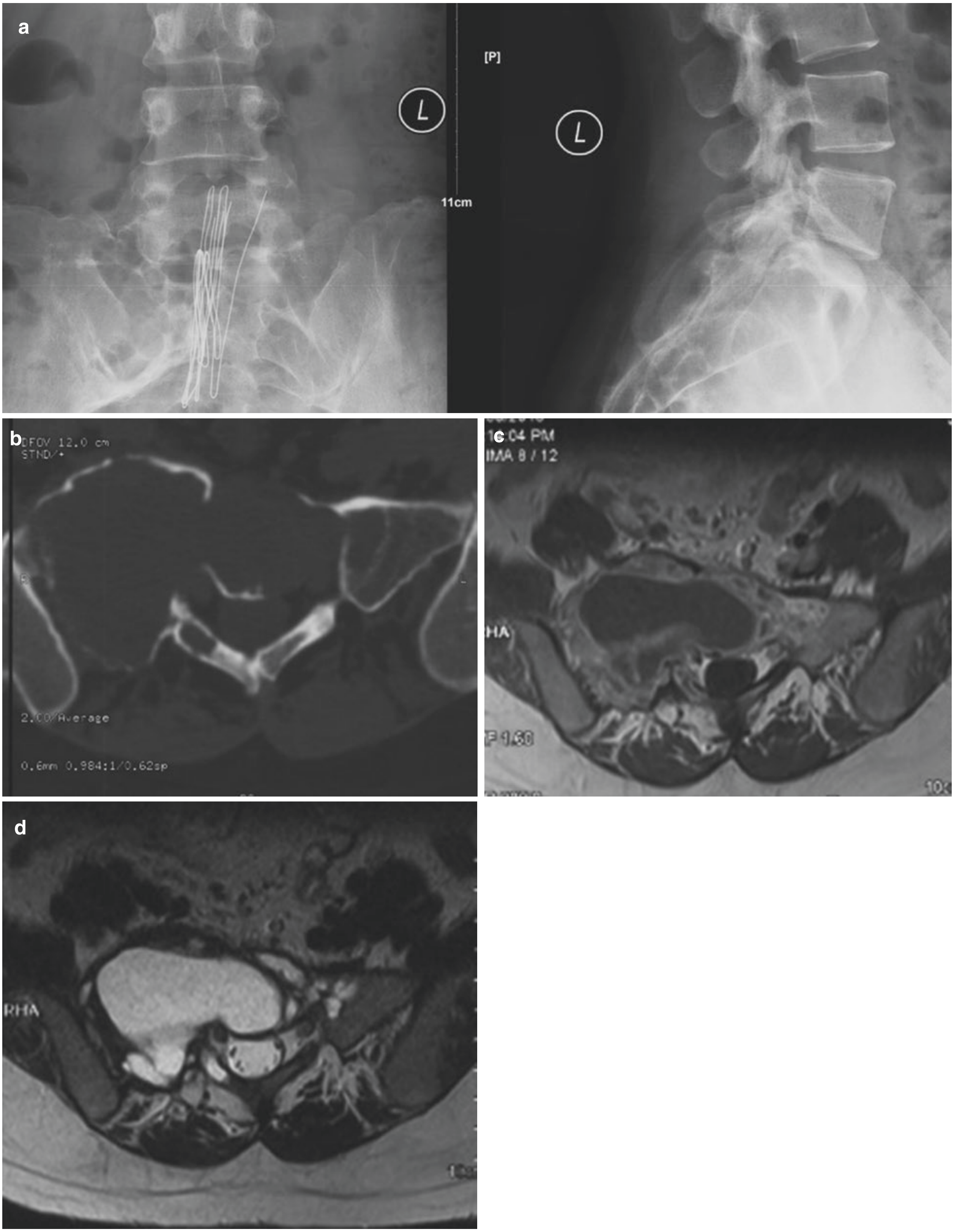

Fig. 22.1 Imaging studies obtained in a 36-year-old woman with GCT of the sacrum. (a) Anteroposterior-lateral radiograph revealing a lytic destructive lesion involving S1–3. (**b**) Axial CT scan confirming the lytic nature of the lesion. In a patient of this age, a diagnosis of GCT should be strongly considered. Magnetic resonance imaging is very useful in suggesting the correct diagnosis. (**c**) Sagittal T_1-weighted MR image revealing that the lesion is of low signal intensity. (**d**) Axial fast-spin echo T_2-weighted MR image demonstrating an inhomogeneous mass that contains several areas of low signal intensity

lower signal intensity is due to prominent collagenous content as well as hemosiderin deposition. Occasionally hemorrhage is detected on both T1- and T2-weighted sequences as high signal intensity (Fig. 22.1c, d). Rarely, fluid-fluid levels may be present. Giant cell tumor may be confused with ABC and benign osteoblastoma.

22.1.2 Aneurysmal Bone Cyst

Aneurysmal bone cyst (ABC) is a benign proliferative lesion of unknown etiology that is neither a true bone cyst, nor an aneurysm, nor neoplastic [5]. It typically affects patients younger than 20 years of age and has a slight female predominance. ABC affects the spine in approx. 10–20% of cases; of these cases, sacral involvement occurs in only 20% [4]. It is an expansile, osteolytic lesion of bone, often with sclerotic margins suggesting a slow growing process. As the lesion progresses, it may assume a soap bubble appearance with eggshell thin cortical margins, which may progressively expand and blow out [6].

Secondary ABC is related to an underlying neoplasm (most frequently GCT, osteoblastoma, chondroblastoma, or osteosarcoma) that causes venous obstruction or an arteriovenous fistula [7]. The majority of ABCs (65–99%) are considered primary lesions, but one should always consider the possibility of an underlying lesion when evaluating them radiologically and histologically.

In the sacrum, the ABC lesion originating in the sacral ala but often extending to involve the VB (Fig. 22.2a). The lesion is lytic, is expansive, and has a thin cortical rim that often seems to contain the lesion. This thin cortical rim may be seen only on CT scans (Fig. 22.2b). On Tc bone scanning, ABC may show peripheral increased uptake, with a photogenic center. This appearance has also been described in GCT. Computerized tomography scanning often demonstrates fluid–fluid levels caused by hemorrhage with sedimentation. Although fluid–fluid levels can be seen on CT scans, they are much more easily visualized on MR images. Although T_1-weighted MR imaging may be nonspecific with the mass showing intermediate signal intensity, it occasionally reveals an increase in signal intensity due to the methemoglobin in the blood-filled spaces. Magnetic resonance imaging with T_2-weighted sequences more reliably demonstrates the fluid-fluid levels (Fig. 22.2c). Note that the high signal intensity methemoglobin may be either the dependent or the nondependent component. Additionally, the useful planes for observing fluid levels are axial and sagittal; coronal imaging masks the fluid levels because imaging is performed with the patient in a supine position.

22.1.3 Osteoblastoma

Osteoblastoma is a rare lesion (1–2% of all bone lesions), but one that has its predominant location in the spine. Forty percent of all osteoblastomas occur in the spine; of these, 17% arise in the sacrum [4]. Osteoblastomas are found predominantly in young adults (90% in the second and third decades, but with a wide age range of 3–72 years). The male/female ratio is 2:1.

Osteoblastomas, like ABC, usually originate in the posterior elements of the spine, but 42% extend into the VB. A small percentage (10–15%) may have an ABC component [1]. Three radiographic/CT patterns have been described. The first is a lytic lesion that may not contain central calcification and which is surrounded by sclerosis. The second pattern, the most common, involves an expanded lesion with multiple small calcifications and a peripheral sclerotic rim. The third pattern is more aggressive, showing bone destruction and infiltration of surrounding soft tissues, often with a matrix. The matrix in osteoblastoma can usually be identified by radiography or CT scanning as osteoid (Fig. 22.3a–c).

Osteoblastoma shows marked uptake on bone scanning. On CT scans, its appearance mimics that seen on radiographs. Its MR imaging appearance is nonspecific, with low to intermediate signal intensity on T_1-weighted images and intermediate to high intensity on T_2-weighted, depending on the amount of matrix present. There may be extensive peritumoral edema [1].

Osteoblastoma should be excised. The lesion recurs in 10–15% of cases, but the rate approaches 50% in the more aggressive variety of osteoblastoma [1]. Therefore, it is important to identify aggressive features by imaging prior to surgery because this may lead to more aggressive surgery as well as more frequent postoperative surveillance. There have been rare reports of malignant transformation of osteoblastoma to osteosarcoma with metastases.

Osteoid osteoma is often considered with osteoblastoma, because 10% occur in the axial skeleton (posterior elements) and because their appearance is similar to the first pattern in the aforementioned description. Only 2% of spinal osteoid osteomas occur in the sacrum [3].

22.1.4 Nerve Sheath Tumors

Nerve sheath tumors (neurofibroma and schwannoma) may arise from sacral nerve roots. They produce an intradural extramedullary mass and therefore are not true sacral neoplasms. They may be large and dumbbell shaped, however, with extradural components that erode and enlarge the neural

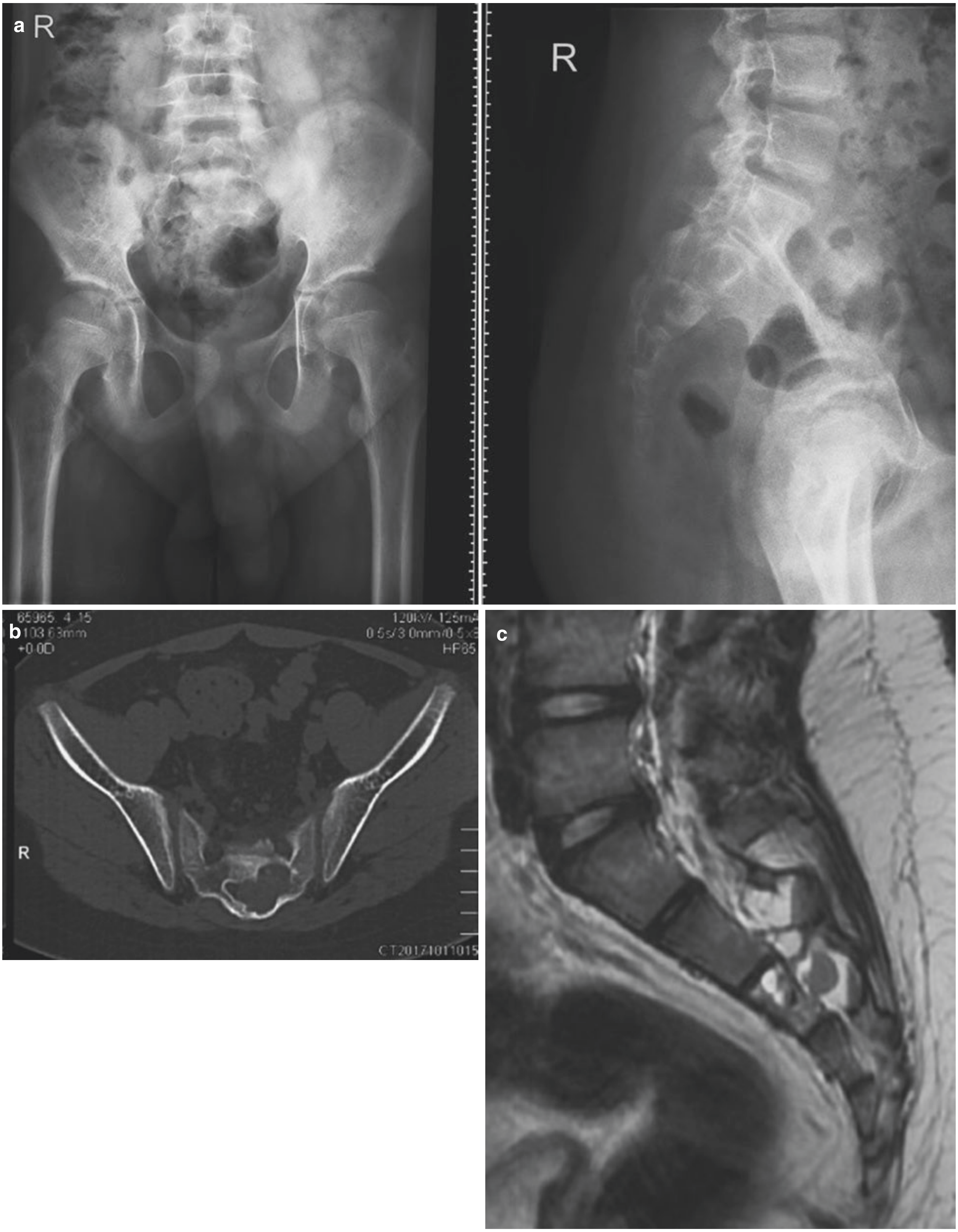

Fig. 22.2 Imaging studies obtained in an 11-year-old boy with ABC. (**a**) The anteroposterior radiograph can be easily misread as normal because of the overlying bowel gas obscuring the sacrum. A lateral radiograph demonstrates only obscuration of the S2–3 posterior elements (*arrows*). (**b**) The lesion is more readily seen on the CT scan. This scan demonstrates a lytic lesion occupying the left S2–3 ala, with a thin cortical rim surrounding the lesion. (**c**) Fluid levels (*short arrow*) are more readily observed on a sagittal T_2-weighted MR image on the sagittal exam. The high signal intensity portion of the fluid is blood. Most, but not all, ABCs contain fluid levels. Conversely, most lesions with substantial fluid levels are ABCs, but such levels may occur in other lesions as well

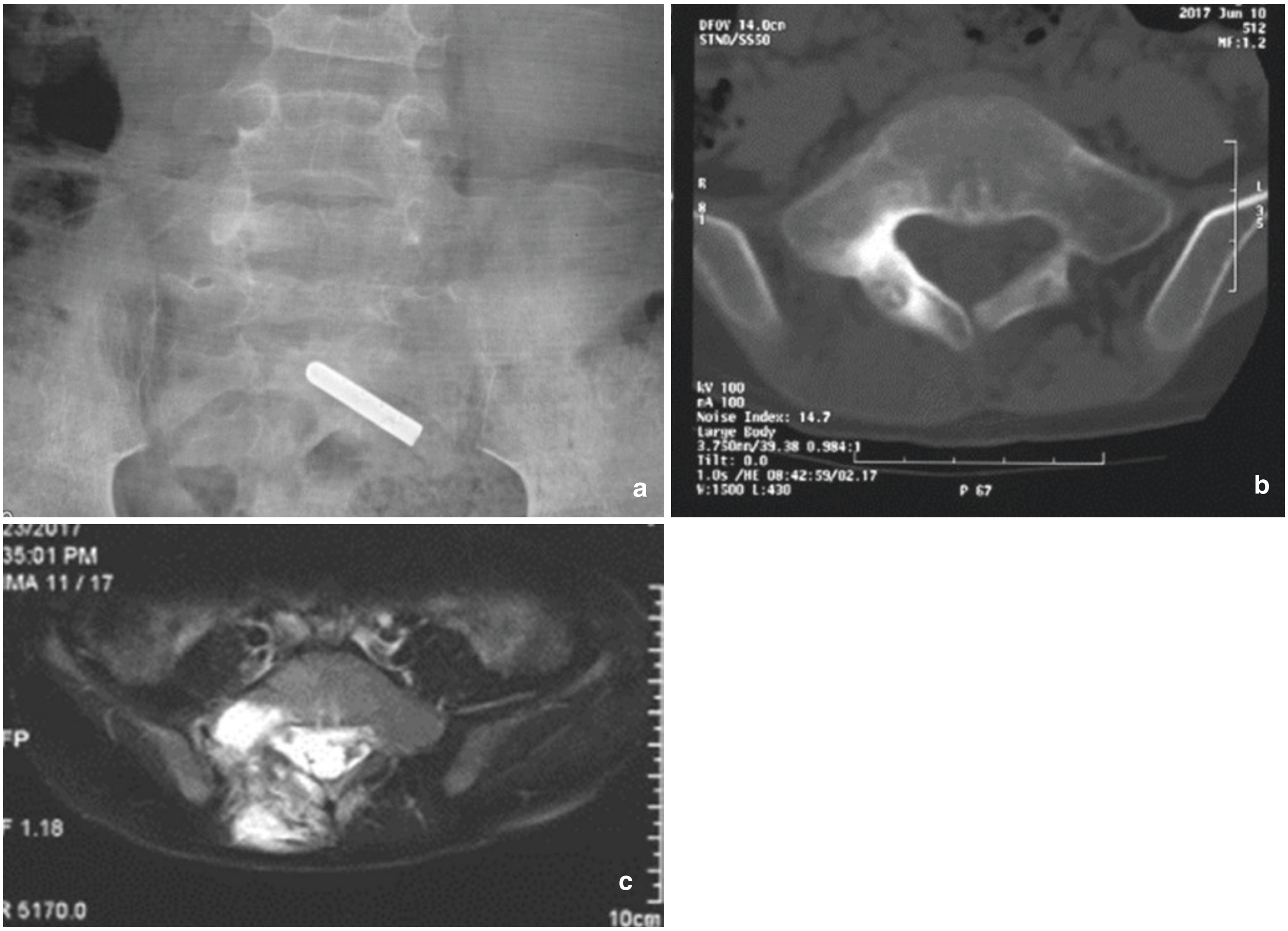

Fig. 22.3 Radiological studies obtained in a 7-year-old man with osteoblastoma. (**a**) Anteroposterior radiograph revealing a subtly osteogenic lesion at S1 (*arrows*). (**b** and **c**) Axial CT and MRI scan demonstrating bone matrix within the minimally expanded lesion, not aggressive in appearance. Thus, based on its location in the spine, presence of bone matrix, and degree of aggressiveness, the lesion must be an osteoblastoma

foramina. This appearance is not difficult to diagnose as a NST. In severe cases, these NSTs may enlarge to such a degree that they result in sacral destruction and large soft-tissue masses. In such cases, the lesions cannot be radiographically distinguished from other benign or even malignant sacral tumors. Both neurofibroma and schwannoma have been described as occasionally having a "target" appearance on T_2-weighted MR imaging, with a low signal center surrounded by high signal mass. Many cases, however, will be nonspecific in appearance (Fig. 22.4a–d).

22.2 Malignant Sacral Lesions

22.2.1 Chordoma

Chordoma is a relatively rare tumor, representing only 2–4% of all primary malignant bone neoplasms. Within the spine, however, chordoma is the most common primary malignant tumor (20–34%) [1]. Furthermore, it is the most common tumor of any type involving the sacrum, representing 40% of all primary sacral neoplasms [3]. Because the lesion arises from notochordal remnants, it is not surprising that its location parallels that of the notochord. Fifty percent of all chordomas arise in the sacrum; within the sacrum, the S4 and S5 levels are the most commonly involved. The lesion develops within the VB but extends, often with a very large presacral mass, into the canal 60% of the cases [8].

The most frequent age range for chordoma is 30–60 years, with incidence peaking in the fifth decade. The male/female ratio is 2–3:1 [1]. Radiographically, the lesion is lytic, is expansive, and may have a very large associated mass (Fig. 22.5a). Intratumoral calcification is seen in 50–70% of cases radiographically but in 90% of cases on CT scanning (Fig. 22.5b). On MR imaging, chordomas display a low to intermediate signal on T_1- and very high signal intensity on T_2-weighted sequences (Fig. 22.5c). If the calcifications are large enough, they may be seen as low signal structures

Fig. 22.4 Imaging studies obtained in a 54-year-old man with schwannoma. (**a**) Anteroposterior radiographs demonstrating a lytic, expansive, destructive lesion involving left part of S2. (**b**) Axial CT scan demonstrating involvement of the left part of the sacrum with a solid mass and extension of the mass well into the presacral space. (**c** and **d**) Sagittal T1-weighted and axial fast-spin echo T2-weighted MR images revealing the extent of the lesion. The lesion is inhomogeneous, containing both low and high signal regions. Schwannoma may exhibit a "target sign" on T2-weighted images. Analysis of a tissue specimen confirmed the diagnosis of schwannoma

within the lesion, but generally low signal is not a common characteristic on T_2-weighted imaging. The lesion enhances after Gd administration.

Primary treatment of chordoma is wide resection, and the prognosis relates to its completeness. Therefore, preoperative cross-sectional imaging and interpretation of findings are key. In cases in which an incomplete resection is achieved, supplemental radiotherapy may be performed, but its efficacy may not be as great as in higher-grade lesions. Local recurrence results in high morbidity rates and is often the eventual cause of death. Metastases eventually develop in 5–43% of patients and may be found in the liver, lung, regional lymph nodes, and unusual sites such as the peritoneum, skin, and heart [9].

22.2.2 Multiple Myeloma

Multiple myeloma is the second most common primary malignant neoplasm of the sacrum. Its incidence peaks in the sixth and seventh decades, and the male/female ratio is 2:1 [3]. Multiple myeloma is seen in the sacrum and other bones as either multiple round "punched-out" lytic lesions, an MR imaging-documented diffuse osteoporosis with an

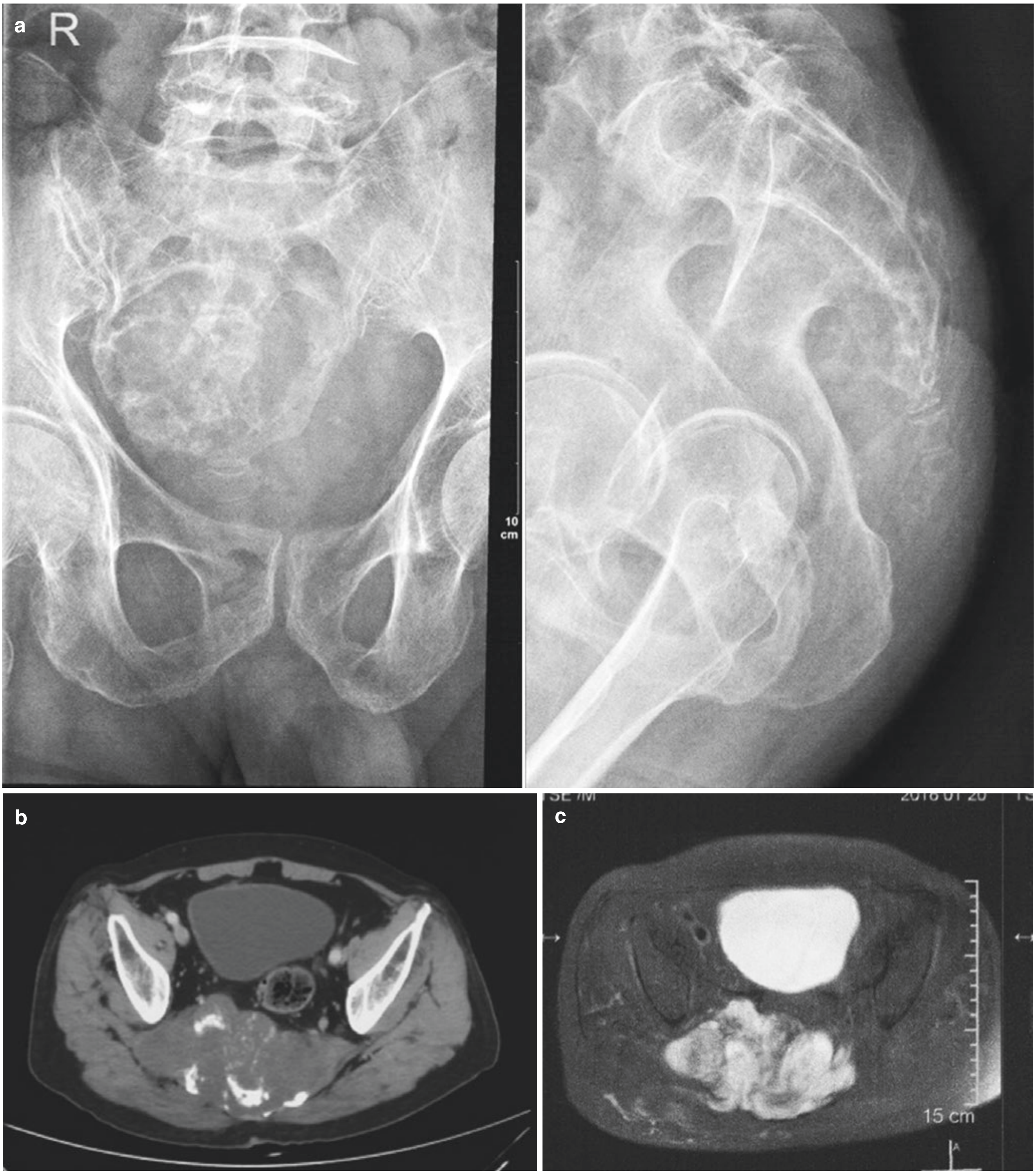

Fig. 22.5 Imaging studies obtained in a 74-year-old man with chordoma involving S2–5. (**a**) Anteroposterior and lateral radiograph revealing *a*n expanded lytic lesion *in sacrum*. (**b**) Axial CT scans demonstrating a large soft-tissue mass extending anteriorly to adjacent to the rectum and posteriorly to invade the buttocks; calcification is seen within the mass. (**c**) Sagittal fast-spin echo T_2-weighted MR images demonstrating the lesion infiltrating the presacral region

Fig. 22.6 Radiological studies obtained in a 34-year-old man with multiple myeloma. (**a**) Anteroposterior and lateral radiograph revealing a highly destructive lytic lesion involving total sacrum. (**b** and **c**) Axial CT and MRI scan confirming involvement of total sacrum, as well as a huge-sized soft-tissue mass. Plasmacytomas may be very large and elicit no osseous reaction, as in this case

infiltrative pattern, or very occasionally multiple sclerotic lesions. The earlier solitary form, plasmacytoma, can be more difficult to diagnose because it has a less distinctive appearance. Plasmacytoma is usually seen as a large, expanded lytic lesion. There may be an associated small- to moderate-sized soft-tissue mass. Magnetic resonance imaging is nonspecific, with low to intermediate signal intensity on T_1- and high signal intensity on T_2-weighted images. Plasmacytoma eventually progresses to multiple myeloma (Fig. 22.6a–c).

22.2.3 Primary Lymphoma of Bone

Primary lymphoma of bone is a rare lesion but is listed as the third most common primary malignant neoplasm of the sacrum [3]. It presents most frequently in the second through fifth decades. It may appear as either an aggressive lesion causing prominent bone destruction, or the osseous structures may remain with an intact appearance but with apparent permeation because there is a large associated soft-tissue mass. Magnetic resonance imaging is nonspecific, as are other imaging techniques (Fig. 22.7a–c).

Fig. 22.7 Radiological studies acquired in a 60-year-old woman. MRI and CT scan demonstrated that there is both an anterior and epidural soft-tissue mass (*arrows*). In a patient of this age, an aggressive sacral lesion most frequently is a lymphoma. (**a**–**c**) Axial CT and Sagittal MRI scan demonstrated that there is a huge soft-tissue mass involving anterior sacrum

22.2.4 Ewing Sarcoma and PNET

Ewing sarcoma and PNET are pathologically distinct lesions that have similar clinical presentations and radiographic appearances. They represent the fourth most common primary malignant tumor of the spine overall [3] and are most frequent in children. They involve the spine primarily in 3–10% of cases but even more frequently involve the spine with their osseous metastases. Within the spine, the sacrum is the most common site of involvement [1]. The age range for Ewing sarcoma is 5–30 years, with 75% occurring in the first two decades. The male/female ratio is 3:1.

Ewing sarcoma/PNET is a highly destructive lesion involving a large soft-tissue mass. The osseous portion may be completely lytic, or portions may be quite sclerotic. The sclerosis has been related to prominent host-reactive bone formation. In one study [10], the author related it to osteonecrosis as well, stating that diffuse sclerosis is found in 69% of spinal lesions. Magnetic resonance imaging in Ewing sarcoma is nonspecific, with intermediate signal on T_1- and intermediate to high signal on T_2-weighted images. Primary therapy in cases of Ewing sarcoma/PNET is radio- and chemotherapy. Many patients, however, require decompressive surgery and stabilization. Unfortunately, these lesions are associated with the worst prognosis when they occur in the sacrococcygeal region, with local control being accomplished in only 62.5% and long-term survival in only 25% [1] (Fig. 22.8a–d).

22.2.5 Chondrosarcoma

Chondrosarcoma is the most common primary malignant bone neoplasm in adults. The spine is the primary site of involvement in only 3–12% of chondrosarcomas, and involvement of the sacrum is unusual [11]. Chondrosarcoma occurs most frequently in patients ranging in age from 30 to 60 years and has a male predominance (2–4:1). At its initial presentation, it is usually low grade [1].

Chondrosarcoma may show mild bone destruction, with extension into the soft tissues. There is usually (although not invariably) chondroid matrix present, which has been described as punctuate, ring, or arclike or having the appearance of "Cs or Js." It should be noted that even in the presence of matrix, the diagnosis of a sacral lesion is more likely to be chordoma than chondrosarcoma, simply because of the relative prevalence of chordoma (Fig. 22.9a–d).

In cases of chondrosarcoma, MR imaging demonstrates low to intermediate signal intensity on T_1-weighted and very high signal intensity on T_2-weighted sequences. The high T_2-weighted signal intensity may have a lobular configuration that can be distinctive for chondrosarcoma. If the matrix is a prominent feature, these regions will show low signal on all sequences.

Treatment of chondrosarcoma is wide excision. Because there are a high recurrence and mortality rates if the resection is incomplete, complete cross-sectional imaging should be performed prior to surgery to evaluate the operative site. Radiotherapy and chemotherapy are used if the surgical margin is not clear, but their effectiveness in low-grade lesions is not proven. Metastases generally occur late and involve the lung and bone.

22.2.6 Osteosarcoma

Osteosarcoma is the most common primary malignant osseous neoplasm in children and has a bimodal age distribution. It is, however, uncommon in the spine. It is the fifth most common primary malignant neoplasm of the sacrum, accounting for only 4% of primary sacral tumors [3]. Many of the osteosarcomas that do occur in the sacrum are secondary to degeneration of Paget disease.

Paget disease occurs in 10% of the individuals greater than age 80 years. Sarcomatous degeneration occurs in less than 1% of these patients, with the prevalence increasing with patient age and number of Paget disease-affected bones. Osteosarcoma is the most frequent sarcoma arising from Paget disease, but chondrosarcoma, malignant fibrous histiocytoma, and fibrosarcoma can also occur.

Paget disease-associated osteosarcoma in the spine may appear lytic in up to 50% of cases [3]. If the amorphous matrix of aggressive osteoid is present, the diagnosis is obvious (Fig. 22.10a–e). In either case, however, other bonesdemonstrating the enlargement and coarsened trabeculae typical of Paget disease should make the diagnosis straightforward.

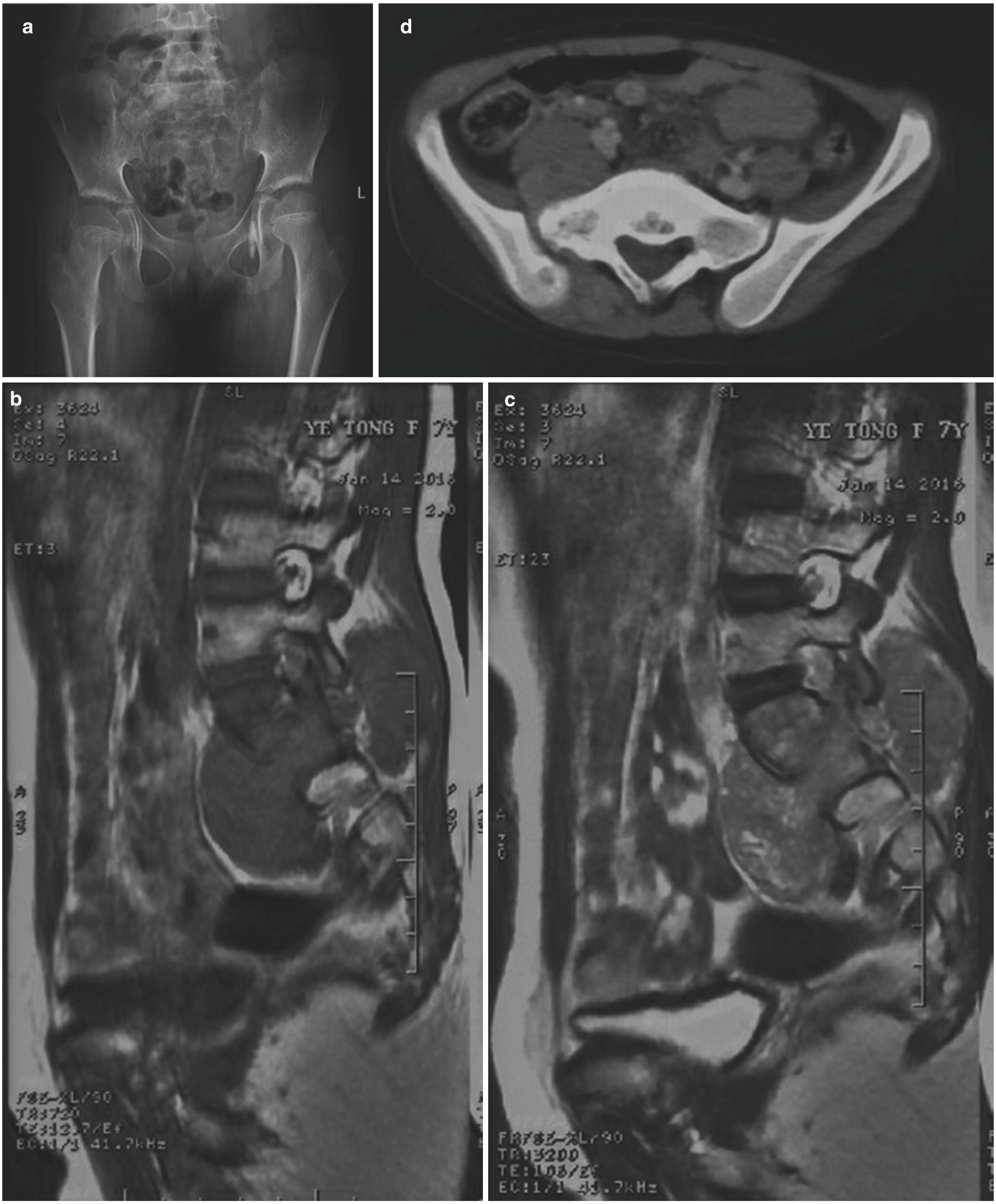

Fig. 22.8 Radiological studies acquired in a 6-year-old girl. (**a**) Anteroposterior radiographs of the sacrum revealing sclerosis of the right side of S1–2. There are no permeative or destructive changes seen. (**b–d**) MRI and CT scan, however, demonstrated that there is both an anterior and epidural soft-tissue mass (*arrows*). In a patient of this age, an aggressive sacral lesion most frequently is a Ewing sarcoma. In this case, the sclerosis is due to reactive bone formation, a common type of host bone reaction to Ewing sarcoma

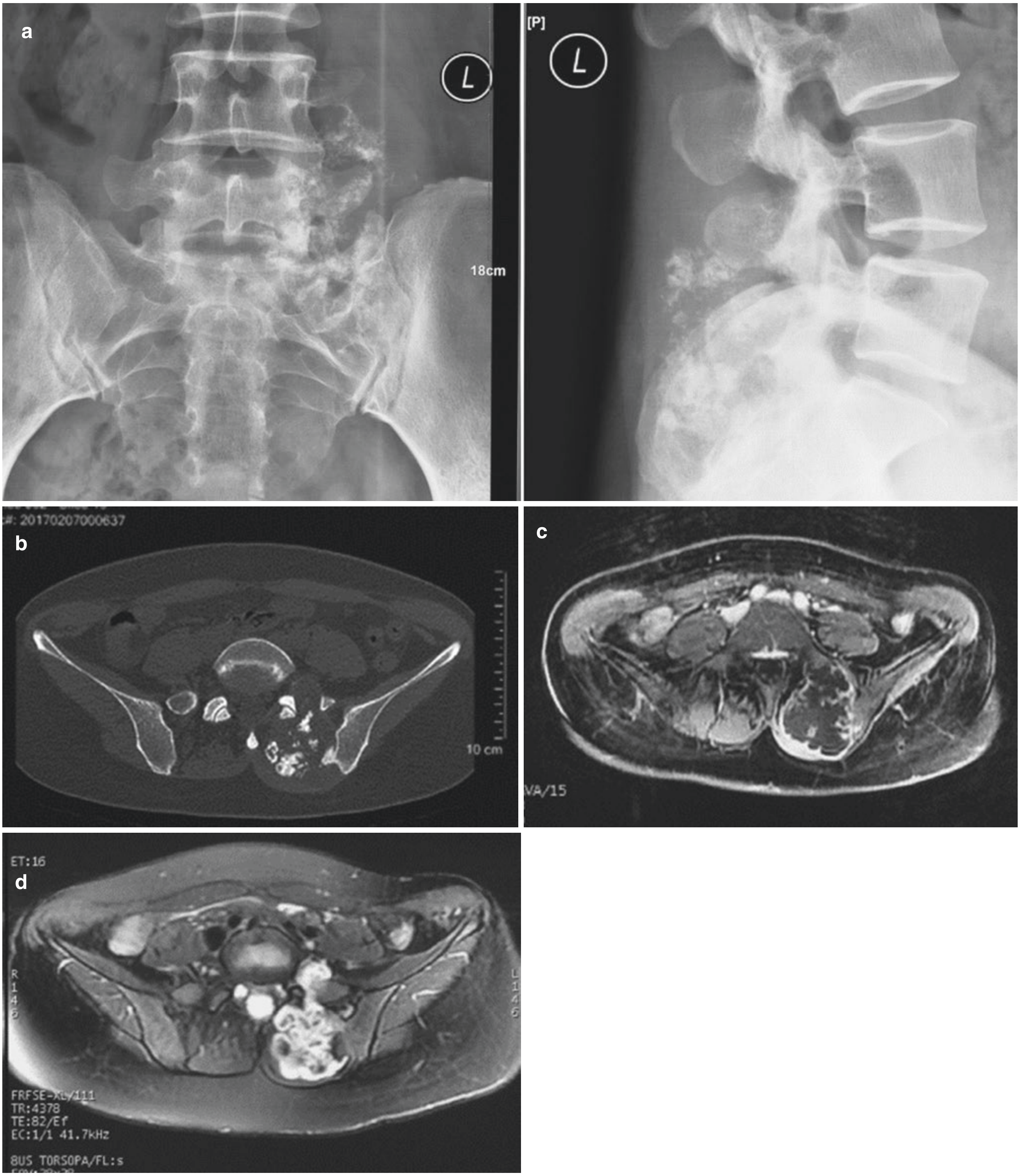

Fig. 22.9 Imaging studies acquired in a 37-year-old woman with chondrosarcoma. (**a** and **b**) Anteroposterior and lateral radiographs and CT scan demonstrating a highly destructive lesion involving L4–S2, with a large soft-tissue mass, which contains chondroid matrix (*arrows*). These features lead to diagnosis of a chondrosarcoma. (**c** and **d**) Axial T_1-weighted (TR/TE 600/9 ms) image revealing a tremendous low signal soft-tissue mass extending well into the back. Axial T_2-weighted fast-spin echo MR images with the patient in a prone position, demonstrating that the mass extends out into soft tissue. The very high signal intensity and lobulated nature of chondroid lesions can be depicted particularly well

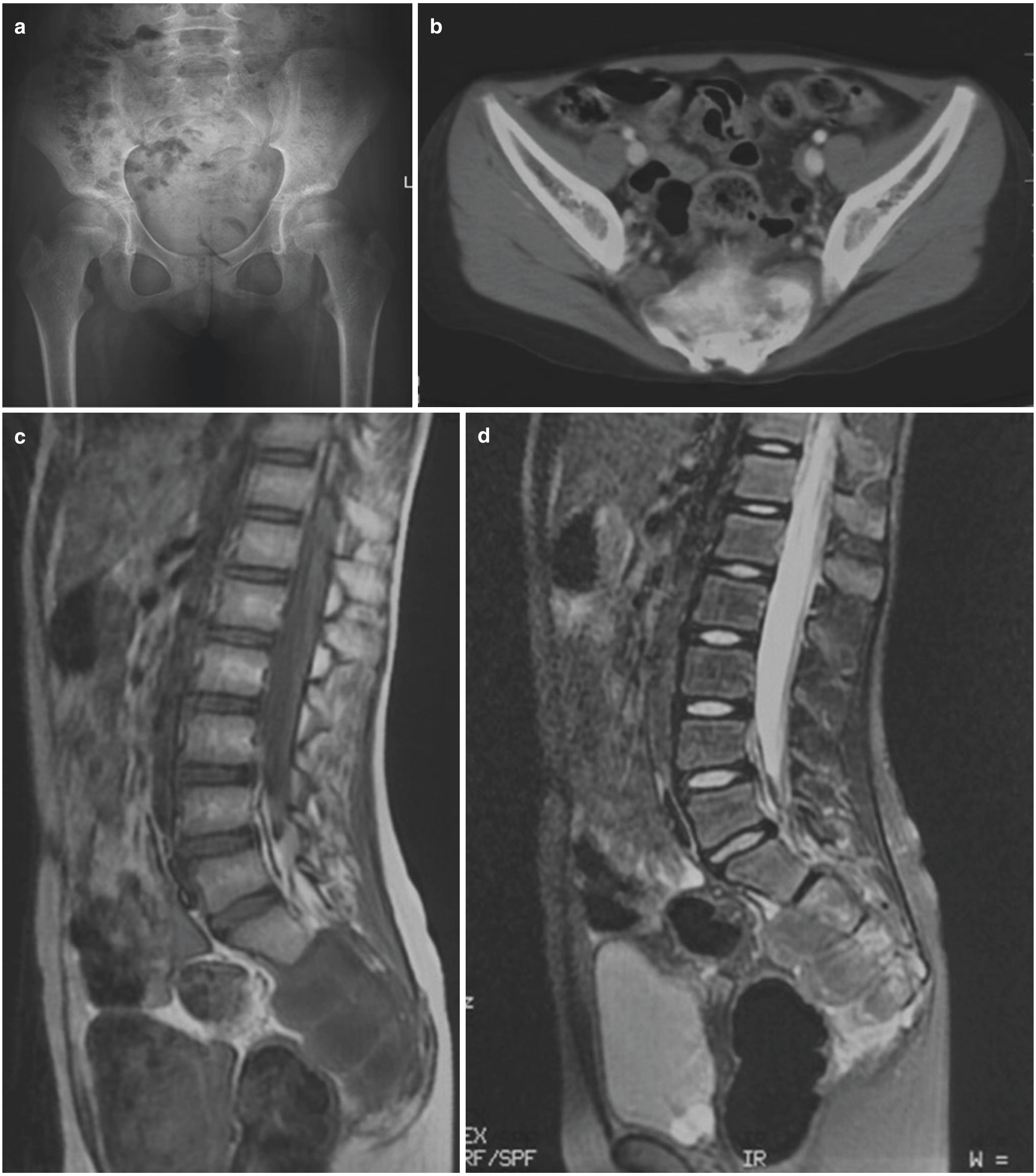

Fig. 22.10 Radiographs obtained in an 11-year-old girl with sacral osteosarcoma. (**a**) Anteroposterior radiographs demonstrating the amorphous bone density replacing the sacrum. (**b**) CT scan showed no remnant of normal bone in that region, and amorphous bone production occurs in osteosarcoma. (**c** and **d**) MR imaging demonstrates low signal intensity on T_1-weighted and also on T_2-weighted sequences. (**e**) Tc bone scan showed a highly predictable osseous distribution in sacral region

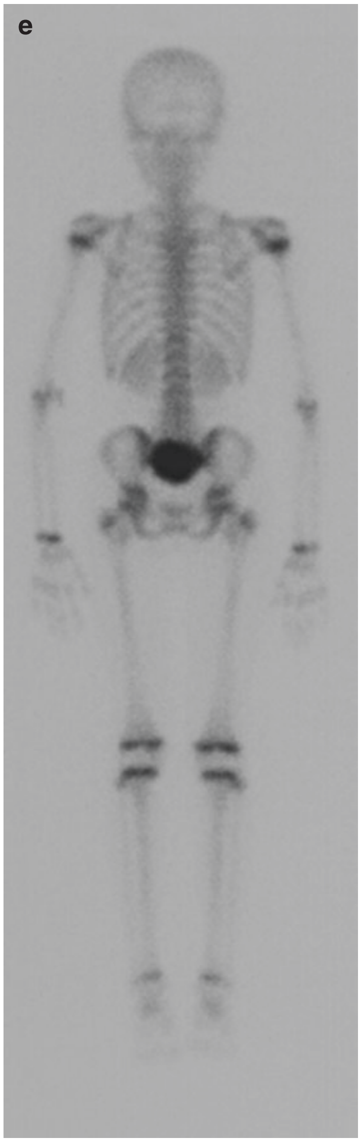

Fig. 22.10 (continued)

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23 Tumors of the Sacrum: Pathologic Aspect

Yi Yang and Wei Guo

The sacrum is composed of the bone, cartilage, soft mesenchymal tissue, bone marrow, and notochordal remnants, which give rise to tumors or pseudotumors. Tumors of the sacrum include primary and metastatic/systemic origin. The primary tumors consist of malignant and benign entities. Systemic diseases include metastases and hematopoietic malignancies such as lymphoma, multiple myeloma, or plasmacytoma.

23.1 Malignant Tumors

Malignant tumors include primary sarcoma, primary hematopoietic malignancy, and metastatic diseases. Primary sarcoma may derive from bone or soft tissue. The histological type of a primary bone sarcoma is often indicative of the tumor grade. The most common bone sarcomas are of high-grade malignancy. Soft tissue sarcoma occurring in the sacrum is rare. Histological type and grade predict tumor behavior. The Fédération Nationale des Centres de Lutte Contre Le Cancer (FNCLCC) grading system is well accepted [1].

23.1.1 Chordoma

Chordoma is the most common primary malignancy occurring in the sacrum arising from notochordal rests [2]. Although it is mainly located in the base of the skull, 29.2–60% of chordomas occur in the sacrococcygeal region [3, 4]. It is usually a slow-growing and low-grade tumor, but metastatic disease is seen more frequently in sacral chordomas than in skull base chordomas [5].

Grossly, the tumor has an expansile, lobulated structure with cortical invasion. The cut surface is gelatinous with chondroid texture (Fig. 23.1). By definition, chordoma is a malignant tumor showing notochordal differentiation [3]. Notochordal differentiation is exhibited by epithelioid cells arranged in nests or cords with clear or eosinophilic cytoplasm; some have vacuolated "bubbly" cytoplasm, so-called physaliphorous cells. The tumor cells are separated by fibrous septa, which give rise to the lobulated appearance

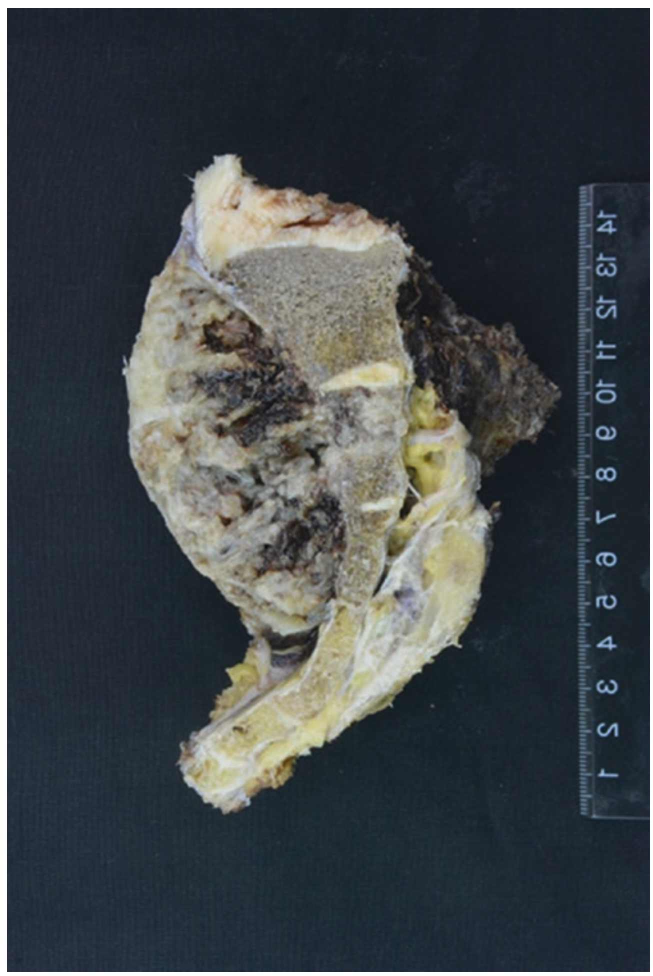

Fig. 23.1 Gross image of a sacral chordoma

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and are embedded in abundant extracellular myxoid matrix. In a low-grade tumor, the nuclei are small with coarse chromatin. In a high-grade tumor, the nuclei may become larger and pleomorphic with greater mitotic activity.

Most of the chordomas are designated as chordomas, not otherwise specified (NOS) [3]. Chondroid chordoma is a rare variant which contains hyaline cartilage component. Its behavior is similar to chordoma, NOS, but it may be confused as chondrosarcoma morphologically. Brachyury is a specific immunohistochemical diagnostic marker for chordoma [6]. Nuclear immunoreactivity to brachyury is seen in chordoma but not in chondrosarcoma and is therefore extremely helpful in the differential diagnosis. Figure 23.2 shows the histological and immunohistochemical features of a chordoma. In practice, we prefer to use the more specific monoclonal antibody of brachyury than the polyclonal to prevent false positivity; we also prefer to use non-decalcified specimen for brachyury testing to prevent false negativity. Other traditional helpful positive diagnostic immunohistochemical markers for chordoma include keratin, epithelial membrane antigen, and S-100 protein. New markers such as loss of PTEN and loss of INI-1 expression have recently been found in chordoma [7, 8]. Dedifferentiated chordoma is a high-grade and biphasic tumor which consists of a high-grade undifferentiated spindle cell sarcoma or osteosarcoma in association to chordoma, NOS [3]. Recognizing the conventional chordoma component is the key to this diagnosis because the dedifferentiated component does not express the diagnostic markers described here.

23.1.2 Chondrosarcoma

Chondrosarcoma is a locally aggressive malignant tumor that produces cartilaginous matrix [3]. There are four histological variants of chondrosarcoma: conventional, dedifferentiated, mesenchymal, and clear cell (Table 23.1). The histological grade is the single most important prognostic factor of conventional chondrosarcoma. Chondrosarcoma can be classified on the basis of its location in the bone.

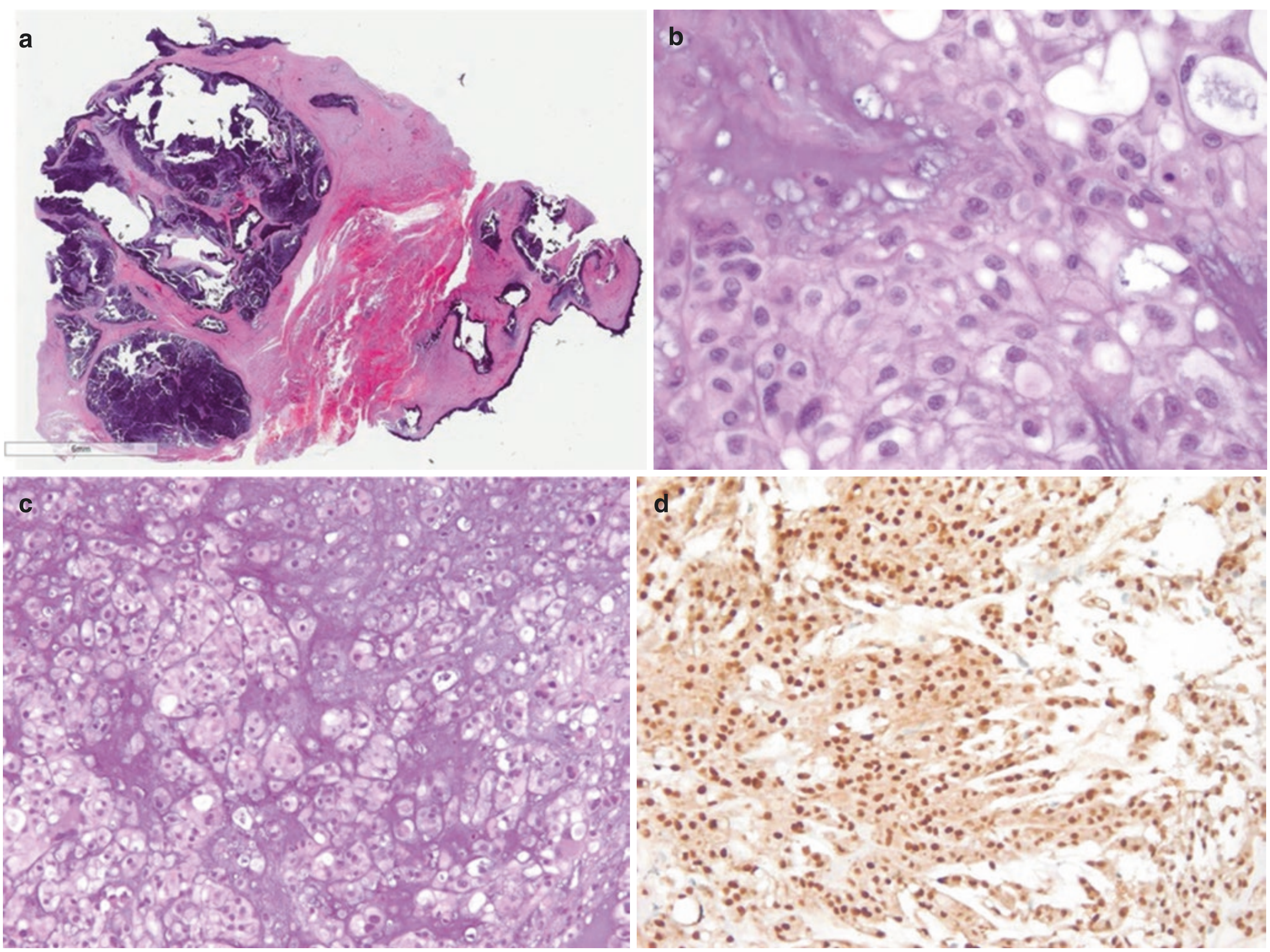

Fig. 23.2 Microscopic images of a chordoma. (**a**) HE digitalized whole slide image. (**b**) HE ×200. (**c**) HE ×600. (**d**) Brachyury stain ×200

Table 23.1 Characteristics of histological variants of chondrosarcoma

Tumor type	Component	Prognosis
Conventional	Chondrosarcoma	Depends on grade
	Grade I	
	Grade II	
	Grade III	
Dedifferentiated	Low-grade conventional chondrosarcoma plus high-grade dedifferentiated sarcoma or osteosarcoma	Poor
Mesenchymal	Low-grade conventional chondrosarcoma plus poorly differentiated malignant small round cells	Poor
Clear cell	Clear cells or chondroblastoma-like cells	Depends on grade
	Note: Usually occurs in the ends of long bones; patients younger than conventional	

Central chondrosarcomas are located in the medullary cavity, peripheral chondrosarcomas arise from the surface of the bone, and periosteal (juxtacortical) chondrosarcomas arise from the surface of the bone and the periosteum [3]. According to its origin, a primary chondrosarcoma arises de novo, and secondary chondrosarcoma is a result of malignant transformation of an enchondroma (central) or osteochondroma (peripheral). All types can affect the pelvic bones, including the sacrum; however, peripheral secondary chondrosarcoma is seen more commonly in younger patients than in central primary chondrosarcoma, which predominantly affects patients more than 50 years of age [9]. The majority of the conventional and dedifferentiated chondrosarcomas exhibit somatic mutations of the *isocitrate dehydrogenase genes 1 and 2* (*IDH1 and IDH2*) [10]. However, this finding is absent in mesenchymal and clear cell chondrosarcoma indicating their different pathogenesis. The presence of (*IDH1 and IDH2*) mutation can be used to differentiate a chondroblastic osteosarcoma when it is deemed necessary. These molecular findings also warrant further investigation for their role as potential therapeutic targets [11].

As shown in Fig. 23.3, conventional chondrosarcomas grossly have the cut surface of hyaline cartilage with irregularly lobular appearance. Myxoid, cystic, and calcification changes can be seen. Microscopically, the distinction between enchondroma and grade I chondrosarcoma can be challenging due to overlapping morphological features. A generally accepted minimum diagnostic criteria for chondrosarcoma include hypercellularity, permeation of the host bone, absence of host bone encasement, open chromatin, mucoid matrix, and older patient (age > 45 years) [3]. After establishing a diagnosis of chondrosarcoma, the next step is to grade the tumor using the following histological features: cellularity, nuclear size, degree of hyperchromasia, and

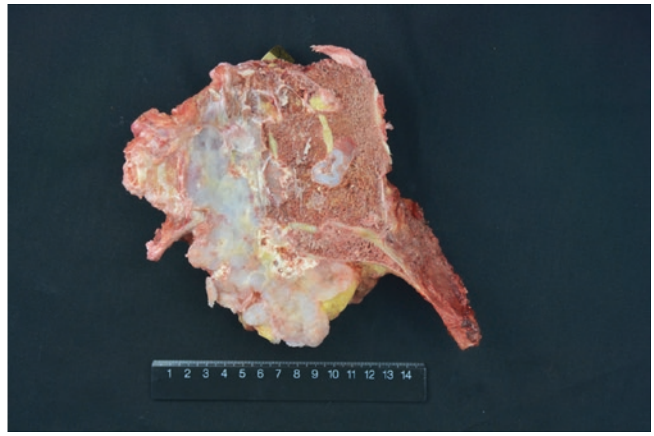

Fig. 23.3 Gross image of a sacral chondrosarcoma

mitoses. The grade I chondrosarcoma has similar nuclear features of enchondroma, except the architectural changes as described above. Grade III chondrosarcoma exhibits high cellularity, markedly enlarged nuclei, pleomorphic nuclei with nucleoli, and frequent mitoses compared to grade II chondrosarcoma. Tumor grade is the single most important prognostic factor of chondrosarcoma [9, 12]. When a chondrosarcoma has a spectrum of histology from grade I to grade III, it is a good practice to report the percentage of the high-grade component which predicts a worse prognosis. Figure 23.4 is the histological illustration of chondrosarcoma of various grades and dedifferentiated chondrosarcoma.

23.1.3 Ewing Sarcoma

Ewing sarcoma is a high-grade malignancy with small, round tumor cells harboring pathognomonic molecular signatures [3]. Approximately 85% of the Ewing sarcoma harbors a somatic chromosomal translocation t(11;22)(q24;q12) which rearrange *EWSR1* gene to fuse with *FLI1* gene [13]. The fusion protein EWSR1-FLI1 is an oncoprotein and is responsible for the pathogenesis of Ewing sarcoma [14, 15]. The *EWSR1* gene also has many other fusion partners such as the *ERG* gene [13]. Molecular testing for the signature gene and products is useful in confirming the diagnosis [16, 17]. While reverse transcription polymerase chain reaction (RT-PCR) may confirm the presence of *EWSR1-FLI1* or *EWSR1-ERG* fusion products, specific for Ewing sarcoma, the detection of rearrangement of *EWSR1* gene by fluorescence in situ hybridization (FISH) is not specific, because other sarcomas may harbor *EWSR1* gene rearrangements [3].

Primary Ewing sarcoma of the spine including sacrum is uncommon (only 3–10%), while metastatic disease from extraspinal Ewing sarcoma is more frequent. The sacral ala is the most common site for primary Ewing sarcoma of the spine [18, 19]. The prognosis is worse for sacrococcygeal

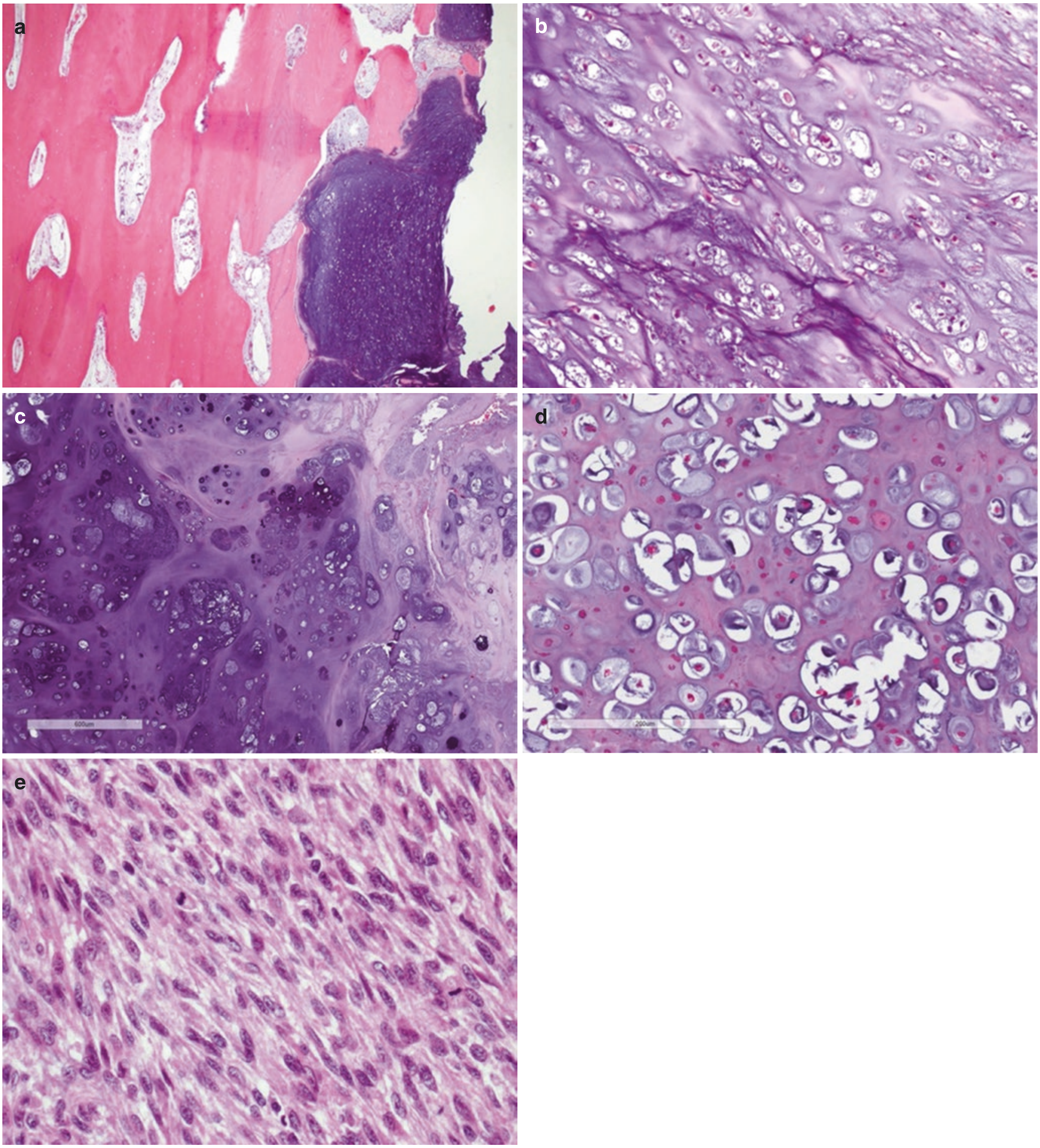

Fig. 23.4 Microscopic images of chondrosarcoma. (**a**) Chondrosarcoma invasion of bone. HE ×40. (**b**) Grade I chondrosarcoma. (**c**) Grade II. HE ×200. (**d**) Grade III. (**e**) Dedifferentiated chondrosarcoma. HE ×400

Ewing sarcoma than for extraspinal Ewing sarcoma, usually due to larger tumor size at presentation because of delayed clinical presentation [20].

Grossly, the tumor has tan-gray cut surface with no bone or cartilaginous matrix. Necrosis and hemorrhage can be seen. In a classic Ewing sarcoma, the tumor is composed of small round cells with scant cytoplasm and round nuclei arranged in a vaguely lobular pattern or completely dyscohesive. This latter appearance resembles lymphoma. However, the cytoplasm of Ewing sarcoma appears clear and contains glycogen, which stains positively with periodic acid-Schiff (PAS). Ewing sarcoma also lacks the lymphoglandular bodies which represent cytoplasmic debris of lymphoma cells. In an atypical Ewing sarcoma, the tumor cells are larger with more pleomorphic nuclei and prominent nucleoli [3]. Neuroectodermal differentiation can be seen with tumor cells forming rosette-like structures. Immunohistochemical stain pattern of Ewing sarcoma includes positive vimentin, CD99 (membranous staining pattern), keratin (aberrantly expressed in 30% cases), neuroendocrine markers, FLI-1, and, rarely, ERG [13, 21, 22]. A histological, immunohistochemical, and molecular illustration of Ewing sarcoma is in Fig. 23.5.

23.1.4 Osteosarcoma

Patients with primary lumbosacral osteosarcoma are older at presentation and commonly males [20]. Secondary sacral osteosarcoma occurs in patients with previous radiation

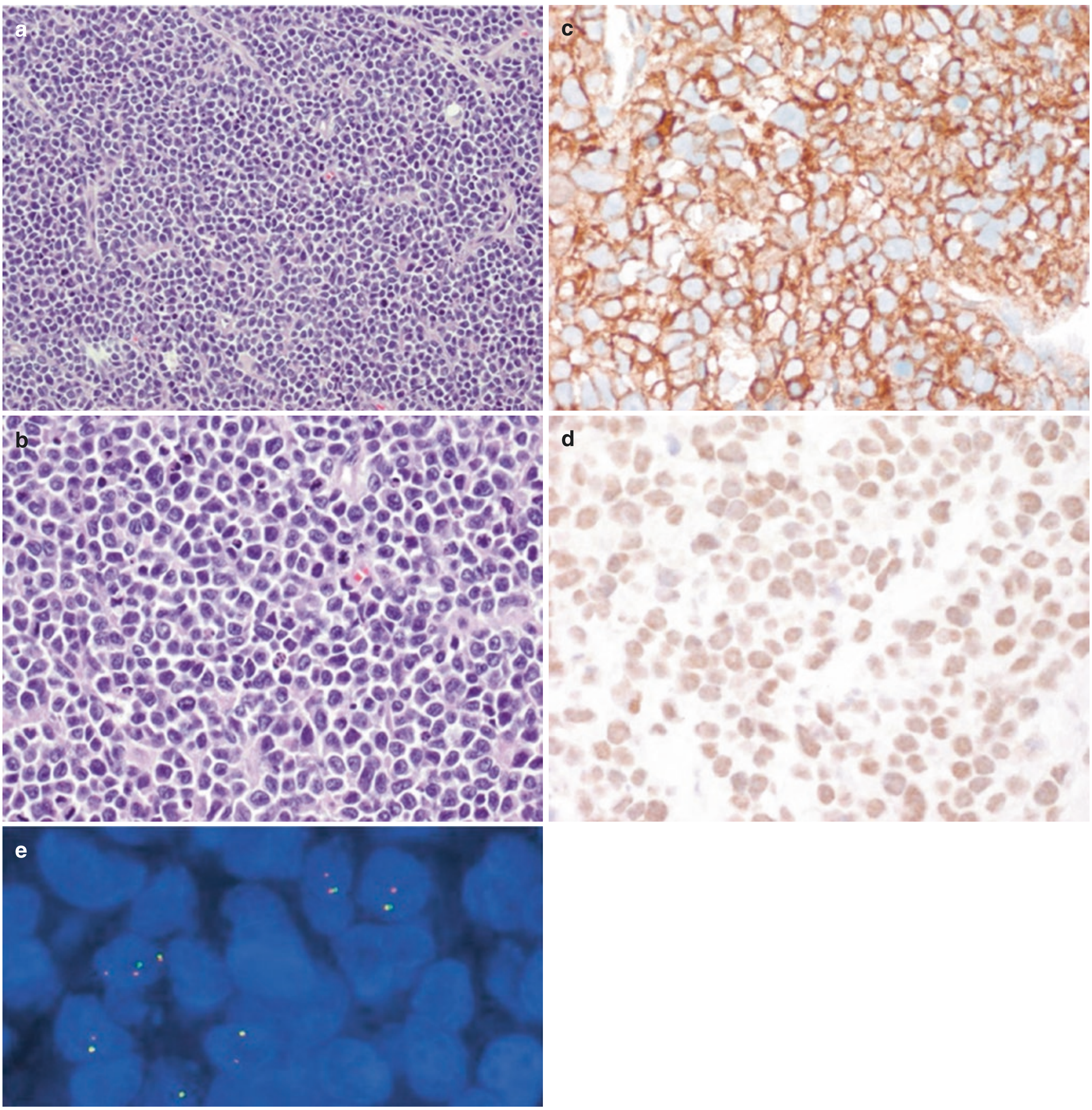

Fig. 23.5 Ewing sarcoma. (**a**) HE ×200. (**b**) HE ×400. (**c**) CD99+ ×400. (**d**) FLI1+ ×400. (**e**) FISH ×1000 showing LSI EWSR1 (22q12) break-apart probe showing EWSR1 rearrangement

treatment or a history of Paget's disease. Elderly patients with polyostotic Paget's disease are most at risk for sarcomatous degeneration [2].

According to its location in the bone, central osteosarcoma is located in the medullary cavity, and peripheral osteosarcoma arises from the surface of the bone [23]. The characteristics of primary central osteosarcoma and surface osteosarcoma are summarized in Tables 23.2 and 23.3. Surface/peripheral osteosarcoma very rarely affects the flat bone.

High-grade osteosarcoma is treated with neoadjuvant chemotherapy. The pathological evaluation of the therapy response is critically important for assessment of prognosis. Osteosarcomas with more than 90% tumor necrosis (less than 10% viable tumor cells) are considered good responders and have better overall and disease-free survival [24]. A generally accepted method of sampling osteosarcoma includes cross-sectioning the central and largest slice of the tumor. The slice is further divided into

Table 23.2 Characteristics of histological variants of primary central osteosarcoma

Tumor type	Component	Prognosis
Conventional	High-grade sarcoma with osteoid formation	High-grade tumor. Subtype does not differ in prognosis and therapy
	Osteoblastic (76–80%) (Fig. 23.6a)	
	Chondroblastic (10–13%) (Fig. 23.6b)	
	Fibroblastic (10%)	
Telangiectatic	High-grade osteosarcoma with characteristic blood lakes and spaces	Similar to conventional type
Giant cell rich	High-grade osteosarcoma with abundant osteoclast-like giant cells (Fig. 23.6c)	Similar to conventional type
Small cell	High-grade osteosarcoma with characteristic small tumor cells (Fig. 23.6d)	Slightly worse prognosis than conventional type
Low-grade central	Low-grade osteosarcoma	Excellent prognosis
	Note: Distinguish from fibrous dysplasia by permeation of the host bone and soft tissue extension; amplification of *MDM2* gene	

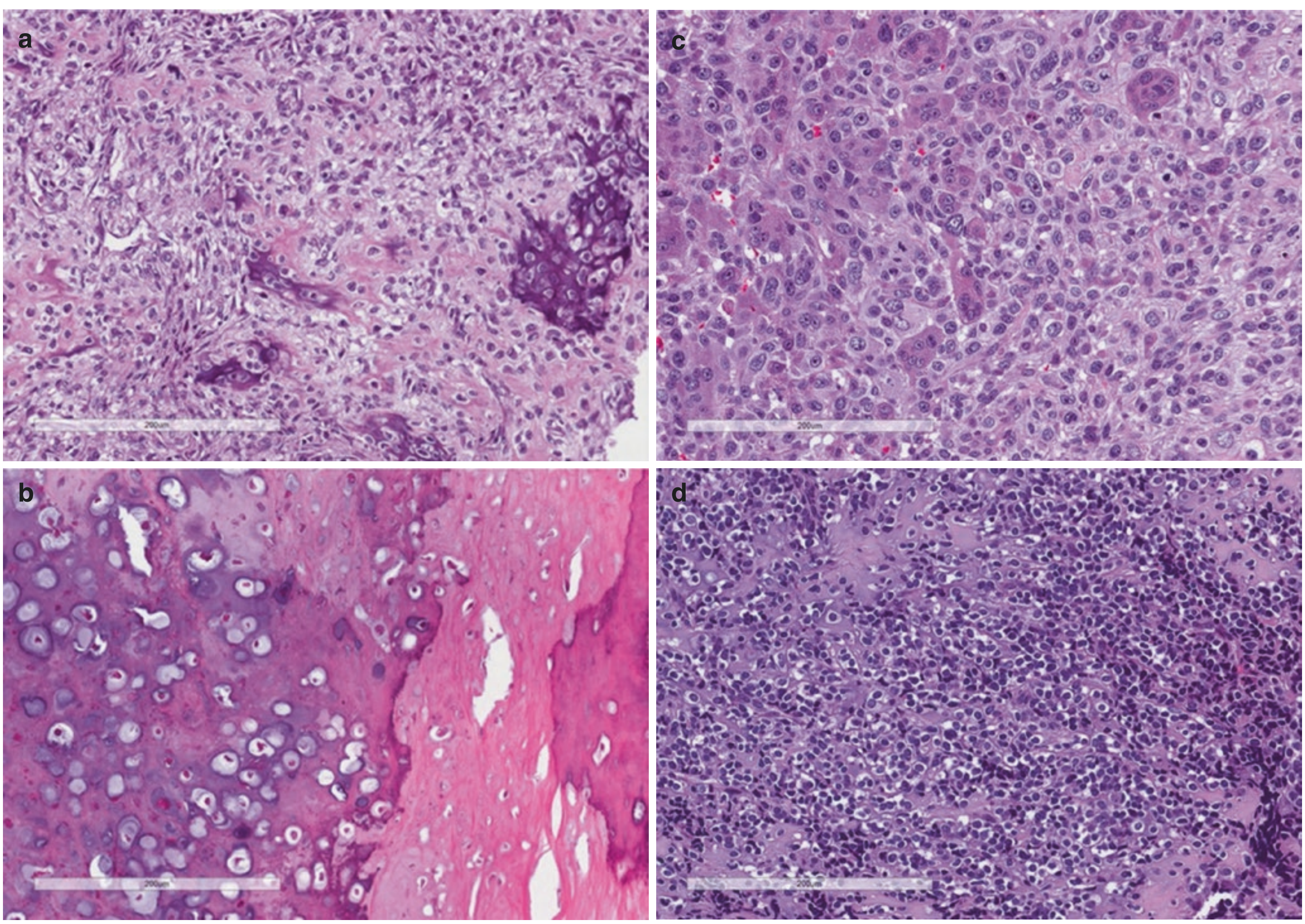

Fig. 23.6 Osteosarcoma. (**a**) Osteoblastic osteosarcoma. (**b**) Chondroblastic osteosarcoma. (**c**) Giant cell-rich osteosarcoma. (**d**) Small cell osteosarcoma

Table 23.3 Characteristics of histological variants of primary peripheral osteosarcoma

Tumor type	Component	Prognosis
Parosteal (juxtacortical osteosarcoma)	Low-grade	Excellent
	Spindle cells with mild to moderate atypia, well-formed bone trabeculae arranged in parallel pattern, and associated benign cartilaginous differentiation	
	Note: Amplification of *MDM2* gene	
Periosteal (juxtacortical chondroblastic osteosarcoma)	Intermediate-grade	Better prognosis than conventional osteosarcoma
	Predominantly atypical cartilage admixed with intermediate-grade osteosarcoma	
High-grade surface osteosarcoma	High-grade osteosarcoma of the surface	Similar to conventional type
	Note: The tumor is predominantly outside the bone; similar variants seen in conventional osteosarcoma	

Fig. 23.7 A gross image of undifferentiated sarcoma

1 cm × 1 cm slices and prepared for microscopic examination. Therapy-induced changes include tumor necrosis, pleomorphic changes, cystic changes, fibrosis, etc. However, only the percentage of tumor necrosis characterized by pyknotic, fragmented, or lysed tumor nuclei, which is reversely related to the percentage of viable tumor cells, is accepted as an independent prognostic factor [25].

23.1.5 Undifferentiated Pleomorphic Sarcoma

Undifferentiated pleomorphic sarcoma (UPS) is a group of high-grade tumors that have no identifiable line of differentiation when analyzed by current technologies and therefore represents a diagnosis of exclusion. Figure 23.7 shows a gross image of a sacral undifferentiated sarcoma. UPS histology is variable and may show several morphologic patterns from storiform areas composed of spindle cells to areas composed of large, pleomorphic neoplastic cells with marked nuclear atypia [3, 26]. Mitotic activity is prominent with atypical mitotic figures.

23.1.6 Metastatic/Systemic Malignancy

Metastatic disease from epithelial malignancies is the most common secondary malignancy of the sacrum [2]. The primary sites include the lung, breast, prostate, kidney, head and neck, and gastrointestinal tract. Melanoma is also a common culprit of metastasis [27]. The most common hematopoietic malignancies of the sacrum are non-Hodgkin lymphoma and multiple myeloma or plasmacytoma. These diseases may be either primary of bone or secondary involvement of the bone in disseminated disease. Our institutional review of primary bone lymphoma (PBL) consisted of 70 patients [28, 29]. PBL cases were included in this cohort using the 2013 WHO criteria for bone/soft tissue tumors [3], as disease was restricted to bone and adjacent soft tissue with or without regional nodes at the time of the diagnosis. Bone lymphoma with distant bone marrow involvement as the only other site of extranodal disease was also included. We found that PBL occurs in the sacrum less frequently than extremities, but diffuse large B-cell lymphoma is the most common variant of lymphoma.

When a primary tumor is present, the diagnosis of metastatic disease is achieved by comparing the histology of sacral lesion with the primary disease. However, when a primary site unknown histomorphology in conjunction with pertinent ancillary testing including immunohistochemistry, flow cytometry and molecular testing are used to render a definitive diagnosis.

23.1.7 Rare Primary Sacral Sarcomas

The following malignant tumors have occasionally been reported occurring in the sacrum [3, 30–32] (*see* Table 23.4).

Table 23.4 Rare primary sarcomas of the sacrum

Tumor type	Definition	Histology and immunophenotype	Prognosis
Undifferentiated/ unclassified sarcoma	High-grade malignancy showing no identifiable specific lineage of differentiation	Pleomorphic, spindle cell, round cell, epithelial cell	Limited data
		No consistent finding	
Angiosarcoma	Aggressive malignancy with endothelial differentiation	Epithelial, spindle	Poor prognosis associated with high grade, presence of macronucleolus, older age, large size, increased mitoses, and high Ki-67 index
		Express vascular markers (CD34, CD31, ERG, and FLI1)	
Fibrosarcoma	Intermediate- to high-grade fibroblastic spindle cell malignancy	Less pleomorphic than undifferentiated pleomorphic sarcoma	Depend on age, tumor site, grade, and stage
		Rare report of slerosing epithelioid fibrosarcoma	
		No consistent finding	
Malignant solitary fibrous tumor	Malignant variant of solitary fibrous tumor	Large tumor size, infiltrative margin, hypercellularity, nuclear pleomorphism, tumor necrosis, and high mitotic activity (>4/10 HPF)	Depend on age, tumor site, grade, and stage
		Express STAT6 (nuclear), CD34, CD99, and Bcl-2	

23.2 Benign Tumors

23.2.1 Giant Cell Tumor of Bone

Giant cell tumor of bone (GCTB) is a benign but locally aggressive tumor. The tumor is composed of numerous characteristic giant cells which are large and osteoclast-like. These cells are impressive morphologically; however, they are the background cells reactive to the true neoplastic cells which are primitive mesenchymal stromal cells. The neoplastic cells are mononuclear and express receptor activator for NF-κB ligand (RANKL), the master regulator of osteoclast differentiation. Macrophages and osteoclasts express RANK. The interaction between the neoplastic mononuclear stromal cells and macrophages/osteoclasts by a RANKL-dependent mechanism via the stimulation of macrophage-colony stimulation factor (MCSF) results in neoplastic proliferation and induces osteoclast formation. During this process, tumor-associated macrophage-like osteoclast precursors, which are also mononuclear cells, are recruited by tumoral stromal cells to participate in osteoclast differentiation and activation. Because osteoclast formation is the major consequence of GCTB, inhibition of osteoclast formation and activity is the key therapeutic approach. For example, bisphosphonate inhibits osteoclast-mediated resorption of bone/osteolysis and anti-RANKL antibody targets the RANKL-dependent mechanism of GCTB formation.

Osteoprotegerin (OPG) is a soluble decoy receptor that is produced by osteoblasts to inhibit osteoclast differentiation through its binding to RANKL, which prevents RANK binding. OPG expression reflects a protective mechanism of the skeleton to compensate increased bone resorption. Bone remodeling is mainly controlled by the balance of RANKL/OPG. Osteoprotegerin ligand (OPGL), also named receptor activator of RANKL, is also expressed in the stroma-like tumor cells of GCTB. The ratio of OPGL/OPG by tumor cells may contribute to the degree of osteoclastogenesis and bone resorption [33].

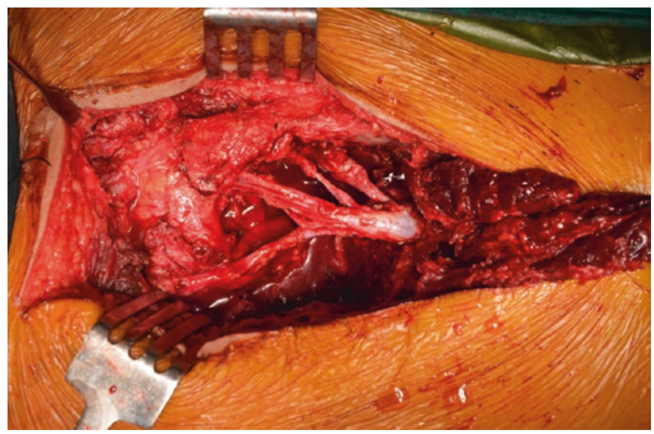

Fig. 23.8 A gross image of a sacral giant cell tumor after curettage

Although giant cell tumors of the bone (GCTB) within the vertebrae are rare (2.7–6.5% of all GCTB), the sacrum may be the most common spinal site for this lesion [34–36]. In a collaborative study with Beijing Jishuitan Hospital, we found that GCTB has significant higher incidences than the Mayo Clinic group [37]. JST group also published an article that described two GCTBs of the sacrum with pulmonary metastasis [38].

Grossly, the tumor is red-brown with hemorrhage (Fig. 23.8). Yellow areas reflect lipid-laden macrophage-rich areas. Histologically (Fig. 23.9) the tumor is composed of numerous giant cells with multinucleation and scattered

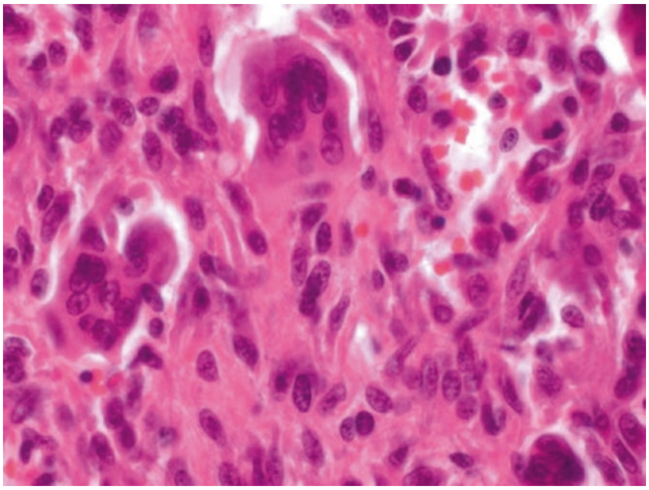

Fig. 23.9 Microscopic images of giant cell tumor

mononuclear cells that are round or spindle. Lipid-laden or hemosiderin-laden macrophages are also present. The tumor is mainly solid and may contain cystic areas. Secondary aneurysmal bone cyst component is seen in 10% of GCTB. The tumor may be mitotically active; however, a benign giant cell tumor typically does not have atypical mitosis or significant nuclear atypia. The latter is associated with a malignant transformation of GCTB. One diagnostic pitfall is to avoid misdiagnosing an osteosarcoma when a pathological fracture is in association with a malignant giant cell tumor.

Fig. 23.10 A gross image of a sacral schwannoma

23.2.2 Benign Neurogenic Tumor

Benign neurogenic tumors occur in paraspinal or presacral locations. Sacral schwannomas or neurofibromas grow within the sacral canal and only rarely expand through the anterior sacral foramina into the presacral space [39]. Schwannoma (Fig. 23.10) consists of well-differentiated Schwann cells and is usually encapsulated, and cut surfaces have a pink, white, or yellow appearance. Classic histology (Fig. 23.11a, b) shows a pattern of alternating Antoni A (cellular areas of spindle cells with occasional palisading) and Antoni B (loose myxoid areas with scattered spindle cells and thick-walled, hyalinized vessels) areas. Degenerative changes such as cyst formation, calcification, hemorrhage, and hyalinization may be present, especially if the tumor has been there for a long duration. Ancient schwannoma is characterized by Schwann cells with large and hyperchromatic nuclei, the manifestation of degenerative change. Schwannomas with increased cellularity and occasional mitoses are referred to as cellular schwannomas, a variant of schwannoma. These variants of schwannoma behave similar to conventional schwannomas. Immunohistochemically, schwannomas express strongly and diffusely S-100 protein as well as SOX10, a new marker of neural crest differentiation [40]. Neurofibroma, originating from sacral nerve roots, is composed of Schwann cells, perineurial-like cells, fibroblasts, and axons and is associated with a myxoid or fibrous stroma.

Malignant transformation can occur, often seen in the setting of neurofibromatosis type 1. Differentiation of benign and malignant neoplasms can be difficult, but increased size, rapid growth, infiltrative border, necrosis, increased cellularity, and increased mitotic activity with atypical mitosis favor malignancy. Cellular schwannoma can be misdiagnosed as malignant peripheral nerve sheath tumor (MPNST) (Fig. 23.11c). Morphologic features distinguishing cellular schwannoma from MPNST are the presence of perivascular accentuation of cellularity, tumor herniation into vascular lumens, presence of necrosis, and loss of expression of H3K27me [41].

Other rare neurogenic tumors include sacral ependymomas that arise from ependymal cells of the terminal filum, expand the sacral canal, and are usually of the myxopapillary type. Sacral meningiomas are even more rare than sacral ependymomas and schwannomas and arise within the sacral

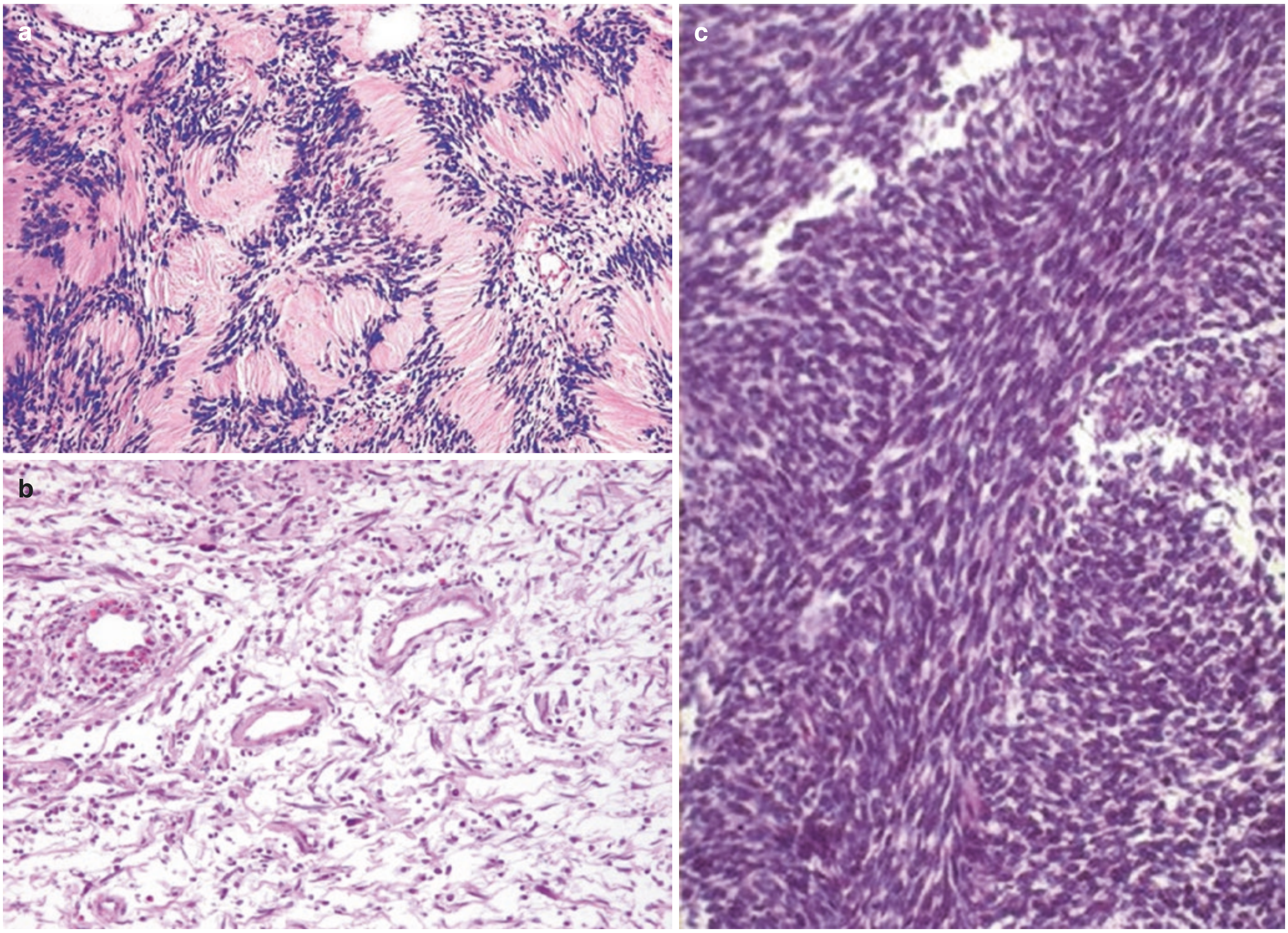

Fig. 23.11 Microscopic images of neurogenic tumors. (**a**) Schwannoma ×200 Antoni A. (**b**) Schwannoma ×200 Antoni B. (**c**) Malignant peripheral nerve sheath tumor

canal. Neuroblastoma is derived from embryonic neural crest tissue and clinically manifests in infancy. Ganglioneuroma is originated from sympathetic ganglion cells [39]. Ependymomas and meningioma have both benign and malignant variants.

23.2.3 Aneurysmal Bone Cyst

Aneurysmal bone cyst (ABC) is a benign tumor. Grossly it is well defined and composed of blood-filled pseudocystic spaces. The cysts lack specific cell lining and consist of a wall of spindle cells with scattered osteoclast-type multinucleated giant cells. The neoplastic cells are spindle shaped and indistinguishable from reactive fibroblasts and myofibroblasts. However, the tumor cells show *USP6* rearrangement in 70% of ABC [42]. The spindle cells are bland. Mitoses may be frequent without atypical forms. Reactive woven bone may be seen with osteoblastic rimming.

23.2.4 Other Rare Benign Primary Intraosseous Sacral Lesions

Although the true incidence has not been established, the lesions listed here (Table 23.5) rarely occur in the sacrum. The diagnosis is mainly based by histologic examination with clinicoradiological correlations. Ancillary testing is not widely used for diagnosis. The ancillary testing described in Table 23.5 is mostly for academic interest except for fibrous dysplasia and Langerhans cell histiocytosis.

23.2.5 Sacrococcygeal Teratoma

Sacrococcygeal teratoma is a germ cell tumor and the most common sacral tumor in neonates, although it is very rare in adults [39]. These tumors are composed of multiple tissues foreign to the tissue in which they arise, which usually include the skin, teeth, central nervous system tissue, and respiratory and alimentary mucosa. Mature teratoma (der-

Table 23.5 Rare benign primary intraosseous sacral lesions

Name	Nature	Histology	Ancillary testing for diagnosis
Chondroblastoma	Neoplastic	Round to polygonal chondroblasts with round to ovoid nucleus exhibit longitudinal grooves and well-defined cytoplasmic borders. Well. "Chicken wire" calcifications and multinucleated osteoclast-like giant cells are seen	S-100 protein and SOX9 expression by IHC
Chondromyxoid fibroma	Neoplastic	A lobular pattern with cellular stellate or spindle cells at the periphery and less cellular center with myxoid or chondromyxoid matrix. Multinucleated osteoclast-like giant cells, hyaline cartilage, and calcification are seen	S-100 protein and SOX9 expression by IHC
Osteochondroma	Neoplastic	Perichondrium, cartilage, and bone	None
Osteoid osteoma	Neoplastic	Nidus consisting of a combination of osteoid and woven bone surrounded by osteoblasts. The nidus is vascular rich with the appearance of granulation tissue. The nidus is surrounded by sclerotic bone	Runx2 and Osterix
Osteoblastoma	Neoplastic	Similar features as seen in osteoid osteoma	None
Fibrous dysplasia	Neoplastic	Bland spindle fibroblastic cells admixed with irregular bony spicules without osteoblastic rimming	*GNAS* mutation
Langerhans cell histiocytosis/ eosinophilic granuloma	Likely neoplastic	Langerhans cells are specialized histiocytes with nuclear grooves (reniform nuclei), which are admixed with inflammatory cells including prominent eosinophilia	CD1a, CD207/Langerin, and CD45 expression by IHC
Paget's disease	Nonneoplastic metabolic disorder of bone remodeling susceptible to deformities and fractures; increased risk to primary bone malignancy	Increase in osteoclastic resorption and secondary bone formation resulting in a disorganized and fragile lamellar bone mosaic pattern	None
	Monostotic or polyostotic		
Simple bone cyst	Nonneoplastic	Unilocular cysts of bone with a fibrous membrane lining. The cyst contains serous or serosanguineous fluid	None

moid cyst) is benign and most common. Struma ovarii is a rare form of mature teratoma that contains mostly benign thyroid tissue. Immature teratoma is uncommon which differs from mature teratoma by the presence of immature tissue and exhibits malignant clinical behavior.

23.2.6 Developmental Lesions

Sacral meningocele is a cerebrospinal fluid-filled protrusion of the meninges through a defect in the sacrum [39]. Benign sacral meningeal cysts are frequent coincidental findings in the radiological examination of the sacrum, and their pathogenesis is poorly understood.

23.2.7 Intraoperative Pathologic Evaluation of Bone and Soft Tissue Lesions

Intraoperative pathologic diagnosis of bone and soft tissue lesions is an important yet challenging tool in clinical musculoskeletal oncology practice. Indications for frozen section include making a diagnosis, evaluating margin status, determining tumor extent/spread, and obtaining an adequate sample for permanent section and diagnosis. Frozen section pathological evaluation provides real-time guidance to therapeutic intervention. In our practice, intraoperative cytology is used as an adjunct to frozen section. This approach has proven useful to enhance the accuracy of diagnosing bone and soft tissue lesion [43, 44], including sacral lesions.

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Preoperative Planning

24

Lu Xie and Wei Guo

All those surgeries of sacral sarcoma are defined as confined operation. The purpose of preoperative planning is to help surgeons get a better understanding of the surgery and help patients prepare themselves for the surgery and recover from it as soon as possible.

Before the preparation of the surgery, surgeons need to study the case discreetly and comprehensively through local plain film, computed tomography (CT), and magnetic resonance imaging (MRI), from which they could plan detailed surgical procedures and steps. Angiography should be undertaken if there might be involvement of the vessels [1]. Sacral tumors usually grow toward the presacral space and present with extremely large dimensions. Digital subtraction angiography (DSA) is thought to be not only a means of examination but also a treatment option, through which surgeons could not only observe the blood supply of the sacral tumors but also embolize the arteries which feed the pathological lesion with most appropriate embolic agent [2]. Occlusion of the vessels decreases the volume of the tumors, but multiple procedures are frequently needed [3].

Although the majority of these tumors are benign, they may become locally aggressive, causing catastrophic neurological impairment and weakening pelvic arch stability. For benign tumors, such as schwannoma, neurofibroma, giant cell tumor, and so on, the standard treatment is intralesional curettage followed by some other adjuvant therapy [4, 5]. However, for malignant or local aggressive tumors, like osteosarcoma, chordoma, chondrosarcoma, and so on, the initial en bloc resection of those tumors may give the patient the best chance of long-term survival [6]. However, surgical procedures might be complicated due to the complex anatomical characteristics of this region, the huge tumor volume, and the abundant blood supply. Besides, the requirement for adequate tumor removal must be balanced against the preservation of nerve function, which all influence surgical margins of those tumors and made local control of those tumors difficult. The conventional transperitoneal approach often requires retraction of abdominal organs for a long time during operation [7], which might had an impact on patients' postoperative recovering. In addition, it is difficult to acquire a sufficient clear absolute visual field to perform the surgery; thus, usually a larger incision was adopted, which resulted in longer rehabilitative time for the wound. Moreover, the lengthy retraction time might increase the possibility of organ or vessel damage. It is possible for surgeons to choose a single posterior approach to complete the surgery of sacrum with satisfactory result [2, 8–11]. Some doctors thought it might be more convenient to use one-stage anterior and posterior combined approach when patients presented with larger tumor [5, 9], while we thought it might not be necessary. With years of practice, the procedure with posterior-only approach brought us local recurrence rate of 18.9% in our 130 cases of sacral giant cell tumors, far lower than incidence of 47% reported in the literatures [12], which, in our opinion, relied on wide resection of most part of tumors and effective control of intraoperative hemorrhage. As we all know, enhanced local tumor control and overall survival could be acquired by wide resection of tumors despite potential complications and neurologic dysfunction [6, 13]. Total sacrectomy is indicated when a malignant or aggressive benign lesion involves the proximal sacrum with anterior extension [14]. Partial sacrectomy is used for tumors with substantial involvement of the sacrum below the S2 segment, which can usually be resected with wide margins and allows patients to maintain bowel and bladder functions without lumbopelvic reconstruction [5]. If complete resection could not be achieved easily, serial arterial embolization and radiation therapy are alternatives [15]; however, because of the limitations of technical merits, it is usually palliative with poor tumor control in China.

Massive retroperitoneal tumor usually involves the ureter, colon, rectum, and some other retroperitoneal viscera. And it is quite common to do urethral shunt, nephrectomy, as well as colectomy first before the actual surgery of the tumor.

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Given the possibility of tumorous involvement of the ureter, catheterization of double-J tube into the ureter of the affected side is necessary. It is appropriate to estimate the blood loss according to individuals' neoplastic condition and take actions to avoid complications. A consultation of general surgeons or urological surgeons is necessary if there is probability of colectomy or bladder bypass surgery. Not all the patients need colectomy; thus, we could not give a clear definition of operative indications of colectomy for sacral tumor. The independent risk factor for early-stage infection of these surgeries is urogenital bacterial and fungal infection, which should be taken into prevention before surgery [16].

24.1 Conventional Preparation

Preoperative communication with patients and their family members should focused on their expectations about surgical and anesthetic procedures, which mainly contributed to diminishing fear and anxiety and better understanding of postoperative recovery [17]. Face-to-face counselling, leaflets, or multimedia information containing explanations of the procedures along with tasks which contain encouragement might facilitate perioperative feeding, early postoperative mobilization, and pain control, which hence reduce the prevalence of complications [18]. Ideally, the patient should meet with the surgeons, anesthetists, and nurses one by one.

Preoperative evaluation should be used to identify medical conditions and risk factors for postoperative morbidity and mortality. Optimization of anemia, diabetes mellitus (DM), and hypertension improves outcomes [19–22]. It is necessary to have a comprehensive understanding of patients' general condition including mental and nutritional condition; the basic function of the heart, lung, liver, kidney, endocrine system, and blood; as well as immunity, from which we could find out various potential factors that may influence the surgery.

Before surgery, patients should be advised to stop excessive smoking or drinking. A recent review upon 11 randomized controlled trials (RCTs) involving 1194 patients concluded that smoking cessation during perioperative period appeared to contribute to reduce perioperative complications [23]. Several other studies have described the association between hazardous intake of alcohol and an increased morbidity because of postoperative infections, cardiopulmonary complications, or bleeding episodes [24].

A recent RCT [25] showed that "prehabilitation" (a program designed to increase functional capacity in anticipation of an upcoming stressor) addresses the impact that physical exercise might have on postoperative functional exercise capacity. The effect of such program regarding outcome remains to be evaluated [26]. Although we encouraged patients to get out of bed and walk as soon as possible after surgery, all those who went through surgeries of sacral tumors experienced huge trauma and sometimes were too weak to get out of bed postoperatively. Thus we need them to practice defecating and urinating at bed a few days before the surgery. What's more, some exercises to strengthen the muscle need to begin before surgery, which means a lot for patients in psychological acceptability [26].

It has been shown that pharmacological prophylaxis against venous thrombosis (VT) reduces the prevalence of symptomatic venous thromboembolism (VTE) without increasing side effects such as bleeding [27]. In addition, use of compression stockings reduces the incidence of VTE [28]. Patients with extensive comorbidity and malignant disease who are taking corticosteroids preoperatively and who have undergone previous pelvic surgery and those in hypercoagulable states have an increased risk of VTE. However, bleeding during or after the sacral surgery is also a life-threatening catastrophe for those patients [1, 13]. Thus prophylaxis against deep venous thrombosis of lower limbs may give way to preventing massive bleeding in the first 1–2 days after surgeries and these prophylactic treatment might be delivered 2–3 days later. In a recent Cochrane report based on four RCTs (n = 1021), it was concluded that prolonged (4 weeks postoperatively) VTE prophylaxis as compared with in-hospital prophylaxis was associated with a significantly reduced prevalence of VTE (14.3 vs. 6.1%, $p < 0.0005$), as well as symptomatic VTE (1.7 vs. 0.2%), without an increase in postoperative bleeding complications or other side effects [27]. We encourage the using of compression stockings to reduce the incidence of VTE. But as for pharmacological prophylaxis, it should depend on the detailed situation of each surgery and the general condition of each individual.

Considering the length of this chapter, we focus on the exceptional preparation for sacral surgeries.

24.2 Preoperative Bowel Preparation

Because of the long duration of sacral surgeries (usually longer than 6 h) as well as massive hemorrhage during operation (blood transfusion exceeding 2000 ml), the preoperative comprehensive evaluation is essential for those patients, mainly on metabolism and the cardiovascular and respiratory system. If the patients' condition is too weak to endure such a surgery, it is more appropriate for the patient to receive radiotherapy rather than to do the surgery. Insufficient preoperative preparation might increase intraoperative bleeding, which could further result in unclear surgical version and unsatisfactory surgical margins for those tumors, which of course further cause disappointed oncologic or functional prognosis. Sometimes even more seriously unsatisfactory preoperative preparation could indirectly lead to patient's death.

The most job of preoperative preparation is intestinal preparation, including using enema and oral laxatives. It is quite common to resect part of the small intestines or colorectum for a sacral surgery because of the neoplastic invasion or adhesion. However, mechanical bowel preparation (MBP) is associated with dehydration and changes in electrolyte balance (particularly in the elderly). A meta-analysis from studies focusing on colonic surgery showed no clinical benefit from MBP [29, 30]. Our patients usually stay at younger age than those with colorectal cancers, dehydration and electrolyte imbalance may be corrected more quickly. We usually combined oral laxatives and enema to prepare the intestinal in order to avoid postoperative wound or other infections. Fasting from midnight has been standard practice in the belief that this reduces the risk of pulmonary aspiration in elective surgery. What's more, our patients even started fluids 2 days before the surgery and oral laxatives 24 h before the surgery [5]. Cleansing enema may be needed at the morning of surgery. If it is estimated that intestinal surgery could not be avoided, preoperative fasting could be as early as 2 days before the surgery with parenteral nutrition.

The estimation of amount of bleeding for the surgery of sacral tumors depended on the location of the tumors, histological types, and the experience of the surgeons [1]. Generally speaking these operations last for a long time and need massive transfusion. Some benign tumor could use autologous stored blood transfusion [31], while majority of these cases require abundant blood storage in case of massive hemorrhage intra- or postoperation. If it is estimated that the blood loss would reach 2000 ml, fresh frozen plasma should be prepared for fear of coagulation disorders (the estimation of amount of blood loss can be seen in Table 24.1 [1]). However it is only preoperative estimation which should be adjusted according to the actual situation during surgery.

Table 24.1 The estimation of the amount of hemorrhage for the surgery of sacral and pelvic tumors [1, 31]

Operation region	Blood loss during surgery (ml)	Blood loss after surgery (ml)
Traditional hemi-pelvic amputation	800–3500	300–600
Hemipelvectomy	1500–5000	300–800
The resection of RegionI[a]	400–2000	200–300
The resection of region II[a]		
Endo-prosthesis	1500–3000	400–600
Arthrodesis	2000–4000	300–600
The resection of region III[a]	400–2000	100–300
Partial sacrectomy	1000–3000	400–800
Total sacrectomy	3000–6000	400–1000

[a]According to [32]

24.3 The Techniques to Reduce Massive Hemorrhage During Surgery

Massive bleeding during sacral surgery can rapidly destabilize a patient. Thus, effective hemostasis is critical due to the fatal course of this complication. Reported methods included packing, thumbtacks, inflatable devices, muscle tamponade, muscle fragment welding, and so on [33]. Local hemostatic agents in conjunction with other methods such as diathermy, cyanoacrylate tissue adhesives, and application of bone wax are also available alternatives. However, we don't think all those mentioned above could effectively control massive bleeding during sacral surgery. Whereas complications related to massive blood infusion, such as coagulopathy, hypothermia, and hypocalcemia might cause morbidity, although rare, but possible [1]. Bleeding can be so extensive at times that complete tumor resection becomes impossible. Blood cell salvage has been used to diminish the requirement for autologous blood transfusion but is not recommended in malignant tumor surgery. Thus, the key to the success of these surgeries is to manage intraoperative hemorrhage. We mainly use three methods to control intraoperative hemorrhage, including preoperative embolization of the tumor-feeding arteries under digital subtraction angiography (DSA), ligation or temporary blocking the affected internal iliac artery and application of a balloon dilation catheter (BDC) [34].

1. Digital subtraction angiography and selective arterial embolism.

   It is highly applicable to do embolization of the tumor-feeding arteries under digital subtraction angiography before surgery, which could not only make the local lesion explicited but also effectively reduce intraoperative hemorrhage [8]. Thus we do DSA and selective arterial embolism routinely for those complicated huge sacral tumors, especially for those hypervascular tumors, such as giant cell tumors (Fig. 24.1), aneurysmal bone cyst, and so on.

   However, so far as we know, the effect of SAE is not satisfactory [35]. Besides, embolization is not always practical in sacral surgery for multiple arteries supply blood to the pelvis and sacrum. Embolizing only the lateral common iliac artery or the internal iliac artery will not completely block the blood supplying the tumor. Besides collateral circulation is usually reestablished within 24 h after embolization [12], tumor resection should be scheduled soon afterward. In addition, some serious complications after embolization have been reported, including neurological deficits and pain. Sometimes the embolization could even lead to wound healing problems. Therefore, we usually choose another

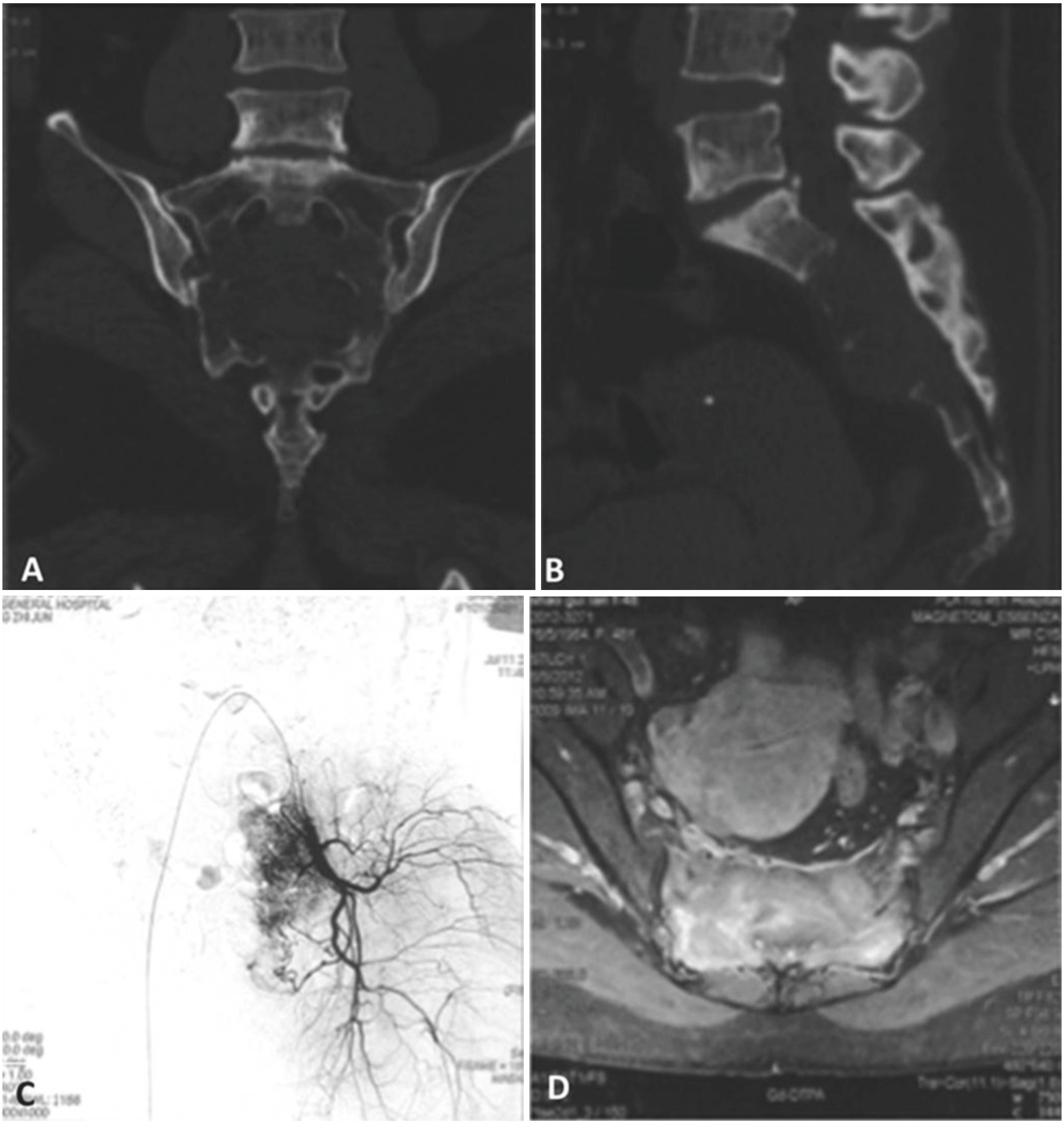

Fig. 24.1 The radiographs of a 48-year-old female patient with sacral giant cell tumor who presented with lumbosacral analgesia for nearly 1 year. (**a**–**c**) showed the CT and MRI manifestation of tumor. (**c**) Digital subtraction angiography showed the hypervascularity of this tumor, and selective arterial embolism was done several times before the surgery. (**d**) The MRI of the local lesion showed the involvement scope of the soft mass

way to control intraoperative hemorrhage such as ligation of affected internal iliac artery and using tape to temporarily block abdominal aorta from anterior approach during surgery.

2. The ligation of affected internal iliac artery and temporary abdominal aorta occlusion (detail seen in Chap. 33).
3. The application of a balloon dilation catheter (BDC) to temporarily block the abdominal aorta (detail seen in Chap. 33).

However there are still some notes special for balloon occlusion. First, to reduce the risk caused by resettlement of the balloon, preoperative ultrasound should be routinely performed to figure out the condition of bilateral femoral artery, external iliac artery, and abdominal aorta. As to elder patients, ultrasound or CTA is especially recommended to exclude vascular disease such as atherosclerotic plaque and vascular malformations, in case of rupture of blood vessels during the placement of the catheter. Second, air in the balloon must be completely evacuated before the catheter is introduced. Liquid was used to inflate the balloon to avoid air embolism in case of balloon rupture during the operation. Third, after the balloon catheter is placed and fixed in position, ultrasonic examination should be available to reconfirm that the balloon is located between the renal arteries and the abdominal bifurcation and that the two renal arteries have not been occluded by the inflating balloon. If the balloon's location is too high, it may cause spinal cord ischemic injury and/or renal ischemia. If urine output is <0.5 ml/kg h, the position of the balloon may be too high and may require an adjustment [36, 37]. The duration of each occlusion should be limited to 1 h to prevent ischemic damage to the spinal cord, pelvic organs, and lower extremities.

Though low-pressure anesthesia has been used for controlling blood loss in resection, it has high demand for intraoperative monitoring and compressive quality of anesthesiologist, because enough blood perfusion for vital organs must be guaranteed throughout the procedure. And it has limited effect and causes high risks [38]. Pre-ligation of internal iliac artery or abdominal artery through abdominal incision is seldom used to control blood loss in clinical, because it causes too much injury and high frequency of postoperative complications [1]. Embolization of bilateral internal iliac arteries and tumor arteries via femoral artery puncture can reduce blood loss greatly and improve surgical safety. However, this method is expensive and time-consuming and may increase the incidence of complications such as lower limb ischemic injury and local ischemic pain. Aortic occlusion with an inflated balloon is deemed to be an effective method for controlling blood loss, which significantly reduces blood loss, eases operation, and offers more time for surgeons to perform. Compared with preoperative iliac arterial or tumor blood supply embolization, balloon occlusion of the abdominal aorta is less complex and safer and is less expensive.

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25 Surgical Approaches to the Sacrum

Taiqiang Yan and Wei Guo

25.1 Posterior Approach

Posterior approach has been performed for two surgical circumstances [1, 2]: One is used in providing exposure for a limited resection of the middle and lower portion of the sacrum (below S2 vertebra) along with the coccyx, such as chordoma involving distal sacrum. The other is in a combined abdominosacral approach for total sacral resection of larger tumor involving S1.

A longitudinal incision (inverted Y type, as shown in Fig. 25.1) is performed through the midline; the two ends curve leftward off the midline. However, in the circumstance of small distal sacral tumor or eccentric tumor, only midline vertical incision or curved longitudinal incision is sufficient to expose the tumor. This incision over the lower sacrum is easy to expose the edge of the sacrum and its bilateral structures, such as the sacrotuberous and sacrospinous ligaments (Fig. 25.2). These ligaments are strong and tense and should be identified and cut off first. The anatomic landmark is the ischiatic notch. It's exposure is to identify sciatic nerve, piriformis muscle, the superior or inferior gluteal artery. The posterior sacral plate is divided at the desired level, and the sacral canal is exposed. Nerve roots can then be ligated and cut off. If possible, unilateral S2 nerve root could be displaced laterally to preserve some sphincter function. The proximal sacral cutting line was defined.

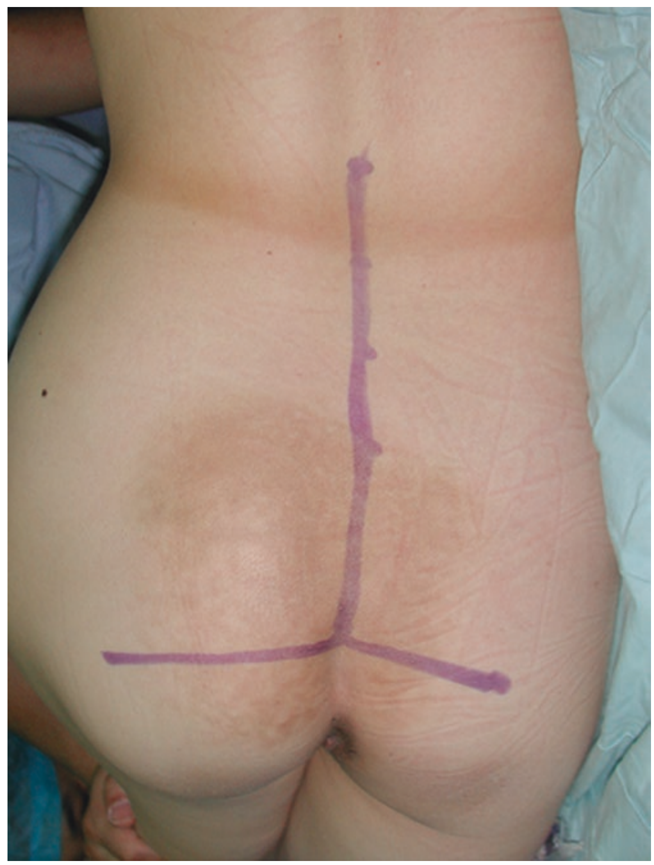

Fig. 25.1 Posterior approach (inverted Y type)

Gaining free access to the ischiorectal fossa is from the edge of the sacrum. Division of the anococcygeal raphe allows one to enter the presacral space. On the anterior surface of the sacrum, blunt finger dissection is used to separate sacrum from the posterior rectum. We packed wet gauze to protect the rectum. What remains is the dissection through the proximal portion of the bone to complete the resection. The transecting sacrum level should be determined on the basis of the preoperative radiologic studies. Because the anterior wall of proximal part of sacrum is thick and hard, we can use rongeur forceps to remove it piece by piece. We should keep in mind that division of this sacrum just below the lower border of S3 vertebra is safe in preserving sphincteric function. Bilateral sacrifice of S2–S4 nerve roots leads to urinary and fecal incontinence and impotence for males. Unilateral preservation of S2 root maintains perfect anorectal continence, and sphincter problems are mild and reversible. Bilateral sacrifice of S1 roots is not possible to restore

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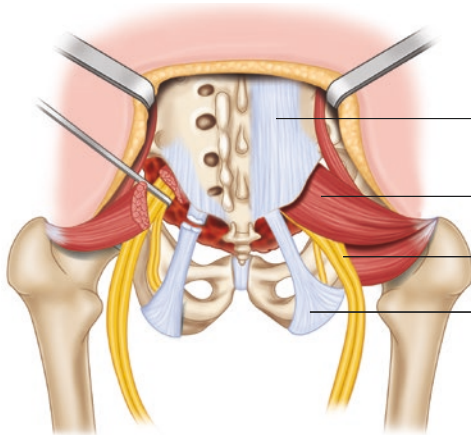

Fig. 25.2 Posterior sacral structure exposure

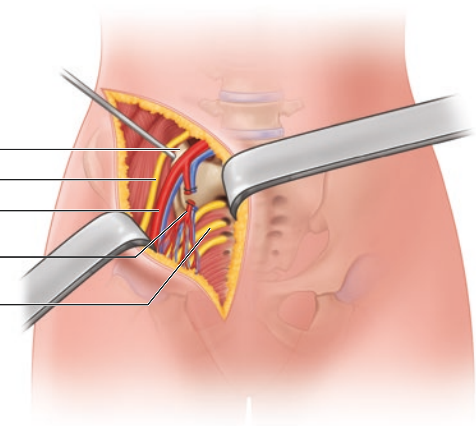

Fig. 25.3 Retroperitoneal space exposure

normal bladder or rectum function even in early rehabilitative treatment after surgery [3, 4, 5].

Following the specimen removal, a large presacral "dead" space may left, and part of the space may be occupied by the rectum. In this situation, the posterior incision would be closed easily. These wounds have a high rate of wound infection, apparently due to the proximity to the anus. On the basis of the preoperative films, it is advisable to perform standard anus finger examination before operation to check whether abnormal adhesion exists between the tumor and the rectum. If so, it is necessary to conduct consultation with the general surgeon. If the rectum rupture was encountered during operation, the general surgeon would repair it; however, in most cases, colostomy may have to be done after the sacral tumor resection [6, 7].

25.2 Retroperitoneal Approach

This approach is able to provide an excellent exposure of the retroperitoneal space (Fig. 25.3). On the one hand, the space from the sacroiliac joint to the greater sciatic notch could be clearly seen. On the other hand, the level of vertebral osteotomy could be identified, including L5/S1 intervertebral disc or lumber vertebral body. The purpose of using retroperitoneal approach is to identity and protect important structures; however, sacrectomy has to be done posteriorly [8].

Angiography should be done preoperatively to learn three important issues: (1) Is the external iliac artery intact? If not, artificial blood vessel transplantation should be performed to restore the limb blood supply during the operation. (2) Is the internal iliac artery involved by the tumor? If yes, the unilateral artery can be ligated safely; if the bilateral internal iliac arteries have to be sacrificed, this is contraindication of operation, because of the risk of bladder, rectum, and uterus necrosis. (3) Abnormal tumor supply should be embolized to reduce blood loss during operation.

25.3 Operation Procedure

Meticulous bowel preparation is advisable preoperatively. A rectal tube is inserted preoperatively with the aim of facilitating to identify the rectum during dissection. In most of these cases, the patient is placed in a supine position or lithotomy position. The incision is performed through the midline, to dissect the three layers of abdominal wall muscle and expose the retroperitoneal space. Large myocutaneous flaps are raised, the spermatic cord is reflected laterally, and incision of inguinal canal is unnecessary. The ureter is identified and traced down to its insertion to the bladder. The neurovascular bundle (femoral artery, vein, and nerve) is reflected laterally. Inferior epigastric artery and veins are ligated, dissociating the external iliac vessels and ureters. The common iliac artery bifurcates along the sacral ala, and the ureter crosses the bifurcation in each side. Careful blunt and sharp dissection is now carried out in the presacral space. Usually, the ureter is easy to recognize; however, it is tough to mobilize when previous surgery in this area is expected to have caused dense adhesions. Preoperative catheterization of the ureter can facilitate to identify the ureter during procedure [1, 2].

The following surgical exposure is to dissect and protect the external iliac vessel, the internal iliac vessel, the common iliac vessel, and the abdominal aorta. The abdominal aorta can be temporarily blocked by the "lace" or Fogarty vascular clamp or aorta balloon to reduce blood loss during operation. Not more than 90 min for each blocking should be kept in mind. The common iliac artery also can be temporarily blocked by the "lace"; the middle sacral artery should be ligated.

The internal iliac vessels usually run under external iliac blood vessels, and the vein is located deeper than artery. Internal iliac vessels can be dissociated if they are on the tumor surface; if not, the artery will be sacrificed. Ligation of unilateral iliac artery does not cause pelvic viscera ischemia.

All vascular branches which supply blood to the tumor should be ligated, including iliolumbar artery, superior or inferior gluteal artery, and obturator artery. The superior gluteal artery should be preserved if it can be dissociated, though the artery is vulnerable to damage and hard to handle. All vessels entering the tumor should be ligated. Usually, it is difficult to dissociate the deep branches of internal iliac vessels, especially when the tumor has a large volume. This can be resolved by posterior approach after sacrum resection.

A lot of reticular veins around the internal iliac vein tend to get damaged. If the hemorrhage is under control, the procedure may have to stop and fill the wound with styptic sponge. In order to avoid this complication, wide exposure may reduce this risk. Vascular tear can be repaired under the condition that the hemostasis by compression is effective for the bleeding vein. Large vascular tear can be repaired.

Upon exposure of the internal iliac artery to the greater sciatic notch, the anterior surface of sacroiliac joints can be identified. Deep vessels can be managed through posterior approach. Cut off part of the iliopsoas muscle if necessary. If there is nerve tumor involvement, it can be sacrificed. Usually L5 nerve roots pass through sacroiliac joints. It is easier to identify the level of division of sacroiliac joints by using Kirschner strings. After touching the anterior surface of the sacrum, blunt finger dissection is used to make sure the transection level and put Mickliz sponge or siliceous membrane on the surface to shield vessels.

Then, the osteotomy line at L5/S1 disc or above will be identified. Vulnerable structures such as major vessels, nerves, or viscus must be identified and mobilized prior to safe resection. Vertebral segment vessels have to be ligated, and some lumbar nerve roots may have to be sacrificed. The disc will be removed as much as possible in this approach, and also the Mickliz sponge or silicon membrane has to shield the vessels.

The rest of the procedure for sacral resection will be done in posterior approach.

25.4 Total Sacrectomy

The combined anterior and posterior approaches are essential for total sacrectomy. This requires repositioning of the patient during operation. A supine position has been advocated for an anterior transabdominal dissection initially, followed by placement of the patient in a prone position.

Anterior approach: A longitudinal incision is performed through the midline, from below the umbilicus 2–3 cm to the pubic symphysis, sparing the rectus abdominis muscle. After opening the posterior peritoneum, careful blunt and sharp dissection is now carried out in the presacral space, separating the bilateral iliac vessels, ligating the bilateral internal iliac artery, and suturing the median sacral artery (Fig. 25.4). Obviously, one should be careful to avoid entering into the tumor as pelvic structures are separated off the anterior sacral surface, and the side peritoneum is incised if necessary. The ureter and inferior mesenteric artery are identified and protected. The "white" lines are then incised along the descending and sigmoid colon in order to allow their mobilization. Free sigmoid colon to the level of anal canal and protect tumor pseudo membrane. After dissecting the rectum, cut off all the nerve roots passing through the tumor, and resect the L5 and S1 intervertebral disc through anterior approach. Sterile gauze is packed on the back of the rectum to fend off

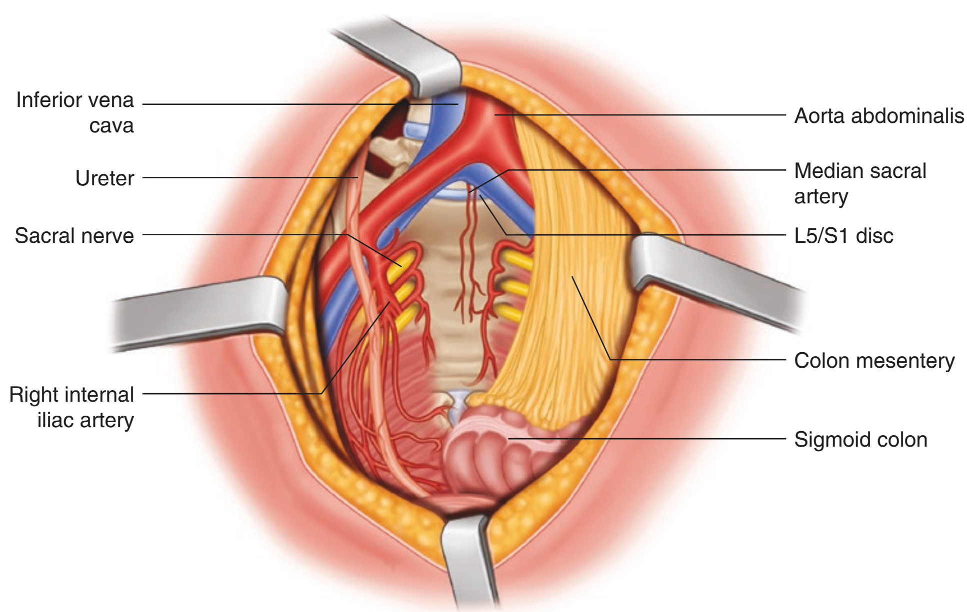

Fig. 25.4 Anterior approach to open the posterior peritoneum and to expose presacral space

the sacrum. Then close the abdominal incision, and start the posterior approach [6, 9].

Wide exposure of the edge of the sacrum and the greater sciatic notch is performed, and the piriformis is cut off. Be sure to fend nerves and vessels off the sacroiliac joint by gauze, and then the osteotomy will be safely done later. The parasacral muscles and gluteus maximus muscles were dissected away, and the L4–L5 spinal process and lamina, posterior cortex of the sacrum, and back aspect of the bilateral sacroiliac joints were exposed. The dura and both sides of the L5 nerve roots were identified and preserved through a laminectomy at the level above the tumor. Then the dura was ligated and cut off at the pre-planned level. Both ends of a plastic tube were inserted from front to back using a thick needle, so that a wire saw could be passed through to cut both sides of the iliac bone during the posterior aspect of the surgery.

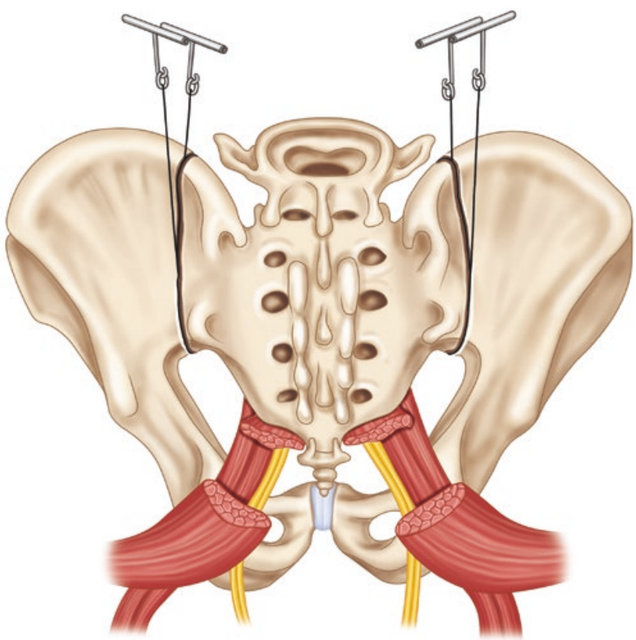

Fig. 25.5 Cutting along the outer edge of the sacroiliac joints via both sides of the iliac bone

To avoid the separation of the spine and pelvis, the temporary internal fixation between the ilium and spine is needed prior to the complete transection of the sacrum.

Pull the sacrum proximally to other side after the dissection on the lateral surface of the sacrum. Careful blunt and sharp dissection is now used to separate the sacrum from the posterior rectum. Both sides of the iliac bone were cut along with the outer edge of the sacroiliac joints using wire saws that were inserted into the silicone tubes placed during the anterior approach (Fig. 25.5). The sacrum was osteotomized at the level of the L5–S1 intervertebral disc, and then the whole sacrum was removed. After all the deep branches of the internal iliac vessels have been ligated and cut off, total sacrectomy resection is completed [6, 9].

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Part VI

Sacral Tumors: Procedures for Sacral Tumor

Two-Stage Total Sacrectomy

26

Francis J. Hornicek

26.1 Introduction

Latin for "sacred bone" and so named for its recurrent role in ancient Greek and Egyptian mythology [1], the *os sacrum* remains the subject of much discourse and scholarship in modern-day musculoskeletal oncology. The treatment of sacral tumors demands attention to the complex interplay of anatomic, biomechanical, and oncologic considerations. However, with meticulous preoperative planning and input from a specialized multidisciplinary team, good functional and oncologic results can be obtained in the surgical management of these tumors.

26.2 Anatomy

The surgical management of sacral tumors demands a detailed understanding of the bony, ligamentous, vascular, and nervous anatomy of the pelvis [2].

Bony: A single bone formed by the fusion of five vertebrae, the sacrum articulates laterally with the ileum via paired L-shaped facets. Inferior articulation (or fusion) with the coccyx involves the two horn-like coccygeal *cornua* and their sacral counterparts. Four pairs of anterior and four pairs of posterior foramina carry the anterior and posterior rami, respectively, of the S1-S4 nerve roots, as they emerge from the sacral canal. The termination of the sacral canal, which itself is the caudal continuation of the vertebral canal, is the sacral hiatus.

Joints and Ligamentous: The articulation between L5 and S1 involves the two zygapophyseal joints and the intervertebral disc. As a result of lordosis at this junction, the L5/S1 disc is wedge-shaped, thicker anteriorly than posteriorly. Stout iliolumbar and lumbosacral ligaments, extending from the transverse processes of L5, reinforce the lumbosacral junction. The synovial sacroiliac joints, prone to fibrosis and fusion with aging, are likewise stabilized by thick anterior and posterior ligaments. Finally, the sacrospinous and sacrotuberous ligaments serve to stabilize the bony pelvis, reinforce the lateral pelvic walls, and define the greater and lesser sciatic foramina.

Muscular: Relevant muscular anatomy about the sacrum includes the paired piriformis and coccygeus muscles, as well as the anococcygeal ligament. The piriformis originates on the anterior surface of the sacrum and exits the pelvis through the greater sciatic foramen, en route to its tendinous insertion on the greater trochanter. The piriformis serves as an important landmark within the greater sciatic foramen; contents of the suprapiriform foramen include the superior gluteal nerve and vessels, while structures exiting inferiorly include the inferior gluteal vessels, sciatic nerve, pudendal nerve, internal pudendal vessels, posterior femoral cutaneous nerve, and nerves to the obturator internus and quadratus femoris.

Contents of the lesser sciatic foramen, separated from its superior counterpart by the sacrospinous ligament, include the tendon of the obturator internus as it exits the pelvis and the pudendal nerve and internal pudendal vessels as they re-enter the pelvis. The coccygeus, which originates on the inner surface of the sacrospinous ligament, inserts on the lateral borders of the sacrum and coccyx. The anococcygeal ligament is at the midline raphe of the left and right levator ani musculature, which form the pelvic floor and help to maintain anal and vaginal closure; the anococcygeal ligament inserts posteriorly on the coccyx.

Peritoneum and Viscera: The sacrum is a retroperitoneal structure. It should be noted that the rectum is retroperitoneal as well; the upper two-thirds of this structure is draped anteriorly by peritoneum, while the lower one-third is completely uncovered by peritoneum.

Vascular: The internal iliac vascular system is relevant to surgery of the sacrum and pelvis. The paired internal iliac arteries typically branch from the common iliac arteries at

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the level of L5/S1, anteromedial to the SI joint. At the superior border of the greater sciatic foramen, the internal iliac artery divides into anterior and posterior trunks, which each subsequently gives rise to multiple named vessels. The posterior trunk of the internal iliac supplies the posterior pelvic wall and gluteal region; branches include the iliolumbar artery, which ascends superiorly out of the posterior pelvis and gives off a spinal branch that passes through the L5/S1 intervertebral foramen; the lateral sacral artery, which gives multiple branches that pass into each anterior sacral foramina; and the superior gluteal artery, which exits the pelvis through the suprapiriform greater sciatic foramen and supplies the abductor musculature.

Relevant branches of the anterior trunk of the internal iliac include the internal pudendal artery, which runs through Alcock's canal with the pudendal nerve and supplies the external genitalia; the obturator artery, which exits the pelvis through the obturator canal into the adductor compartment of the thigh; the inferior gluteal artery, which exits the pelvis through the infrapiriform greater sciatic foramen and supplies the gluteus maximus and piriformis; and multiple branches to the pelvic viscera. The median sacral artery, an unpaired, midline vessel, branches off the abdominal aorta just above its bifurcation and travels down the anterior surface of the sacrum and coccyx.

Nervous: The sacral and coccygeal plexuses, formed by the anterior rami of S1-Co with contributions from L4-L5, carry primarily somatosensory fibers, with sympathetic and parasympathetic components as well. As noted above, four paired anterior and four paired posterior foramina transmit the anterior and posterior rami of the S1-S4 nerve roots. Each anterior ramus, except at the S4 level, in turn divides into ventral and dorsal divisions.

The sacral plexus gives rise to multiple somatic nerves, including the sciatic (L4-S2), superior and inferior gluteal, and pudendal nerves, as well as smaller motor branches to the quadratus femoris, gemelli, obturator internus, levator ani, and coccygeus muscles, and two sensory nerves (the posterior femoral cutaneous and perforating cutaneous). The pudendal nerve is of particular importance to surgery of the sacrum and pelvis. Arising from the ventral divisions of the anterior rami of S2-S4, the pudendal nerve exits the pelvis through the infrapiriform greater sciatic foramen, passes dorsal to the sacrospinous ligament, and re-enters the pelvis through the lesser sciatic foramen. As it does so, it courses along the lateral wall of the ischioanal fossa within Alcock's canal (pudendal canal), inferior to the pelvic floor and accompanied by the internal pudendal vessels. The pudendal nerve innervates the skeletal muscle of the external anal and urethral sphincters as well as the levator ani and provides sensory innervation to much of the perineum and penis or clitoris. Compression or injury to the pudendal nerve or its sacral nerve roots can result in bowel, bladder, and sexual dysfunction, which is of great concern in the management of sacral tumors as discussed further below.

In addition to its somatic nervous functions, the sacral plexus provides parasympathetic visceral innervation through its contributions to the inferior hypogastric plexus of the pelvis. The S2-S4 nerve roots contribute to parasympathetic pelvic innervation, including vasodilation of the erectile tissue of the penis or clitoris, stimulation of bladder contraction during micturition, and modulation of activity of the descending colon and rectum. This parasympathetic outflow is carried by the pelvic splanchnic nerves, which originate from the S2-S4 roots. The pelvic splanchnic nerves join fibers from the superior hypogastric plexus, which descends along the posterior abdominal wall and carries both sympathetic and parasympathetic fibers, to form the inferior hypogastric plexus.

Sympathetic functions of the inferior hypogastric plexus include innervation of smooth muscle within the pelvic vasculature, internal anal and urethral sphincters, and reproductive tract (i.e., critical for the processes of ejaculation). It should be noted that sympathetic fibers within the inferior hypogastric plexus are supplied by the roots of T10-L2, but *not* by sacral nerve roots.

Finally, the somatic coccygeal plexus, with contributions from S4, S5, and Co, gives rise to the anococcygeal nerve, which contributes to motor and sensory innervation of the perineum.

26.3 Surgical Anatomy

The bony, ligamentous, and nervous anatomy of the pelvis, as related to pelvic biomechanics and function, must be considered in the course of preoperative planning. Partial transverse sacrectomies have been classified with respect to nerve root anatomy, wherein preservation of S3, S2, and S1 correspond to low, middle, and high sacrectomies, respectively [3]. The level of resection is of critical importance not only with respect to margin status but also with respect to preservation of mechanical stability and bowel, bladder, and sexual function. It should also be noted, however, that the level of bony resection does not necessarily correlate with extent of sacral root sacrifice: intraoperative oncologic considerations might require sacrifice of more cephalad nerve roots in order to achieve negative margins, or conversely, tumor location might allow for the sparing of nerve roots contralateral or caudal to the bone cut [4].

26.3.1 Biomechanical Considerations

Spinopelvic fixation is typically performed after total sacrectomy; in the absence of reconstruction, as elaborated by one author, the resultant "flail axial skeleton precludes the ability to ambulate" [5]. On the other hand, low partial sacrectomy does not warrant reconstruction. However, the indications for reconstruction after high or middle partial sacrectomy are less clear. Early biomechanical work performed by Gutenberg [6] found that resection through (or just cephalad to) the S1 foramina weakened the pelvic ring by 50%, while resection between the S1 and S2 foramina resulted in only 30% weakening. Regardless, the former was thought to preserve sufficient bony stability to allow for standing, and therefore it was concluded that reconstruction is not required for resections that preserve any amount of S1 body.

In contrast, a more recent cadaveric study, which purported to model physiologic loading more accurately, found that pelvises with sacral resections just caudal to the S1 foramina could withstand forces associated with postoperative mobilization, while those with resections just cephalad to the S1 foramina could not [7]. These authors highlighted the importance of at least partial preservation of the sacroiliac joint to pelvic stability and noted that bone cuts just cephalad vs. just caudal to the S1 foramina are associated with preservation of 75% vs. 84%, respectively, of the sacroiliac joint—perhaps signifying a biomechanically significant cutoff point within that range. Clinical outcomes data have largely confirmed these findings. One review found that three of nine sacrectomies involving the S1 body failed via fracture, ultimately requiring reconstruction [8]; another series reported a 76% rate of postoperative sacral insufficiency following high sacrectomy (and adjuvant radiation), as compared with 0% after low sacrectomy [9].

It should be noted, however, that not all authors advocate for reconstruction after total or high sacrectomy. Ruggieri et al. [10] have suggested that muscle and scar tissue may form a “biologic sling” between the unreconstructed pelvis and lower lumbar spine, which may migrate inferiorly toward the ilia; the use of a lumbosacral corset may increase stability and ambulatory ability in these cases [11].

26.3.2 Neurologic Considerations

The S3 nerve roots are thought to play a critical role in supplying normal bowel, bladder, and, to a lesser extent, sexual function. Sacrectomy with nerve root sacrifice cephalad to S3 may result in loss of normal bowel and bladder function in many [3, 8, 12] or all [6, 13] patients. Specifically, S2 may allow for weak internal and external anal sphincter activity but neither for discrimination between different rectal contents or sensation of rectal distention nor for maintenance of the micturition reflex. A review of sacrectomies performed at the Mayo Clinic found that preservation of bilateral S3 nerve roots ensured maintenance of normal bowel and bladder function in 100% and 69%, respectively, of patients; unilateral S3 preservation ensured maintenance of function in approximately two-thirds of patients. All patients with sacrifice of bilateral S2 roots had abnormal bowel and bladder function, and a minority of patients enjoyed normal bowel and bladder function with bilateral S3 sacrifice [8]. However, a more recent case series [13] found slightly improved outcomes with S3-sacrificing sacrectomies: normal bowel and bladder function in 63% and 71%, respectively.

Preservation of the S2 nerve roots—and perhaps even S1 nerve roots alone—might be sufficient for maintenance of sexual function [6, 14, 15]. Unilateral S1-S5 resection, with preservation of contralateral nerve roots, has minimal impact on bowel, bladder, or sexual function [3, 6, 8].

Investigations of patient-reported outcomes at our center, utilizing PROMIS questionnaires, have confirmed the negative impact on pain and quality of life resulting from more proximal resections. A study of 74 patients undergoing total or partial transverse sacrectomy identified significantly lower physical and mental health scores in patients for whom S3 was the most cephalad nerve root spared, as compared with those in whom S3 was preserved. Additionally, sparing of S2 resulted in higher orgasm scores. Interestingly, no difference in quality of life metrics was observed in patients with and without colostomy creation [4].

Evaluation of PROMIS questionnaire data for patients with low sacral resections (bony resection through S4 or S5) demonstrated particularly good functional outcomes. These patients, unsurprisingly, reported better physical health, mobility, and ability than patients undergoing total sacrectomies. However, quality of life and functional scores following low sacrectomy were in fact equivalent or superior to *normative*, general population PROMIS data [16]. High partial and total sacrectomies were also consistently associated in chronic pain in the reviews of PROMIS data [4, 16]; this finding was echoed in another review of 21 patients treated with sacrectomy for chordoma, which found that neuropathic pain persisted at final follow-up in 8 of 11 patients who presented with this complaint preoperatively [17]. As with bowel, bladder, and sexual function, therefore, chronic pain must be addressed as a known risk of high sacrectomy in preoperative patient discussions.

Of course, sacral nerve root status is only one of a number of factors that may contribute to post-sacrectomy function. High partial sacrectomies may necessitate lumbo-pelvic fixation, as noted above, which can be associated with sciatic nerve dysfunction or pain-generating hardware failure. Patients undergoing more proximal resections are also typically those with larger tumors or more significant preoperative sacral root involvement. Indeed, it has been demonstrated that the greatest predictor of postoperative bowel and bladder outcome is preoperative function [13]. Disruption of the hypogastric plexus, as may occur during a direct approach to the spine or direct injury to the pudendal nerves, can also result in bowel, bladder, or sexual dysfunction, even if the sacral nerve roots proper are preserved.

26.4 Clinical Management of Sacral Tumors

Sacral tumors may present with pain, perineal sensory changes, and sexual, bowel, and bladder dysfunction, the latter of which may be the result of either nervous or direct visceral compression. Additionally, given their location, sacral tumors may be quite large before symptoms arise. As with evaluation of any bone tumor, initial work-up should consist of history, physical examination, and imaging studies, including plain radiographs, CT and MRI of the sacrum and pelvis, and bone scan and CT of the chest for staging. Complete imaging of the mobile spine should be performed as well; additional lesions may be present in 17% of patients with sacral chordomas [18].

Tissue is necessary for histologic analysis and should preferably be obtained through image-guided needle (aspirate or core) biopsy. Open incisional biopsy of chordomas, especially when performed outside of the ultimate treating center, is associated with a higher risk of local recurrence, metastasis, and tumor-related death [3, 19, 20]. Analysis of sacral mass biopsy tissue should include immunohistochemical staining for cytokeratins, EMA, and vimentin, which stain strongly in chordomas, as well as for Ki-67, which is associated with a poor prognosis when present with a high degree of proliferative activity [19].

Tumor characteristics dictate surgical management, including extent of resection and use of adjuvants. Chordomas are the most common primary sacral malignancy; sacral chondrosarcomas, osteosarcomas, and Ewing's sarcomas are seen as well. Benign primary tumors include giant cells tumors, aneurysmal bone cysts, and osteoid osteomas/osteoblastomas. Metastatic disease, multiple myeloma, and lymphomas are commonly encountered. Teratomas are the most common sacral tumors in children [21].

26.4.1 Management of Chordomas

Deriving from notochordal tissue, chordomas represent 1–4% of all primary malignant bone tumors and occur more commonly in the sacrum (50%) than in the skull base (35%) or the mobile spine (15%) [22]. Intralesional resection (or resection with inadequate margins) is associated with a higher rate of local recurrence (up to 83%) and, in turn, a lower survival [10, 12, 19, 22–24]. In one series, local recurrence was associated with a 23-fold increased risk for metastases and a 21-fold increased risk for tumor-related death [19]. Another large series found a more modest 2.5-fold increase in metastatic risk among patients with local recurrence, and survival among those with metastatic chordoma ranges from 20 months for cervical disease to 130 months for sacral disease [25]. Wide, en bloc resection is therefore recommended in the management of chordomas.

It has been suggested that tumor invasion into the piriformis or gluteus maximus muscles, or involvement of the sacroiliac joints, is an independent predictor of local recurrence, regardless of margin status at the time of resection [23]. Indeed, local recurrences may tend to occur in the posterior musculature, and wider margins may be required posteriorly as compared with anteriorly, where the presacral fascia may pose a barrier to tumor spread [23, 26].

High-dose (~70 Gy) proton/photon beam radiation therapy is now standard in the management of chordomas and other spinal malignancies at our institution. For sacral tumors, preoperative radiation of ~20 Gy is administered, with the remainder administered as a postoperative boost. A phase II clinical trial performed at the Massachusetts General Hospital, evaluating patients undergoing surgical resection of spinal chordomas and sarcomas (predominantly sacral), demonstrated that high-dose radiotherapy in addition to surgical resection was associated with a 74% rate of local control at 8-year follow-up [27, 28]. Notably, local control for primary tumors (94% at 5 years and 85% at 8 years) was far superior to that for locally recurrent tumors (~50% re-recurrence rate).

Chordomas, in particular, appear to benefit from high-dose radiation. A retrospective review of spinal chordomas treated at our institution demonstrated 5-year overall survival and local control of 81% and 62%, respectively [24]. This study found further improvement in local control when surgical resection of primary chordoma was accompanied by neoadjuvant and adjuvant radiation, as opposed to adjuvant radiation alone: 85% vs. 56% at 5 years. Most strikingly, among the 28 patients in this series who underwent en bloc resection and received both neoadjuvant and adjuvant radiation, *no* cases of recurrence were observed. Even in cases of margin-positive primary resection, good results may be salvaged: the use of adjuvant high-dose radiation achieved local control at 8.8 years in 10 of 11 patients with primary sacral chordomas (but in 0 of 5 patients with recurrent disease) treated at out institution [29]. Taken together, these results high lighted not only the excellent results achieved with neoadjuvant and adjuvant radiation and wide resection, but also the critical importance of initiation of aggressive treatment at first presentation.

Additionally, in patients for whom surgical resection is not feasible, due to risk of intraoperative neural injury (i.e., high sacral tumors) or medical comorbidities, definitive management with high-dose radiation is reasonable. In a review of 24 spinal chordomas, of which 19 were located in the sacrum, treated at our institution with proton or photon radiotherapy alone, local control rates were 90.4% and 79.8% at 3 and 5 years, respectively. All surviving patients maintained ambulatory status [30]. A follow-up study noted an ongoing tumor volumetric reduction up to 5 years after definitive radiation treatment [31]. However, high-dose radiation therapy to the sacrum can be associated with significant adverse effects. Specifically, delivery of greater than 77 Gy, which is required in cases of definitive treatment with radiation alone, has been associated with higher rates of neuropathy and erectile dysfunction, with lower rates of neural injury in patients receiving ~70 Gy or less [24, 27, 28, 30]. Sacral insufficiency fractures have been noted to occur in roughly half of all patients with sacral chordomas treated with definitive high-dose radiation [30, 31] and in over three-quarters of patients undergoing high sacrectomy [9]. For this reason, we avoid radiation doses greater than 70 Gy for patients undergoing surgical resection.

26.4.2 Management of Other Sacral Malignancies

Chondrosarcoma of the sacrum represents approximately 20% of spinal chondrosarcomas and 5% of all chondrosarcomas [32] and is the second most commonly resected primary sacral tumor after chordoma [3, 28, 33]. As is the case with other sacral malignancies, en bloc resection with negative margins is likely associated with decreased rates of local recurrence and improved disease-free survival [32, 34, 35]. Chondrosarcomas, like chordomas, are treated with high-dose neoadjuvant and adjuvant radiation, in addition to wide surgical resection, at our institution [28]. Spinal osteosarcoma is rare, but sacral involvement has been reported in 31–68% of cases [36, 37]. At our institution, treatment includes en bloc resection, when feasible, as well as high-dose radiation therapy and neoadjuvant and adjuvant chemotherapy [37], though outcome is poor and prognosis is worse for osteosarcoma of the sacrum as compared with that of the mobile spine [36].

Locally recurrent rectal cancer may be treated with aggressive re-resection, to include partial sacrectomy in cases of cortical invasion or tumor adherence to bone. Overall mean survival of 22–40 months has been reported following re-resection with sacrectomy, with improved outcomes in cases of negative margins [38–40]. The use of pedicled omental or rectus abdominis flaps has been described as a means of decreasing wound complications [41].

26.4.3 Management of Giant Cell Tumors

Traditionally, giant cell tumors (GCTs) of the sacrum have been treated with intralesional curettage, but high rates of recurrence—up to one-third to one-half of patients—have been reported [42, 43]. The authors of a review of a pooled cohort of 166 patients with sacral GCTs, therefore, recommended wide surgical resection for lower sacral lesions and for recurrent proximal sacral lesions. Notably, this study reported a 23% disease-related mortality, of which approximately one-third was related to treatment complications, at 8-year follow-up. Radiation may be utilized in cases of large or challenging sacral GCTs but is associated with high recurrence rates when used alone or as an adjuvant following curettage [42]. Radiation-related malignant transformation is also a concern [42–44]. Arterial embolization may represent a more successful nonoperative modality, and good results with respect to symptomatic improvement and low recurrence have been reported with the use of serial arterial embolizations (typically every 4–6 weeks) as monotherapy for sacral GCTs [45–47]. Additionally, embolization may be performed concurrently with local intra-arterial injection of cisplatin [47] or may be employed as a preoperative adjunct to limit surgical bleeding [48].

26.5 Total and Partial Sacrectomy: Surgical Techniques

Meticulous preoperative planning is a necessity for successful surgical management of sacral tumors. Preoperative considerations include options for preservation of fertility, plastic surgery consultation, and vascular embolization, while intraoperative considerations include choice of approach (combined anterior-posterior vs. posterior-only), use of computer-assisted navigation, and method of spinopelvic fixation, when needed.

26.5.1 Preoperative Preparation

We routinely refer reproductive-aged female patients for gynecologic evaluation prior to initiation of oncologic treatment. If desired, laparoscopic oophoropexy may be performed prior to initiation of radiation therapy, relocating the ovaries within the abdominal cavity such that they are outside of the planned radiation field, thereby preserving future fertility options.

Preoperative consultation with a plastic surgeon should also be pursued prior to sacral resection; wound closure often proves difficult, given location, extent of surgery, prior radiation, and poor patient nutrition. Options for soft tissue wound closure include mobilization of pedicled omental flap; transpelvic vertical rectus abdominis myocutaneous (VRAM) flap, fed by the inferior epigastric vessels; and local tissue advancement techniques. A reconstructive algorithm put forth by Miles et al. [49] recommends that bilateral gluteus advancement flaps are associated with the lowest complication rate among reconstructive options and therefore should be the first choice for post-sacrectomy closure if the gluteal vessels are intact; transpelvic VRAM reconstruction should be performed in the setting of preoperative radiation, though is contraindicated for patients with prior laparotomy; and free flap coverage may be performed if other options are unavailable. In their review of outcomes following surgical management of sacral chordomas, Schwab et al. [12] found a significant decrease in wound complications in patients in whom rectus flap coverage was performed, as compared with patients for whom no flap coverage was performed. Reconstruction of the pelvic floor can also prove problematic; sacroperineal hernia is a rare complication but a risk in cases of large pelvic floor defects. Good results have been reported with reconstruction utilizing acellular dermal collage grafts [50] or mesh [51].

Intraoperative blood loss may be minimized by bilateral internal iliac ligation, which is the standard practice at our institution; however, preoperative embolization and intraoperative aortic balloon pump occlusion are options as well. Embolization of the internal iliac, median sacral, and other tumor-feeding arteries, typically within 24 h of surgery, may be performed with Gelfoam, thereby potentially minimizing longer-term devascularization of healthy tissues [52]. Additionally, the use of occlusive aortic balloon pump has been reported as a technique to reduce intraoperative blood loss in sacral tumor resections [53, 54]. In their review of 215 sacral resections, Tang et al. [53] found that balloon occlusion, utilized in 120 patients, was associated with significantly lower blood loss (2.2 vs. 3.9 L).

26.5.2 Staged Anterior and Posterior Approaches for Total and Partial Sacrectomy

26.5.2.1 Anterior Approach

Following induction of general anesthesia, administration of antibiotics for coverage of both bowel and skin flora, and establishment of central venous access, the patient is positioned supine on a flexible Jackson table, broken in the middle to improve intraoperative sacral exposure. With the assistance of a general surgeon, a low midline incision is made from the pubis to the umbilicus, and the peritoneal cavity is entered. An Omni retractor is utilized. The rectosigmoid colon and left ureter are mobilized away from the tumor, pelvic side wall, and sacrum; this process is repeated on the right side. Great care must be taken not to inadvertently enter the tumor in the course of mobilizing the overlying tissues. For chordomas in particular, violation of the tumor pseudocapsule confers a poor chance for curative resection. Subsequently, following identification and mobilization of the common, internal, and external iliac arteries, the trunks of the left and right internal iliac arteries and veins are ligated just distal to their respective bifurcations; large branches, such as the gluteal and iliolumbar vessels, may be identified and individually ligated as well. Exposure of the anterior sacrum will typically also require ligation of the median sacral artery. The L5 nerve root, which is closely associated with the anterior surface of the sacral ala, is identified and protected. The L5 root may be traced distally to facilitate further exploration of the lumbosacral plexus and identification of the sciatic nerve within the greater sciatic foramen.

En bloc partial sacrectomy: If a partial sacrectomy is to be performed, a high-speed burr is used to create a transverse osteotomy through the anterior sacral cortex, cephalad to the tumor. An intraoperative radiograph is made to assess the level of the osteotomy. The osteotomy is started in the midline, advanced laterally toward the left and right SI joints (or toward the greater sciatic foramina, in cases of more distal resections), and continued posteriorly until the posterior sacral cortex is reached. As the osteotomy is extended laterally from the midline, exiting nerve roots cephalad to the level of the cut are protected and gently retracted laterally; the more proximal L5 nerve root will need to be retracted as well. Additionally, osteotomies at the S1-S2 level may place the superior gluteal vessels at particular risk [55]. A Cobb elevator can be used to deepen the osteotomy and score the posterior cortex. Throughout the creation of the osteotomy, hemostasis is maintained with silk ligatures, monopolar and bipolar electrocautery, Floseal and Surgicel, epinephrine-soaked sponges, and bone wax. Prior to closure, a Gore-Tex patch may be placed ventral to the sacrum but dorsal to the rectum and left in place until the second stage, at which point it will serve as a landmark for the safe anterior extent of the dissection.

En bloc total sacrectomy: Following anterior approach and exposure, a spinal needle is placed into the L5/S1 disc space (for tumors that do not extend cephalad to S1), and correct level is confirmed on a lateral radiograph. As noted above, the L5 nerve roots and internal iliac vessels (in particular, the artery on the right) should be identified and protected in the course of dissection anterior to the L5/S1 junction and sacral ala. Discectomy is performed at this level with curettes, Kerrison rongeurs, and endplate elevators, extending dorsally to the posterior annulus. Using a high-speed burr, longitudinal osteotomies are created through the anterior cortices of the posterior ilia bilaterally. The resultant troughs are packed with bone wax for hemostasis; osteotomies will be completed posteriorly during the second-stage procedure. The location of the osteotomies with respect to the sacroiliac joints is dictated by tumor size and margin considerations; for small, midline sacral tumors, the osteotomies may even be created within the sacral ala, thereby preserving the sacroiliac joints.

Coverage with a pedicled rectus abdominis flap is standard practice at our institution. Following completion of the anterior osteotomy, the VRAM flap is harvested by the plastic surgeon. Typically, the flap, once raised, is dunked into the pelvis and secured with a Prolene suture to assist in subsequent orientation. Blake drains are placed within the anterior pelvis. The patient is taken to the intensive care unit for further resuscitation between stages. Vena cava filter is typically placed on the first postoperative day, to obviate the need for chemical DVT prophylaxis. Additionally, we obtain a pelvic CT scan between stages for assessment of the osteotomy level. The posterior procedure is performed 3–5 days later.

26.5.2.2 Posterior Approach

The patient is placed prone, in a position of neutral lumbar lordosis, and a midline incision is made. For large masses with dorsal extension into the superficial soft tissues, a skin paddle may be removed with the specimen; biopsy tracts should also be ellipsed out with the incision. Dissection proceeds through fascia to the spinous process of L5. Dissection proceeds laterally, elevating the paraspinal musculature proximally, and more distally and laterally, elevating or transecting the gluteus maximus to expose the ilium. Medially, the sacral ala is exposed, as are the adjacent transverse process and lamina of L5. After removal of spinous processes with a Leksell rongeur, laminectomy just cephalad to the level of the planned sacral osteotomy is performed with a diamond-tipped burr and Leksell and Kerrison rongeurs. The dorsal aspect of the dura is dissected free of adhesions to the overlying lamina with a Stevens scissors and Penfield 4. The laminectomy may be widened laterally with a small osteotome. The bilateral nerve roots at L5, S1, and any additional caudal levels to be preserved are exposed laterally and mobilized with a Kerrison rongeur and Stevens tenotomy scissors. The thecal sac is then circumferentially exposed at the level of the planned resection, just caudal to the axilla of the exiting nerve roots, and a right-angled clamp is passed ventral to the dura. The thecal sac is ligated with 2–0 silk suture and clips and then sharply transected. Later in the procedure, after removal of the mass, the transected end of the thecal sac should be covered with dural sealant as well.

Dissection is carried laterally and caudally, elevating the gluteus maximus musculature; a Versa-Trac self-retaining retractor is helpful in maintaining exposure. The extent of lateral exposure will be dictated, in part, by the soft tissue extension of the tumor. Dissection proceeds caudally along the lateral borders of the sacrum, exposing and identifying the piriformis, as well as the sacrotuberous and sacrospinous ligaments. Sizeable traversing vessels are typically encountered about these structures and will need to be ligated; the use of a bipolar sealer (such as the Aquamantys) can be of benefit here. The piriformis and ligaments are subsequently divided, allowing access into the pelvic cavity through the greater sciatic foramen. Now, using blunt dissection, a plane may be developed ventral to the tumor and dorsal to the rectum. The sciatic nerve should be identified and protected throughout. Dissection should be continued caudally until the tip of the coccyx is palpable.

En bloc partial sacrectomy: An intraoperative radiograph is made to assess the correct level for completion of the sacral osteotomy. A high-speed burr and Cobb elevator are used to complete the osteotomy in the midline. As the osteotomy is continued laterally, nerve roots cephalad to the level of resection should be identified, dissected free from overlying bone with a high-speed burr, and protected. The osteotomy is eventually completed laterally into the left and right sciatic foramina. The mass is now lifted caudally, carefully elevating it off of the adjacent rectum; a Gore-Tex patch, if placed ventral to the sacrum during the first stage, will serve to identify the safe plane between the sacrum and rectum. Nerve roots passing through the tumor are sacrificed. Vessels extending into the specimen are clamped and ligated. Remaining soft tissue attachments to the pelvic floor are transected, the specimen is passed off, and hemostasis of the resection bed is obtained.

En bloc total sacrectomy: Posterior exposure is performed as detailed above. The thecal sac is ligated just cephalad to the L5/S1 disc space after laminectomy is performed at this level. The posterior L5/S1 discectomy is completed. The bilateral iliac osteotomies, initiated during the anterior approach, are completed posteriorly using a high-speed burr and Cobb elevator. The sacrum and mass are lifted out of the wound, as described above.

Spinopelvic Reconstruction: As discussed in greater detail above, the extent of sacral resection impacts the degree of biomechanical instability imparted and therefore the need for spinopelvic fixation. Total sacrectomy warrants spinopelvic reconstruction. Multiple techniques for reconstruction of the bony defect after sacrectomy have been reported. Dickey et al. [5] described a triangular construct in which two segments of fibular autograft are oriented in an inverted "V," such that the apex is "docked" into the inferior endplate of L5 and each limb is buttressed against the inner table of the ilium, along the iliopectineal line. The authors note that the oval "receptacles" in ilium need to be fashioned, utilizing a high-speed burr, during the anterior approach. This construct allows for the structural bone graft to be placed along the force transmission lines between the acetabulum and lumbar spine. As discussed in greater detail below, vascularized fibular grafts can be used in a similar fashion.

Instrumentation is then performed: first, a wide osteotome is used to remove the dorsal cortex of the posterior superior iliac spines, which may be morselized for use as bone graft. Two iliac screws are placed on each side; orienting the pedicle probe toward the ipsilateral greater trochanter helps with proper placement. Standard pedicle screws are placed bilaterally at L4 and L5. A total of four lumbo-pelvic rods are used: on each side, rods are fixed to the heads of the L4, L5, and proximal iliac screws. Rod-to-rod connectors are used to incorporate two additional, parallel rods, one on each side. A horizontal transiliac rod is fixed to the two distal iliac screws.

In cadaveric biomechanical testing, this dual-rod, double iliac screw construct has been shown to be stiffer than constructs involving single-rod, single iliac screws, or Galveston rods (in which the distal end of the spinopelvic rod itself is driven between the inner and outer tables of the pelvis) [56]. Cross-link connectors may be used proximally between the parallel lumbo-pelvic rods and distally between the lumbo-pelvic and transiliac rods, to provide additional stability. Burr decortication of the lumbar posterior elements is performed, and morselized autograft and allograft bone is laid down to promote fusion. We routinely make use of calcium phosphate scaffold graft as well.

Additionally, our practice is to perform limited spinopelvic reconstruction for partial sacrectomies cephalad to the S4 foramina, especially in cases of neoadjuvant radiation, which we feel significantly retards the fusion process. In these cases, L5-ilium instrumentation is performed with single rods and double iliac screws bilaterally and a horizontal transiliac rod. Complete exposure and thorough decortication of the remnant sacroiliac joints will increase the probability of fusion.

Vascularized autograft reconstruction: Though not routinely performed at our institution, the use of vascularized fibular autografts in reconstruction after total sacrectomy has been described. Choudry et al. [57] reported on the use of bilateral vascularized fibular grafts, fashioned into an inverted V at the spinopelvic junction as described above; the pedicles are anastomosed with the internal iliac vessels (presumably prohibiting intraoperative iliac ligation). In this study, bony fusion was observed on CT scan at 6 months. Anastomosis of free fibular grafts into the deep inferior epigastric pedicle of the mobilized VRAM flap has also been described [58]. This “vertical rectus abdominis musculocutaneous flow-through flap” technique has the advantage of permitting ligation of the internal iliac vessels during anterior approach. These authors noted arthrodesis on imaging within 3 months of surgery. It should be noted that the added complexity of this procedure, which in some cases required the use of a saphenous vein graft to increase fibular pedicle length, necessitated a three-stage procedure in some cases.

Computer-assisted navigation: Intraoperative CT-guided navigation is now routinely used at our institution. Following anterior sacral exposure during the first-stage procedure, the anterior iliac crest is exposed through a small longitudinal incision. The navigation system tracker is affixed to two Steinman pins driven through the iliac crest, between the inner and outer tables. Intraoperative CT scan is performed. Navigation subsequently aids in identification of the correct osteotomy level and assessment of anterior-posterior depth during creation of the osteotomy. During the posterior procedure, the tracker is affixed to one of the lumbar spinous processes, and CT scan is performed after laminectomy and nerve root exposure. Navigation is then used to confirm that the posterior osteotomy is being performed at the same level as the anterior osteotomy.

26.5.2.3 Rebar Technique for Reconstruction After Curettage of Proximal Sacral Tumors

In select circumstances, benign tumors (i.e., GCTs) within the proximal sacrum, treated with curettage of the S1 body, may be amenable to reconstruction with an anterior “rebar” technique and limited posterior spinopelvic stabilization. We prefer, in these cases, to perform the posterior intralesional resection and spinopelvic instrumentation as a first stage. Laminectomies of the proximal sacrum and L5 are performed, the filum terminale and sacral nerve roots are dissected and protected, and a Gore-Tex patch is placed ventral to the nervous structures; residual, inaccessible anterior tumor is left for completion of resection in the second stage. As compared with that for a total sacrectomy, less extensive instrumentation is required: single, bilateral L5-ileum spinal rods, with a single horizontal trans-iliac rod, are sufficient. The second stage involves a standard transperitoneal approach and resection of the remnant anterior tumor and S1 body. A high-speed burr, curettes, and Kerrison rongeurs are used to excise the tumor in a piecemeal fashion. If the posterior approach was performed previously, the Gore-Tex patch, placed ventral to the sacral nerve roots, will be encountered and will signify the dorsal extent of safe dissection.

Reconstruction commences with the placement of one or two large fragment screws into both the inferior endplate of L5 and exposed superior aspect of S2. The screws may be placed in a retrograde or antegrade fashion to maximize cortical purchase, such that either the heads or the tips are exposed within the defect. Antibiotic-laden bone cement is then introduced into the defect and molded around the exposed screws. A Gore-Tex patch or segment of Esmarch tourniquet may be utilized as a temporary dorsal “backstop,” preventing damage to the underlying nerve roots as the wet cement cures.

26.5.3 Posterior-Only Approach for Total Sacrectomy

While staged anterior-posterior approaches are the preferred option for total sacrectomy at our institution, other authors have advocated for posterior-only approaches. Good oncologic outcomes with low rates of bowel or vascular injury have been reported with posterior-only total sacrectomy [17, 20]. The use of radiopaque gauze, packed anteriorly to the sacrum, from above the iliac crests and through the greater sciatic foramina, has been described as a method of tamponading anterior bleeding and displacing the iliac vasculature and pelvic viscera away from the osteotomy sites [17].

Disadvantages of a posterior-only approach include blind dissection of the rectum off of the anterior surface of the sacrum and inability to ligate the internal iliac vasculature as a method of decreasing blood loss. Additionally, it is impossible to raise a rectus abdominis flap from the posterior-only approach, though proponents have argued that preservation of the gluteal vessels (and internal iliac system as a whole) decreases soft tissue ischemia and allows for successful closure with gluteus maximus myocutaneous flaps [20]. Potential contraindications to posterior-only total sacrectomy include direct tumor invasion of the rectum or iliac vessels or tumor extension cephalad to the L5/S1 disc space [20].

Our preference is for staged anterior-posterior approaches for both total and partial sacrectomies, especially for malignant tumors in which curative resection is the goal. In addition to the aforementioned benefits of decreased blood loss (after internal iliac ligation) and safer dissection about the rectum and pelvic vasculature, we feel that negative margins can be more reliably obtained from a combined approach. Tumor visualization is best when approached anteriorly, and "scoring" the anterior cortical osteotomy during the first stage prevents subsequent "lifting up" and splintering of the anterior cortex, which can occur in posterior-only osteotomies and which can result in less precise margins.

26.6 Complications

Complications following sacrectomy are common, including surgical site infection, hardware failure, vascular or visceral injury, and CSF leak and pseudomeningocele formation; postoperative bowel, bladder, and sexual dysfunctions are typically expected, as discussed above, following sacrifice of the upper sacral nerve roots. Incidence of major complications is likely increased in total and high partial sacrectomies: 85% vs. 29% for resections above vs. below S2 in one review [59]. In a review of 46 patients undergoing partial or total sacrectomy, Sciubba et al. [60] reported postoperative infection in 39% of patients. Identified risk factors included prior lumbosacral surgery and number of surgeons scrubbed into the case, regardless of length of operation. While *Staph. aureus* was the most frequently cultured organism, the relatively high rate of gram negative infection led the authors to posit that proximity to the rectum, especially in the context of bowel (or bladder) dysfunction, might increase the risk of surgical site infection after sacrectomy. Interestingly, another study of postoperative complications described two *delayed* rectal perforations, both within 2–3 weeks of sacrectomy, thought to result from bowel ischemia in the setting of internal iliac ligation [51]. This series, reporting on complications in 34 patients with primary sacral tumors, identified five infections, three cases of postoperative foot drop (likely resulting from L5 nerve root injury), one case of cutaneous CSF fistula formation, and three perioperative deaths, highlighting the significant risks associated with these surgeries. Hardware failure occurred after total sacrectomy in 16% of patients included in a systematic review [33], though our experience suggests that the use of adjuvant high-dose radiation significantly increases the risk of postoperative fracture (up to 57% [9]), as well as nonunion and hardware failure. Radiation likely increases the risk of infection as well. A recent report from our institution identified wound infections in 10 of 60 patients (17%) undergoing resection of spinal chordomas after neoadjuvant radiation and in 7 of 58 patients (12%) undergoing resection with adjuvant radiation alone [24].

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One-Stage Total Sacrectomy

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Jie Zang and Wei Guo

27.1 Combined Anterior and Posterior Approach

While primary sacral tumors are not common, chordoma and giant cell tumor are the two most common tumor types involving the sacrum. Satisfactory local control of sacral tumors is difficult to achieve due to the anatomic complexity, massive intraoperative bleeding, and challenges realizing adequate margins in this region. Wide resection with a safe margin is the only practical approach to achieve the best local control and cure the disease for patients with primary malignant sacral tumors.

Very few studies have addressed total sacrectomy, for which the most common procedure is a two-stage operation using a combined anterior and posterior approach. In the two-stage operation, the bilateral internal iliac arteries are mobilized from the sacrum by ligating the branches of the iliac vessels, and the soft tissue in front of the tumor is dissected to facilitate resection of the L5–S1 disc using an anterior approach by laparoscopic or open surgery. The total sacrum is then removed using a posterior approach. The two surgical stages are usually performed a few weeks apart. Intra-abdominal surgery often leads to intestinal obstruction. To address these issues, we performed total sacrectomy with a one-stage operation that was achieved using a combined anterior and posterior approach. In this chapter, we describe this surgical procedure carried out in our center.

The indication for one-stage total sacrectomy is malignant sacral tumors invading S1–S2. The relative indication for this procedure is a giant cell tumor extensively invading S1–S5.

Surgical procedure: Bilateral ilioinguinal incisions are made to visualize the retroperitoneal space. The bilateral iliac vessels are mobilized from the sacrum by ligating the branches of the iliac vessels. Soft tissue in front of the tumor is dissected away, and the L5–S1 disc is resected as much as possible. After identifying the upper and lower edge of the sacroiliac joint, two ends of a plastic tube are introduced from anterior to posterior with a thick needle. A wire saw is then passed through the tube to accomplish the osteotomy from the outer edge of the sacroiliac joint via the posterior approach (Fig. 27.1).

A longitudinal midline incision is made in the posterior approach. The gluteus maximus muscles and parasacral muscles are dissected away, and the lamina and spinal process of L4–L5, posterior aspect of the sacrum, and bilateral sacroiliac joints are visualized. Four transpedicle screws are inserted into the L4 and L5 vertebrae before resection of the sacrum. After visualizing the posterior aspect of the sacrum, the anterior aspect of the sacrum is dissected from the surrounding tissue. The rectum is identified from the posterior approach after resection of the sacrotuberous ligament, sacrospinous ligament, and sacrococcygeal ligament. The rectum is carefully separated and gauze packed into the space between the rectum and anterior aspect of the sacral tumor to push the rectum forward so that the integrity of the rectum is maintained during tumor dissection. The inferior articular process of L5 and the lamina above the tumor are resected, and the bilateral L5 nerve roots and dura sac are carefully preserved. Osteotomies of the bilateral iliac bone are performed along the outer edge of the sacroiliac joints with wire saws passed through the silicone tubes that were placed during the anterior approach. The sacrum is resected at the level of the L5–S1 disc, and then the whole sacrum is removed (Fig. 27.2).

After total sacrectomy, four pedical screws are placed into the residual ilium, and two or four rods are applied to connect the screws in the ipsilateral lumbar region and ilium. Complex reconstruction with a screw-rod system and a fibular bone graft is preferred for young patients.

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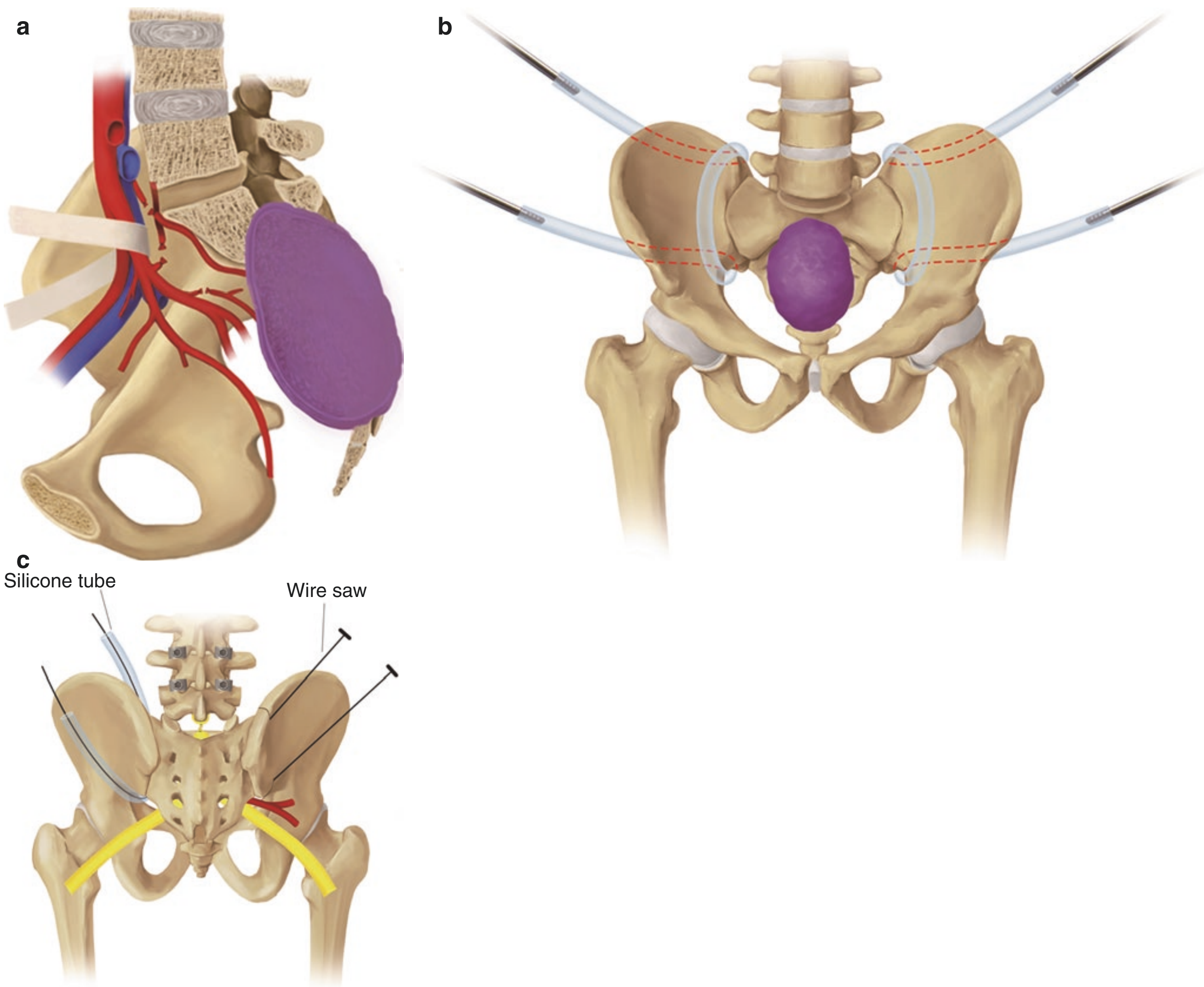

Fig. 27.1 (**a**–**c**) Schematic diagram of surgical procedure for one-stage total sacrectomy (**a**) Coronal view revealing the mobilization of the bilateral iliac vessels from the sacrum after ligation of the branches of the iliac vessels. (**b**) Frontal view revealing application of the silicone tube from the anterior to posterior aspect of the sacrum. (**c**) Dorsal view showing the passage of the wire saw though the silicone tube

27.2 The Single Posterior Approach

Before performing total en bloc sacrectomy using a posterior-only approach, the surgeon must consider three major issues: (1) carrying out an osteotomy through the ilium without injuring the viscera, lumbosacral trunk, and bilateral iliac vessels; (2) mobilizing the internal iliac vessels from the soft tissue anterior to the tumor; and (3) excising the L5–S1 disc without injuring the surrounding vital vessels attached tightly to the L5 vertebrae.

Indications for total en bloc sacrectomy using a posterior-only approach in our center are (1) primary malignant sacral tumor with S1–S2 involvement; (2) tumor not involving the internal iliac vessels and bowel, evaluated by imaging studies and digital rectal examination; and (3) recurrent sacral tumors without a history of an anterior procedure.

Surgical procedure: Prior to surgery, the main arteries feeding the sacral tumor are selectively embolized by the radiologist. The arteries are embolized based on the blood supply to the sacrum to occlude the middle sacral, iliolumbar, and lateral sacral arteries. The bilateral internal iliac arteries are not commonly embolized. After preoperative embolization, the patient is taken directly to the operation room, receives general anesthesia, and is placed in a supine position, and an aortic balloon is inserted into the abdominal aorta by femoral access. The aortic balloon is inflated during the tumor resection to reduce intraoperative blood loss. We recently described this technique, and it is

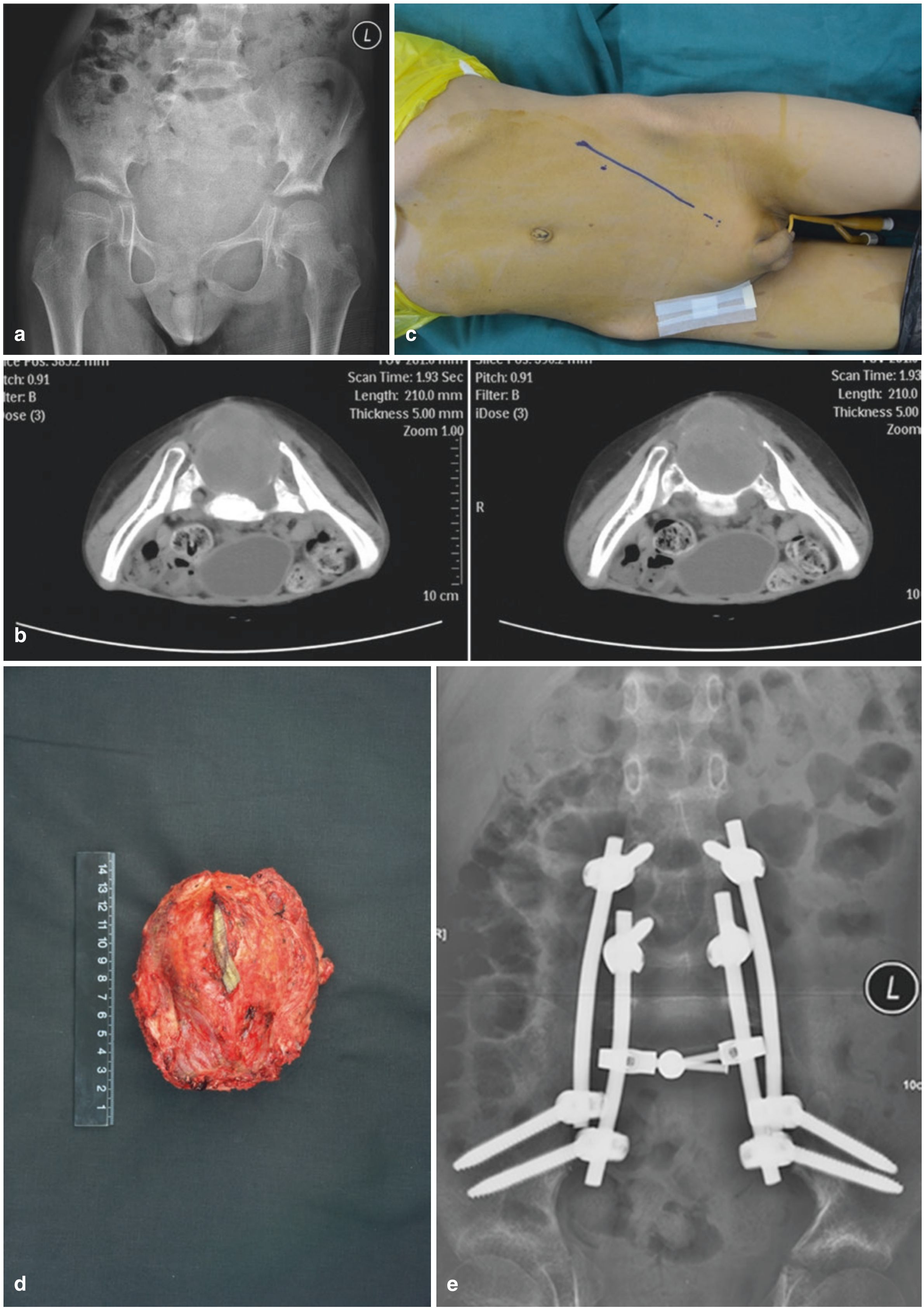

Fig. 27.2 (**a**–**e**) A male patient with malignant peripheral nerve sheath tumor involving the sacrum treated with total sacrectomy. (**a**) Plain film revealing osteolysis in the sacrum. (**b**) CT image revealing a soft tissue mass and destruction of the sacrum. (**c**) Abdominal incision used in the patient. (**d**) Removal of the whole sacrum and L5 vertebra. (**e**) Plain film revealing reconstruction of lumbosacral stability after resection

performed in patients with large sacral tumors requiring extensive dissection. The patient is then moved into a prone position. An inverted "Y" incision is applied to reveal the posterior aspect of the sacrum and the adjacent ilium. A posterior longitudinal midline incision is made in the lumbosacral region, and two other incisions are made beginning from the end of the midline incision and going along the fibers of the gluteus maximus muscle. To reveal the posterior aspect of the bilateral ilium and the sacrum, and the spinal process and lamina of L4–L5, the gluteus and the parasacral muscles are carefully dissected away. Before resection of the sacrum tumor, four pedicle screws are placed into the L4 and L5 pedicles. After revealing the posterior aspect of the sacrum, the anterior aspect is separated. Visualization of the rectum is achieved after resecting the sacrotuberous, the sacrospinous, and the sacrococcygeal ligaments. The rectum is carefully separated, and gauze is packed into the space between the rectum and anterior aspect of the sacral tumor to push the rectum forward to maintain the integrity of the rectum during the tumor dissection (Fig. 27.3). The inferior articular process of L5 and the lamina above the tumor are resected, and the bilateral L5 nerve roots and dura sac are carefully preserved. Ligation and division of the dura sac below L5 are then performed. Resection of the L5–S1 disc is carried out using a posterior approach. Excision of the bilateral transverse processes of L5 is necessary so that blunt dissection from the lower and upper edge of the sacroiliac joints can be achieved. To maintain the integrity of the internal iliac vessels and lumbosacral trunks during the osteotomy, the internal iliac vessels and lumbosacral trunks are pushed away from the iliac osteotomy lines by packing gauze in front of the sacroiliac joints. After identifying the lower and upper edges of the sacroiliac joint, a silicone tube is placed in the anterior aspect of the sacroiliac joint. A wire saw is passed through the tube to achieve the osteotomy along the outer edge of the sacroiliac joints with an adequate margin. After osteotomy of the bilateral ilium, the sacrum can be easily lifted up, which may facilitate the visualization of the sacral nerves, lumbosacral trunks, rectum, and iliac vessels. The branches of the internal iliac vessels to the sacrum and the middle sacral artery are identified and ligated. The anterior aspect of the tumor is carefully dissected, and the sacral nerve roots are resected. The total sacrum is then removed (Fig. 27.4).

After total sacrectomy, four pedical screws are placed into the residual ilium, and two or four rods are applied to connect the screws in the ipsilateral lumbar region and ilium. Complex reconstruction with a screw-rod system and a fibular bone graft is preferred for young patients. We recently performed reconstruction with a 3D-printed custom-made prosthesis after the tumor resection (Fig. 27.5).

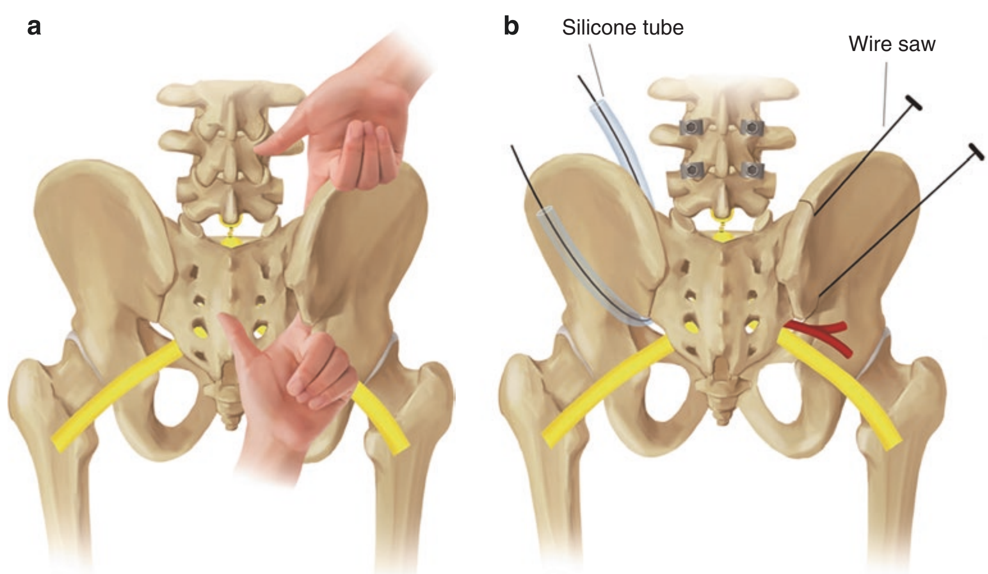

Fig. 27.3 (**a** and **b**) Schematic diagram of the operation procedure for total en bloc sacrectomy. (**a**) Blunt dissection by fingers from the lower and upper edge of the sacroiliac joint. (**b**) Application of the silicone tube in the anterior space of the sacroiliac joint and passage of the wire saw though the silicone tube

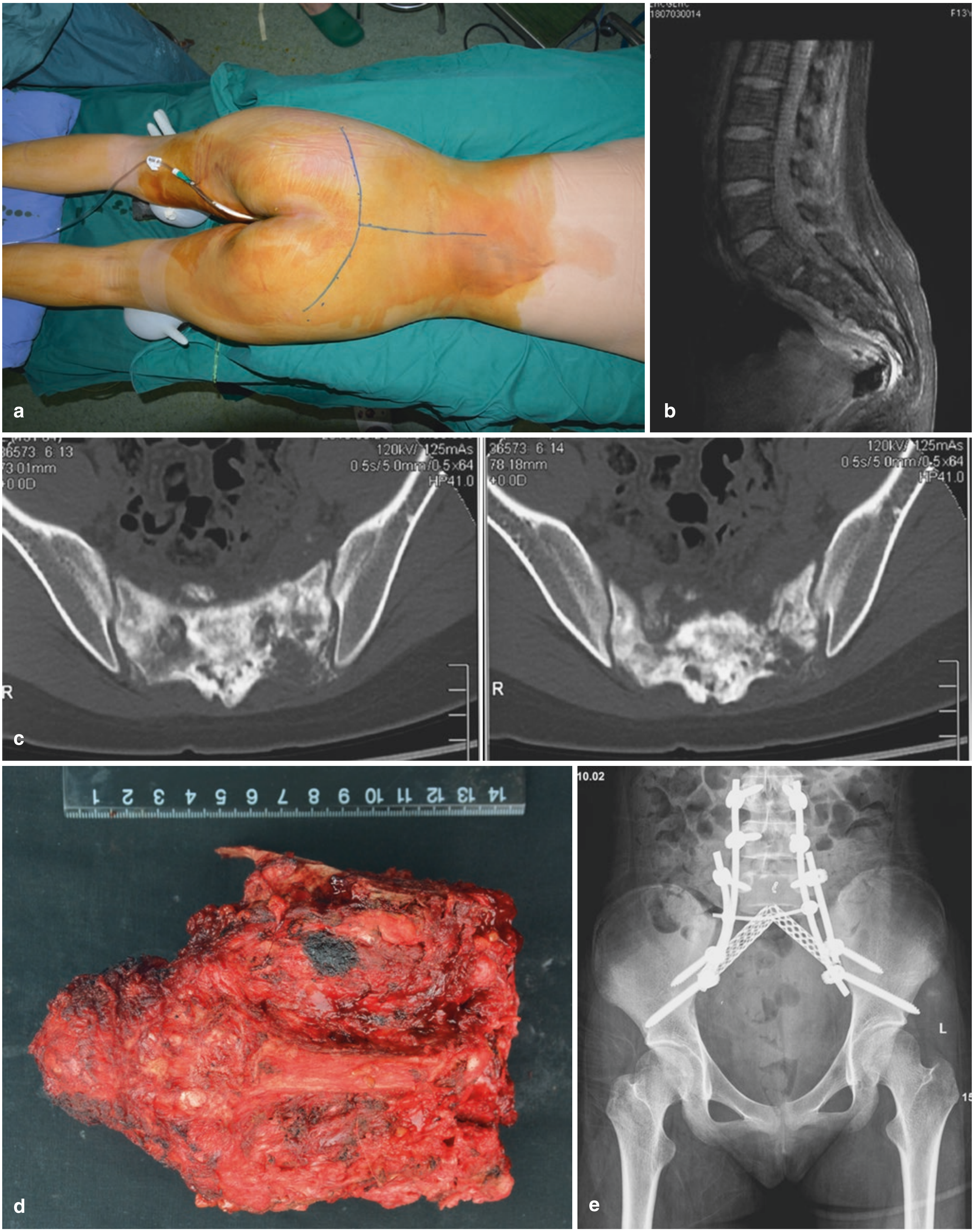

Fig. 27.4 A female patient with sacral osteosarcoma involving S1–S5 treated with total sacrectomy. (**a**) The patient was placed on the supine position and an inverted "Y" incision was used. (**b**) Magnetic resonance image showing low signal intensity on T1WI in the sacrum. (**c**) CT image showing a soft tissue mass resulting in the destruction of the sacrum. (**d**) Image of the entire sacrum. (**e**) Plain film showing the reconstruction of the lumbosacral stability after total sacrectomy

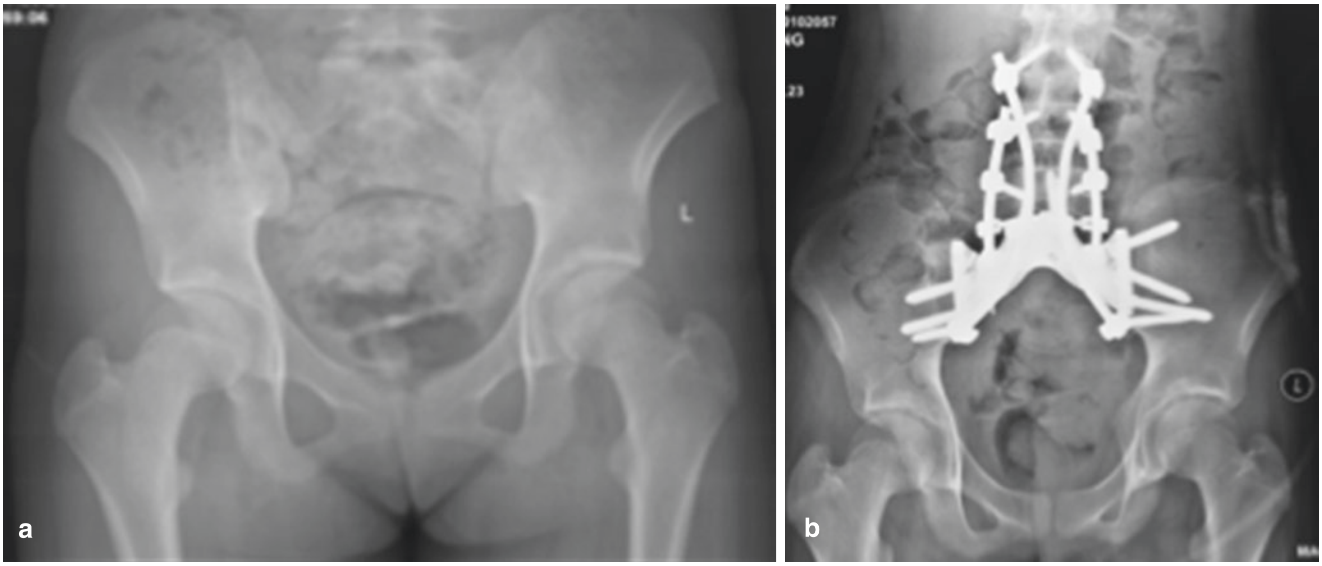

Fig. 27.5 (**a**) Preoperative plain film of a 14-year-old female patient with sacral osteosarcoma. (**b**) Postoperative plain film of the reconstruction with a sacral prosthesis

Using 3D Printing Sacral Endoprosthesis for Spinopelvic Reconstruction

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Wei Guo

Primary sacral tumor is rare, for which surgical resection is the cornerstone of therapy [1, 2]. For primary sacral malignancies involving the upper sacrum, the main treatment is total en bloc sacrectomy (TES). Although the functional outcome of TES-treated patients without spinopelvic reconstruction has been reported as acceptable [3], the bone defect resulting from TES which leads to the discontinuity between spine and pelvis often requires reconstruction because of the facilitation for early mobilization which precludes the complications in patients who are bedridden for a long time [4]. According to the classification proposed by Bederman et al., the reconstruction methods after TES can be categorized into three types: spinal pelvic fixation (SPF), posterior pelvic ring fixation (PPRF), and anterior spinal column fixation (ASCF) [5]. It was suggested that a combined reconstruction including ASCF would be the optimal reconstructive method after TES [5]. However, the combined reconstruction including ASCF conceivably has an increased risk of prolonged surgical time and massive intraoperative hemorrhage, which would impair the safety of the procedure. To address this problem, several unconventional reconstruction methods aiming at synthesizing SPF/SPF+PPRF and ASCF, such as reimplantation of extra-corporeally irradiated sacrum and endoprosthesis replacement, had been reported [6, 7], which, however, could hardly show advantages over another due to the limited number of cases. In general, the standardized reconstructive method for TES-treated patients remains controversial.

We designed and applied a 3D-printed sacral endoprosthesis for the reconstruction of spinopelvic stability after TES to synthesize the biomechanical characteristics of SPF, PPRF, and ASCF in one step with the induction of the bond in-growth on bone-endoprosthetic interfaces by 3D printing trabecular structure [8–11].

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28.1 The Design and Manufacture of 3D-Printed Sacral Endoprosthesis

The morphological design of 3D-printed sacral endoprosthesis was based on the database including computed tomography (CT) scanning data of nearly 100 patients who underwent TES in our center. The design of the prosthesis stemmed from the concept of an endoprosthesis with porous bone–implant interfaces that could connect lumbar spine and ilium, connect both sides of ilium, and rebuild the structure of loading transfer through anterior spinal column in one step while conducive to bone in-growth to the trabecular pores [8]. Biomechanically, it has been confirmed by finite element analysis that the endoprosthesis we designed showed similar diffuse distribution of stress compared to the combined reconstruction including ASCF (Fig. 28.1).

The endoprosthesis consisted of three bone-contacting surfaces: the proximal surface fit to the contour of inferior endplate of L5 vertebrae to reconstruct the lumbarsacral joint; the surfaces on both flanks were matched to bilateral iliac osteotomic planes to reconstruct both sacroiliac joints. Screw holes were predrilled on every bone-contacting surface for fixation. Two screw heads were placed on the dorsal surface to connect with the pedicle screws of lumbar spine with titanium rods (Fig. 28.2).

The endoprosthesis was produced from titanium alloy and manufactured by 3D printing technique. Electron beam melting (EBM) was used in fabrication by successive layering of melted titanium alloy. The bone-contacting surfaces were porous to facilitate the bone in-growth. The endoprosthesis was manufactured in three different sizes to fit the real size of the intraoperative bone defect. The plastic models, of which the shapes were consistent to the corresponding endoprosthesis, were simultaneously manufactured by 3D printing technique to facilitate the selection of appropriate size of endoprosthesis during surgery (Fig. 28.3).

Although the endoprosthesis was designed and manufactured modularized, before surgery, the CT scanning data of the patient would be imported to the computer to simulate

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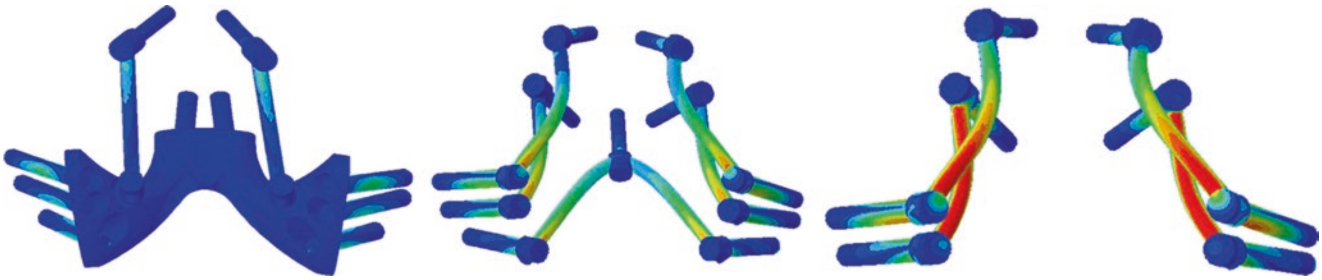

Fig. 28.1 The distribution of von Mises stress on different reconstructive systems. Left, 3D printing sacral endoprosthesis; Middle, combined reconstruction including ASCF; and right, SPF

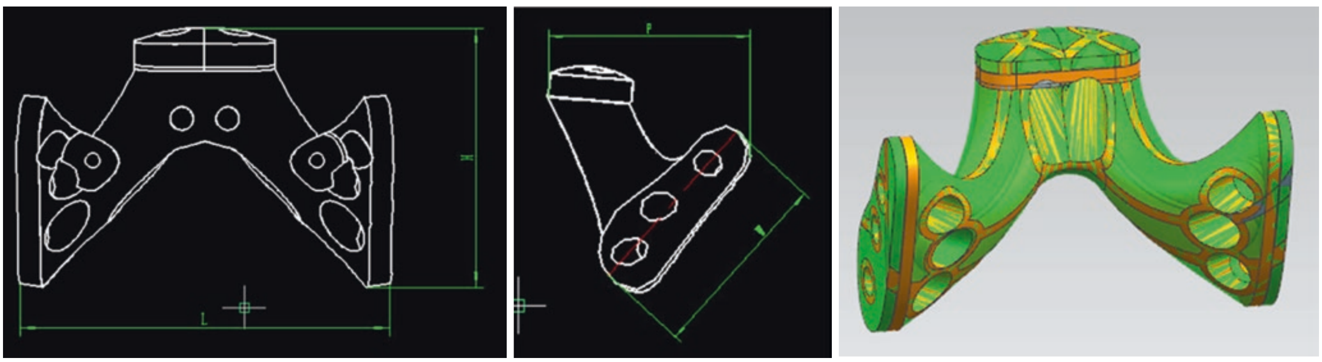

Fig. 28.2 The blueprints of 3D printing sacral endoprosthesis

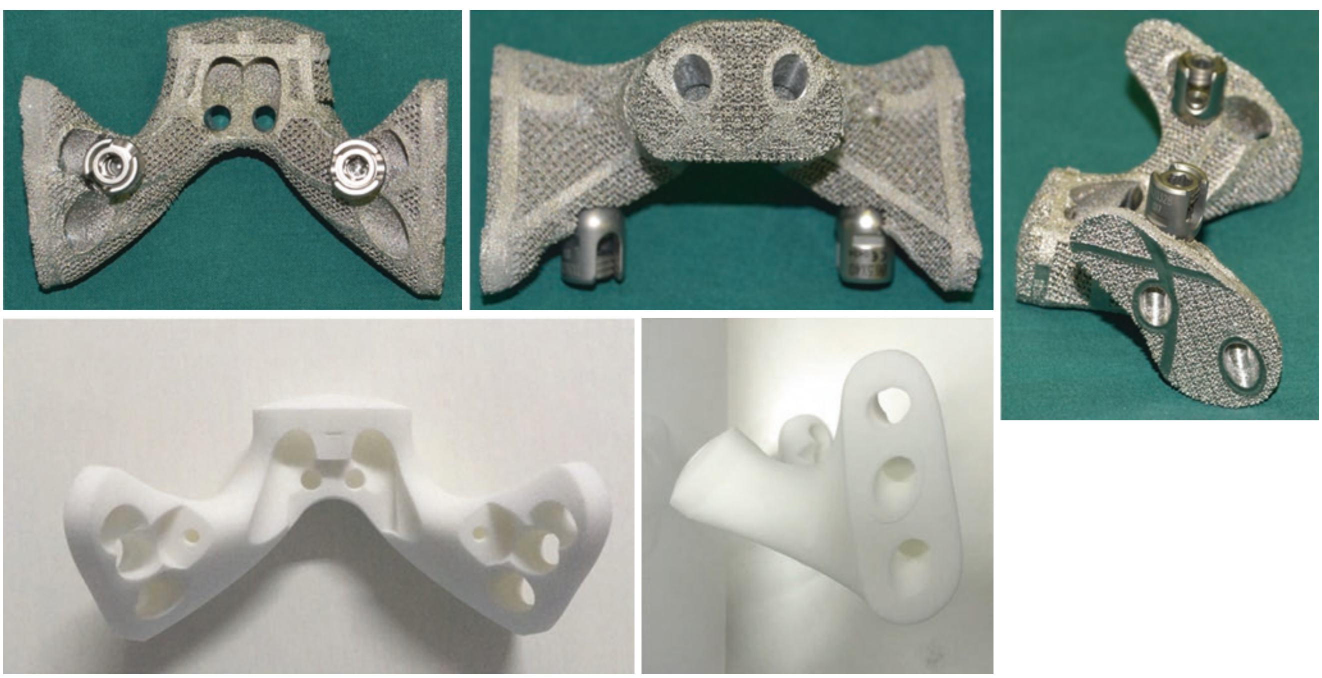

Fig. 28.3 The 3D printing sacral endoprosthesis and plastic models

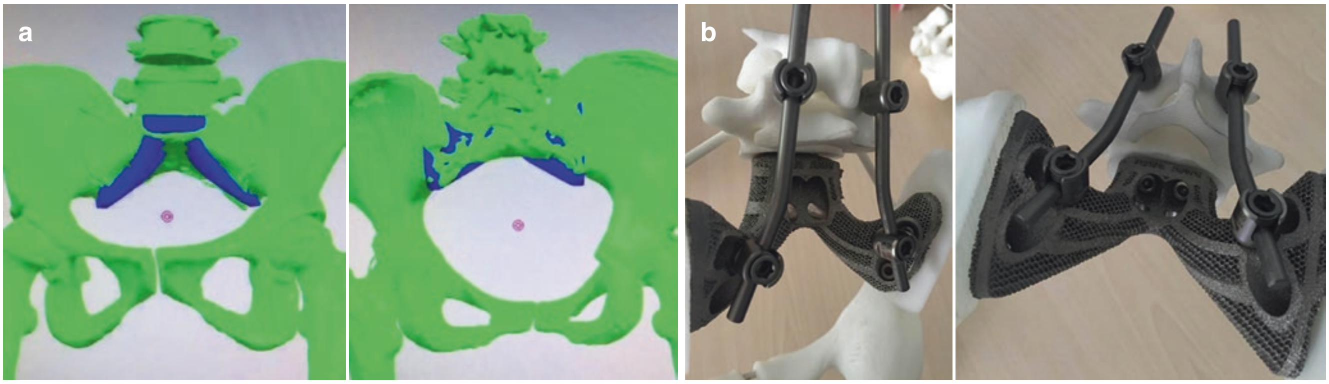

Fig. 28.4 The preoperative simulation of 3D printing sacral endoprosthesis installation. (**a**) Computer simulation; (**b**) rehearsal of the TES and endoprosthesis installation on plastic model

the procedure of TES and installation of the endoprosthesis and determine if the endoprosthesis could be used for reconstruction (Fig. 28.4a). If the endoprosthesis was found to hardly match the potential bone defect during simulation, a custom-made endoprosthesis would be warranted to be manufactured. During the preliminary stage of using the endoprothesis, using computer simulation, the plastic model of the particular patient's spinopelvic anatomic structure would be manufactured by 3D printing technique to rehearse the procedure of TES and endoprosthesis installation (Fig. 28.4b).

28.2 Reconstructive Procedure

After the tumor-bearing sacrum was en bloc resected, the plastic model was placed into the bone defect to determine the size of endoprosthesis. The corresponding endoprosthesis was then settled in the bone defect and fixed to the L5 vertebrae and both sides of ilium by screws through the predrilled holes. Then rods were installed to connect the spine and the endoprosthesis through lumbar pedicle screws and screw heads on the endoprosthesis. In addition to these fixations, SPF and/or PPRF could be supplemented in order to strengthen the reconstruction system (Fig. 28.5).

28.3 Illustrative Cases

A 14-year-old female patient had previously developed severe pain in the buttocks and left lower extremity. A sacral Ewing's sarcoma/PNET involving the upper sacrum was identified by biopsy (Fig. 28.6a). She received neoadjuvant chemotherapy and underwent one-stage TES through posterior approach and 3D-printed sacral endoprosthesis combined SPF reconstruction (Fig. 28.6b). The surgery took 330 min and was accomplished smoothly, during which the volume of bleeding was 2400 ml. The postoperative X-ray is shown in Fig. 28.6c.

The postoperative pathological diagnosis was confirmed to be Ewing's sarcoma/PNET. During the perioperative period, she had a wound healing problem, which was cured by a debridement. The follow-up time was 16 months. At 1 year after surgery, X-ray showed no evidence of implant failure (Fig. 28.6d) and CT scan showed new bone formation in the bone–implant interfaces (Fig. 28.6e). She could walk without aids at last follow-up (Fig. 28.6f).

28.4 The Advantages of Using 3D Printing Sacral Endoprosthesis for Reconstruction of Spinopelvic Stability After TES

To identify the advantages of using 3D printing sacral endoprosthesis for reconstruction of spinopelvic stability after TES, we summarized the clinical data of TES-treated patients reconstructed by endoprosthesis and compared it to that of patients who received combined reconstruction including ASCF (optimal conventional reconstructive method) and received only SPF reconstruction (elementary reconstruction) respectively in corresponding period.

The spinopelvic stability, implant survival (IS), surgical time, intraoperative hemorrhage and perioperative complication rate of patients were documented and compared. In light of the reconstructive method, we categorized patients into three groups: endoprosthesis group included 10 patients, combined reconstruction group included 14 patients, and SPF group included 8 patients. The spinopelvic stability was assessed using the scoring of pain and motor in the scoring system for evaluating neurologic deficit after sacral resection, which was proposed by us [12].

In endoprosthesis group, the mean surgical time and intraoperative hemorrhage was 392.5 min and 3530 ml, respectively.

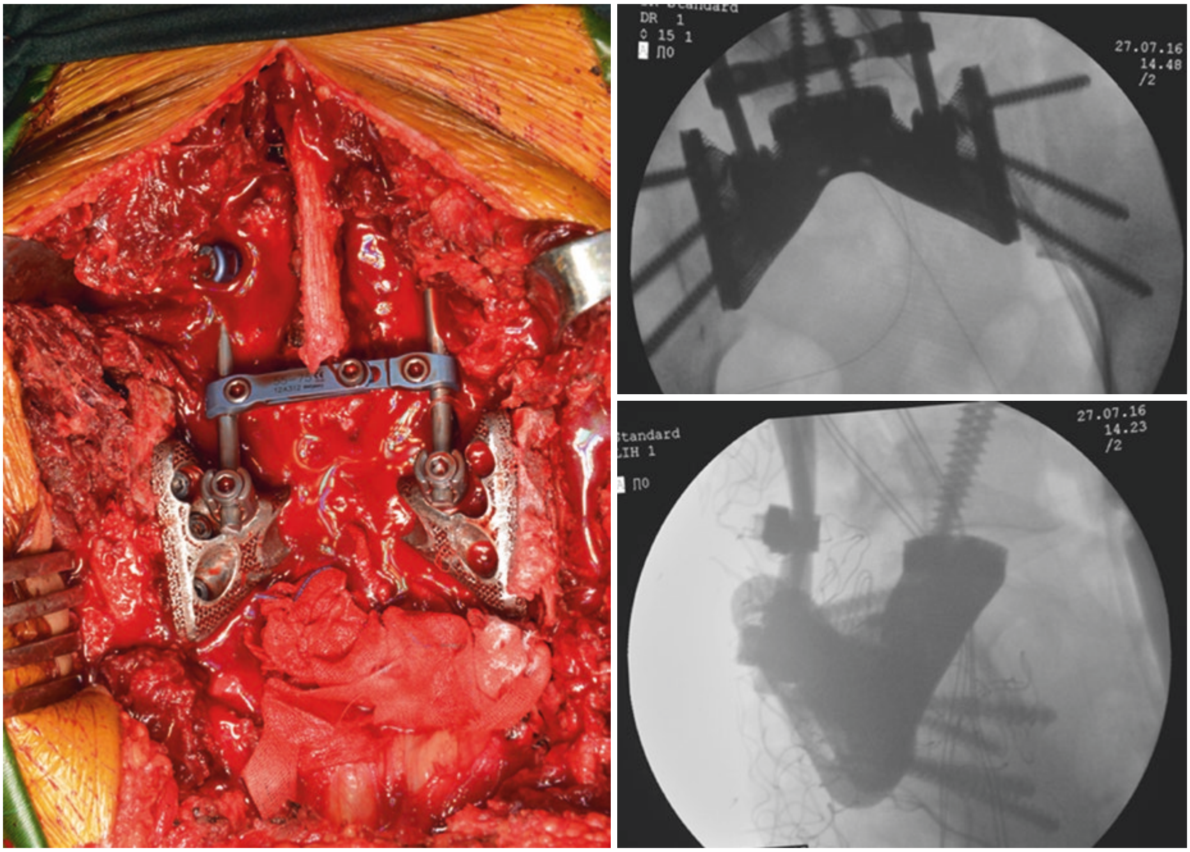

Fig. 28.5 Intraoperative photograph and X-rays showing the endoprosthesis fixed in the bone defect between lumbar and pelvis by screws and rods

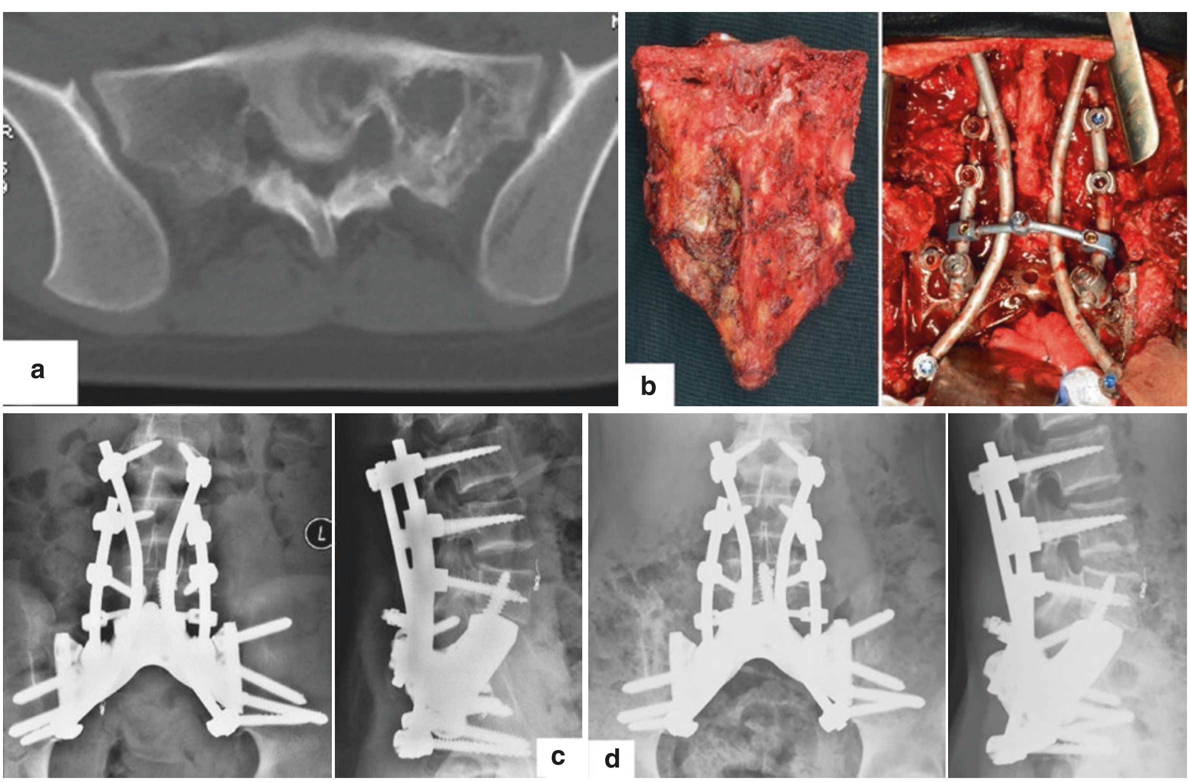

Fig. 28.6 Female, 14 years, sacral Ewing's sarcoma/PNET. (**a**) Preoperative CT. (**b**) Tumor-bearing sacrum was en bloc resected and endoprosthesis was settled. (**c**) X-ray 3 weeks after surgery. (**d**) X-ray 12 months after surgery. (**e**) CT scan 12 months after surgery showed new bone formation (arrow). (**f**) She could walk without aids at last follow-up

Fig. 28.6 (continued)

Perioperative complications occurred in 2 patients and all were wound healing problems. After a mean follow-up of 21.3 months, 9/10 patients could walk without aids and 8/10 patients were without using analgesic. The imaging evidence of implant failure was found in 3 patients and all of them were breakage of screws and/or rods, of whom only 1 patient with local recurrence received reoperation in which the rigid bone-endoprosthetic osseointegration was found, while other 2 patients dispensed with reoperation. The mean IS using reoperation as endpoint was 39.4 months.

Compared to combined reconstruction group and SPF group, the spinopelvic stability, i.e. the pain and motor scores, in endoprosthesis group were significantly better than those of SPF group and were similar to those of combined reconstruction group. Regarding the IS, in the case of using reoperation as endpoint, the implant failure rate of endoprosthesis group was significantly lower than that of SPF group and was similar to that of combined reconstruction group, and the IS in endoprosthesis group was similar to that of combined reconstruction group and was significantly better than that of SPF group. Moreover, the surgical time, intraoperative hemorrhage, and perioperative complication rates of patients in endoprosthesis group showed no significant difference compared to combined reconstruction group and SPF reconstruction group (Table 28.1) (Fig. 28.7).

In general, the advantages of using 3D printing sacral endoprosthesis could be concluded in three facets.

28.4.1 3D Printing Sacral Endoprosthesis Can Provide Optimal Reconstruction of Spinopelvic Stability After TES

According to our results, the spinopelvic stability results of patients who received endoprosthetic reconstruction were similar to those of patients who received combined reconstruction including ASCF, which had been identified as the optimal reconstructive method, and were significantly superior than those of patients who received SPF only. The endoprosthesis is a one-step reconstructive solution composing three key structures, SPF, PPRF, and ASCF. The necessity of the three structures was proven by finite element analysis.

28.4.2 3D Printing Sacral Endoprosthesis Can Reduce the Risk of Long-Term Implant Failure

According to our results, the IS of patients who underwent endoprosthetic reconstruction was similar to those of patients who received combined reconstruction including ASCF and

Table 28.1 The comparison of surgical safety, spinopelvic stability, and implant survival between three reconstructive options

			Combined reconstruction	*P*	SPF	*P*
Endo-prosthesis	N	10	14		8	
	Pain score	2.5	2.3	0.50	1.8	0.047[a]
	Motor score	2.2	2.1	0.68	1.4	0.028
	Imaging implant failure	3	1	0.18	5	0.18
	IS using imaging implant failure as endpoint (Mos)	16.5 (95% CI 13.8–19.2)	45.1 (95% CI 39.5–50.6)	0.18	24.8 (95% CI 12.1–37.5)	0.33
	Implant failure	1	1	0.67	5	0.032[a]
	IS using reoperation as endpoint (Mos)	39.4 (95% CI 34.6–44.2)	45.1 (95% CI 40.0–50.3)	0.51	24.8 (95% CI 12.1–37.5)	0.032[a]
	Surgical time (min)	392.5	416.4	0.76	368.7	0.80
	Intraoperative hemorrhage (ml)	3530.0	2771.4	0.39	4637.5	0.27
	Perioperative complication	2	8	0.08	2	0.62

[a]Significant difference

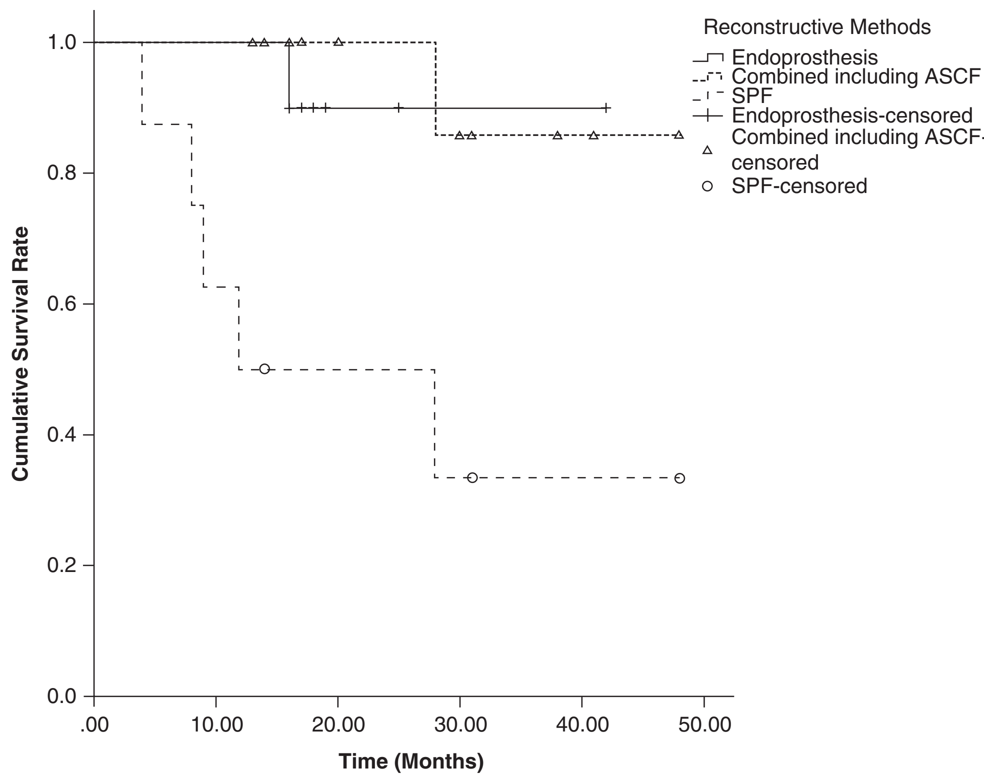

Fig. 28.7 Kaplan-Meier curves show differences in IS using reoperation as endpoint between different reconstructive methods

was significantly better than those of patients who received SPF only. Moreover, the situation of 3 patients with imaging implant failure in endoprosthesis group is worth noting: 2 patients showed no symptoms of spinopelvic instability and dispensed with revision surgery, and 1 patient who received reoperation in which we found the endoprosthesis was hard to take out due to solid scar tissue. It implies that even if the screws and rods failed, the long-term spinopelvic stability can be secured by the rigid osseointegration, which attributes to the bone-ingrowth on the bone–endoprosthesis interface induced by 3D-printed trabecular structure.

28.4.3 Using 3D Printing Sacral Endoprosthesis for Reconstruction Does Not Complicate the Surgical Procedure

According to our results, the surgical time, intraoperative hemorrhage, and perioperative complication rates of patients with 3D-printed endoprosthesis reconstruction were in between of those of patients with combined reconstruction including ASCF and SPF reconstruction in this study without statistical significance. It demonstrates that with a similarly high-level postoperative spinopelvic stability and IS to combined reconstruction including ASCF, using 3D-printed sacral endoprosthesis can simplify the reconstructive procedure to some extent, which may attribute to the one-step reconstruction realized by using preset screw holes and heads on the endoprosthesis for fixation.

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29 Resection of Hemisacrum Vertically and Sacroiliac Joint

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During en bloc sacrectomy, the bilateral sacral nerves usually have to be sacrificed. Postoperative complications, including urinary and fecal incontinence, partial dysfunction of the sciatic nerves, and sexual dysfunction, are usually inevitable [1–3]. *In a small number of cases, sacral malignances may originate from an ala sacralis and only involve the ipsilateral sacral nerves. If the contralateral sacral nerves are not affected by the tumor and are located beyond the resection line, en bloc resection of the hemisacrum vertically is theoretically possible. Thus, a much better postoperative function could be expected in this scenario. This procedure is reserved for a very limited number of cases. Although it is simple in concept, it is much more demanding than one-stage total sacrectomy* [4–6].

29.1 Indication and Contraindication

Contralateral sacral nerve roots are located beyond the planned resection line and can possibly be preserved without sacrificing the resection margin on high-resolution MRI and CT.

If a tumor invades the ipsilateral lumbosacral trunk or the common and/or the external iliac vessels, hemisacrectomy plus external hemipelvectomy might be a better choice.

29.2 Surgical Technique

In hemisacrectomy, the extent of resection includes half or more than half of the sacrum and the ipsilateral SI joints. The sagittal cut line in the sacrum is located between the posterior median line and the medial side of the contralateral sacral foramina (Fig. 29.1). All sacral nerve roots of the contralateral side are preserved during tumor resection.

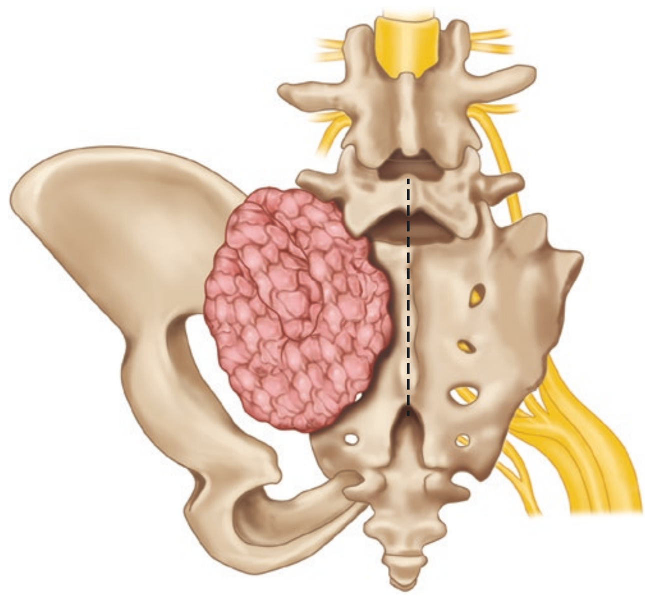

Fig. 29.1 The sagittal cut line in the sacrum is illustrated by a dotted line

Use intra-abdominal aortic balloon occlusion to decrease blood loss during sacral tumor resection. Preoperative embolic occlusion of the internal iliac arteries and/or their branches should be considered for tumors with abundant blood supply.

A combined posterior–anterior approach is the most common approach.

Place the patient in a prone position. Be sure that bolsters are placed longitudinally under the patient's sides, to allow the abdomen to be entirely free, thus reducing venous plexus filling around the spinal cord by permitting the venous plexus to drain directly into the inferior vena cava. Make a posterior vertical midline incision, to expose the posterior aspect of the sacrum and the adjacent two or three lumbar vertebrae. Detach the paraspinal muscles subperiosteally from the bone, as one unit. Expose the sacrum clearly. Remove the L5

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spinal process and perform sacral laminectomy with a safe margin. If the dural sac and the contralateral sacral nerve roots are not invaded by the tumor and can be safely isolated after a careful exploration, hemisacrectomy can be performed.

Subsequently, each ipsilateral nerve root must be identified individually. Double ligate and cut off the ipsilateral sacral nerve roots along the dural sac. Resect the ipsilateral L5/S1 facet joint, cut off the ipsilateral half of the posterior longitudinal ligament, and remove the ipsilateral half of the L5–S1 disc. Expose and cut off the anococcygeal ligament, the ipsilateral piriformis, the sacrotuberous ligament, and the sacrospinous ligament with a safe margin. Dissect the rectum bluntly from the front of the sacrum and pack gauze into the presacral space, to protect the rectum. Perform a standard pedicle screw placement procedure in the bilateral pedicles of L4–L5 and the contralateral pedicle of S1. Connect the three screws on the contralateral side with a rod. Close the posterior incision temporarily.

Turn the patient to a lateral decubitus position. Follow an ilioinguinal incision and elongate it posteriorly to the posterior midline incision. Using a retroperitoneal approach, identify and protect the ipsilateral ureter, mobilize the rectum until exposure of the gauze packed in the presacral space, and ligate the ipsilateral internal iliac artery and the middle sacral vessels. Ligate and cut off the ipsilateral sacral nerves with a safe margin from the tumor. Identify and protect the ipsilateral lumbosacral trunk well. Ligate blood vessels to the tumor and dissect the anterior aspect of the tumor and the ipsilateral sacrum and ipsilateral S-I joint from the noninvolved tissues. Cut off the ipsilateral half of the anterior longitudinal ligament and remove the residual ipsilateral half of the L5–S1 disc. Expose the anterior aspect of the ilium and confirm the anterior cut line in the ilium.

Remove the temporary stitches from the posterior approach. Dissect the ipsilateral gluteus from the sacrum and ilium and expose the sciatic notch. Confirm the safe cut line in the ipsilateral ilium posteriorly. Protect the gluteal blood vessels and nerves, as well as the sciatic nerve, and cut the ilium along the cut line using a Gigli saw or a milling burr. Expose the planned cut line at the anterior bony surface of the sacrum well and cut the sacrum sagittally through a safe margin along the midline or the contralateral sacral foramen using a milling burr or Gigli saw. Remove the specimen en bloc.

A vertebral pedicle screw and rod system is used to reconstruct the bone and joint defect. Two or more pedicle screws are placed in the residual ilium and are connected to the screws placed in the ipsilateral lumbar pedicles or lumbar bodies, using two or more rods. Bone grafting using an allograft or an autograft, such as the fibula, is performed. Wound closure is then performed in the customary manner.

With the development of surgical techniques, resection of the hemisacrum vertically together with the ipsilateral sacroiliac joint can now be performed through a posterior approach in one stage at our center [7, 8].

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Part VII

Sacral Tumors: Special Topics

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30 Lumbosacral Resection and Reconstruction

Matthew T. Houdek, Peter S. Rose, and Michael J. Yaszemski

30.1 Introduction

Sacrectomies (either partial or total) are indicated for a primary malignancy or recurrent pelvic tumors that require a resection for cure. For these patients an en bloc resection is indicated, which can compromise spinopelvic continuity. The extent of resection varies based on the location and type of tumor, and most types of partial sacrectomy do not require reconstruction. Since oncologic outcome is favorable when an en bloc resection can be obtained with negative margins, frequently the reconstructive procedures are hindered with large bony and posterior soft-tissue defects. This combined with the complex anatomy of the pelvis, vascular, and visceral structures make surgery and reconstruction technically complex. The purpose of this chapter is to describe the indications and also techniques for sacral reconstruction.

30.2 Indications for Reconstruction

Through the sacroiliac joints, the sacrum is the only mechanical connection between the axial skeleton and the lower extremity. This joint is highly constrained and has to resist not only compression but also rotation. Biomechanical studies have shown that many partial sacrectomies are well tolerated and those that do not violate the sacroiliac (SI) joints do not need reconstruction. In normal conditions, the wedge shape of the sacrum between the iliac wings provides an inherent stability to prevent caudal migration. Similarly, the broad and irregular surface of the SI joint forms an interlocking articulation which resists motion. This is further stabilized by the stout sacroiliac, sacrotuberous, sacrospinous, and lumbosacral ligaments.

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Although debated, reconstruction of the sacrum is thought to be necessary, since following the resection, the axial skeleton becomes supported only with soft tissue. Cadaveric work by Gunterberg and colleagues has shown that transverse resections of the proximal sacrum involving the ala weaken the ability of the pelvis to resist an axial load applied to the lumbosacral junction [1, 2]. Although the resections weakened the pelvis by up to 50%, based on the published literature at that time, the authors felt it did not weaken the pelvis to an extent that would prevent safe weightbearing [2].

Further work by Hugate and colleagues on a cadaveric model showed that if a transverse partial sacrectomy was performed cranial to the S1 nerve root (Fig. 30.1a), then reconstruction should be performed; however if it were distal to S1 (Fig. 30.1b), the authors felt the pelvis would be able to withstand postoperative mobilization [3]. In cadaver specimens from younger patients, Yu and colleagues showed that resection involving S1 resulted in rotational instability and with further resection cephalad through half of S1 resulted in compressive instability [4]. Based on these findings, the authors recommended reconstruction when the osteotomy was at or above the S1/S2 level [4]. Currently, at our institution we advocate for reconstruction if there is a transverse osteotomy above the S1/S2 level or removes at least one of SI joints. Functionally, it has been our experience that the best results were observed when the continuity between the pelvis and sacrum was either maintained or reconstructed [5, 6].

30.3 Types of Reconstruction

Pedicle and iliac screw fixation is currently the standard technique to stabilize the lumbosacral junction following sacrectomy. Although it restores spinal balance, historically this has been associated with a high failure rate related to screw loosening and breakage, especially with single-screw fixation and an all-posterior spinal column support [7, 8]. Anatomically the ilium is separated into two columns, the upper (posterior superior iliac spine to the iliac crest) and

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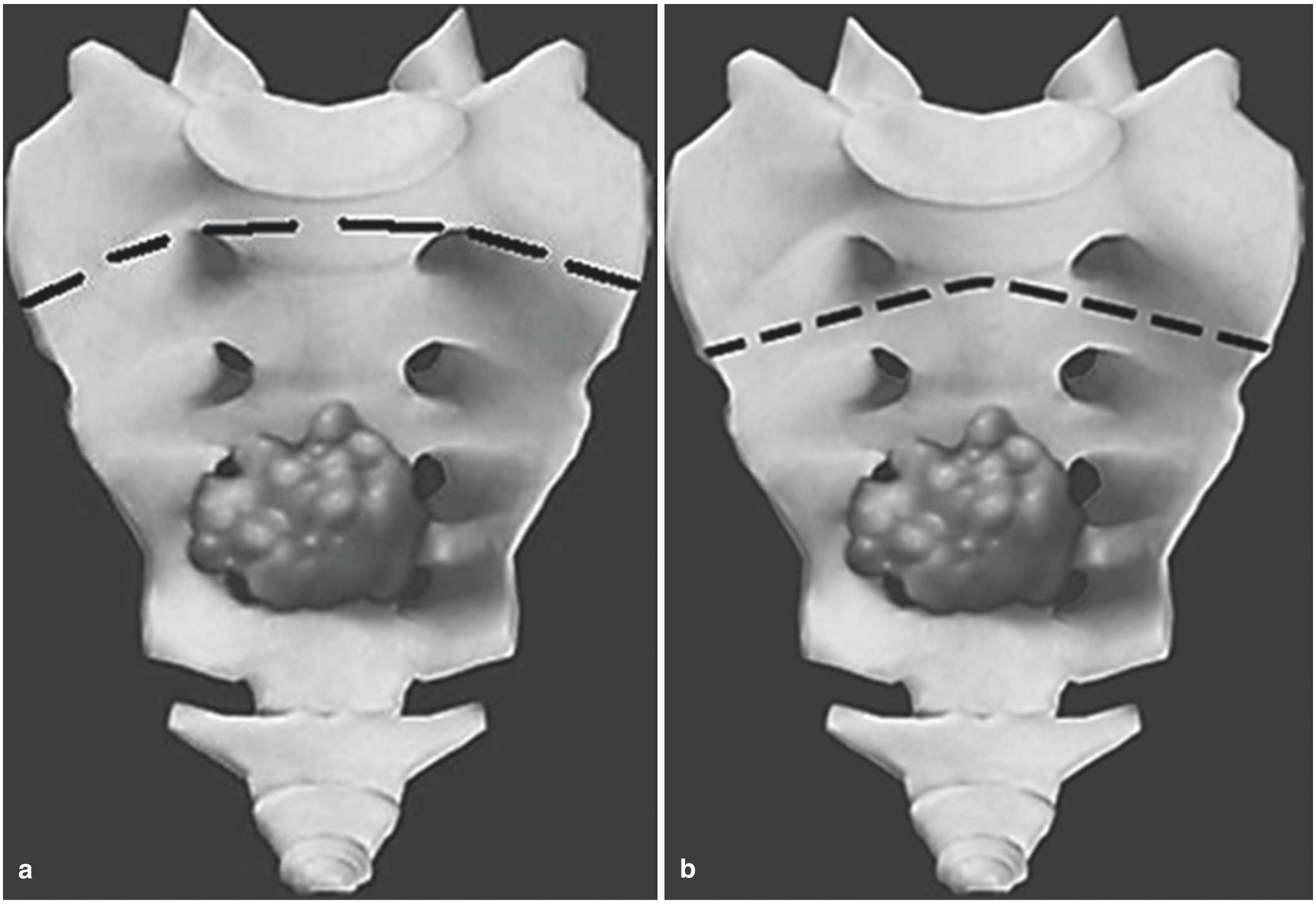

Fig. 30.1 Sacrectomies performed above the S1 neural foramen (**a**) were found to be unable to withstand postoperative mobilization compared to osteotomies performed below the S1 level (**b**). Due to these findings, reconstruction is recommended, when an osteotomy is performed cranial to the S1 foramen

lower segment (posterior superior spine to the anterior inferior iliac spine). These areas have abundant cancellous bone, which provides sufficient space for anchoring iliac screws. Work by Yu and colleagues has shown superior resistance to compression and torsional forces if dual iliac screw constructs are utilized, especially with dual lower segment fixation [9].

In addition to the use of dual iliac screws, a change in rod instrumentation and supplementation has increased the resistance of failure from historic reconstruction techniques. Kelly and colleagues were able to show in an in vitro total sacrectomy model that the four-rod reconstruction technique provided significantly greater stability in flexion and extension compared to the traditional two-rod construct [10]. Similarly, if cross-links were added to the four-bar construct, there was a significant increase in the stability in rotation [10]. In addition to a four-rod reconstruction, the addition of strut grafts increases the stability of the reconstruction, forming a “cathedral” reconstruction of the sacrum, which is currently our technique of choice [6].

At our institution, a reconstruction is performed when the SI joint has been disrupted based on the Mayo Sacral Tumor Classification (Fig. 30.2), either unilateral hemisacrectomy (Mayo Type 2) or total sacrectomy (Mayo Type 1A); above the level of the S1 foramen (Mayo Type 1B); or in the setting of an amputative procedure such as a hemipelvectomy and hemisacrectomy (Mayo Type 3) or total sacrectomy and hemipelvectomy (Mayo Type 4).

30.3.1 Cathedral Reconstruction

The total sacrectomy is performed in a staged anterior and posterior procedure, with a third stage for reconstruction if needed (Fig. 30.3a). Prior to the extirpative surgery, a rectus abdominis musculocutaneous flap (RAM) is elevated. The largest RAM flap that can be elevated and can still allow the abdomen to be closed is planned (Fig. 30.3b). The flap is either a vertical rectus abdominis musculocutaneous (VRAM) or a transverse rectus abdominis musculocutaneous (TRAM)

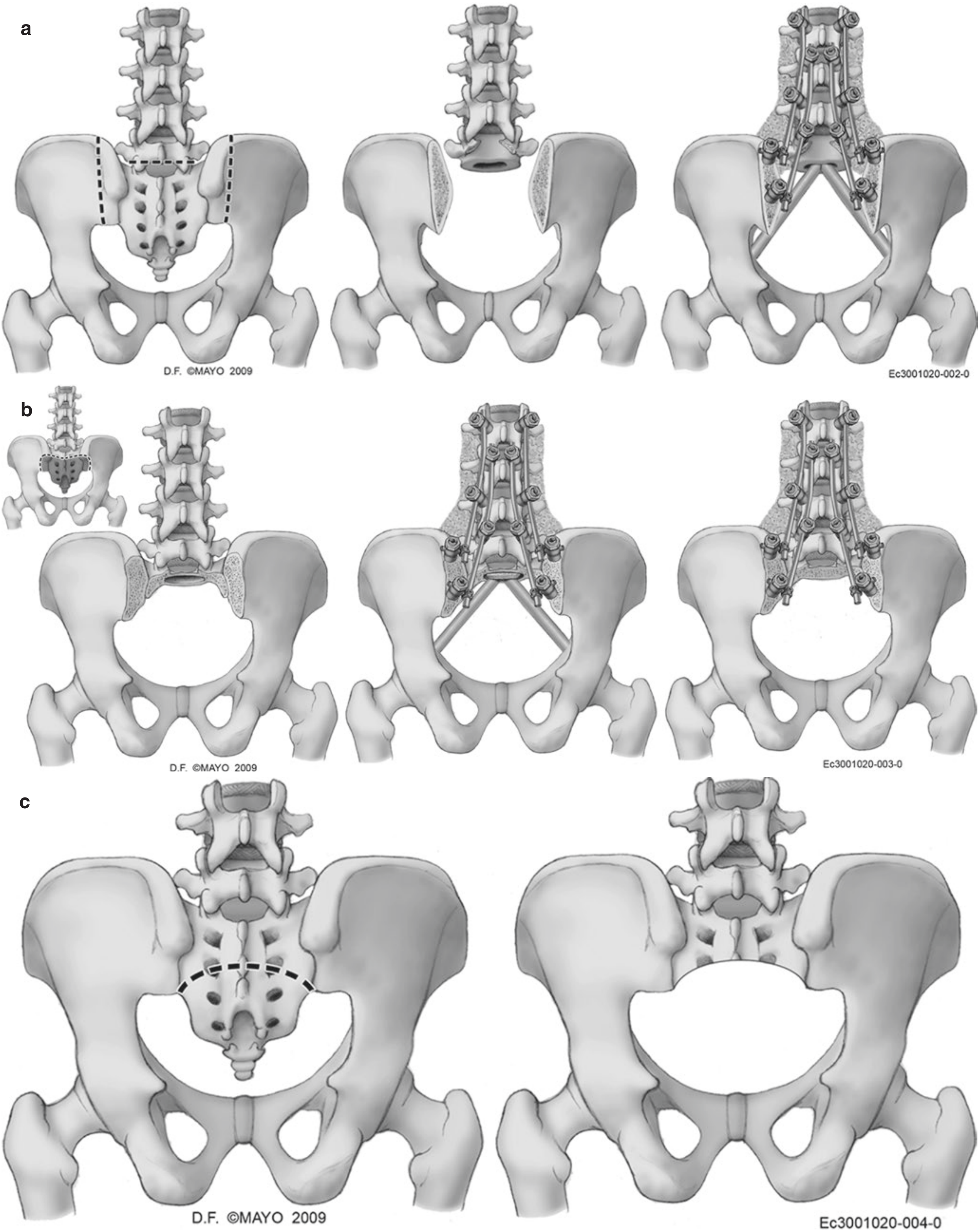

Fig. 30.2 The Mayo Sacral Tumor Classification assists in the planning of reconstruction based on the extent of resection and also if there is an amputative pelvic procedure. We perform a spinopelvic reconstruction for cases of Mayo Type 1A (total sacrectomy, **a**), Type 1B (subtotal sacrectomy above the S1 foramen, **b**), Type 2 (hemisacrectomy involving the ipsilateral sacroiliac joint, **d**), Type 3 (hemipelvectomy and hemisacrectomy, **e**), and Type 4 (total sacrectomy and hemipelvectomy, **f**). Since the SI joints are not disrupted in a Type 1C (subtotal sacrectomy below the S1 foramen, **c**), a reconstruction is not typically performed

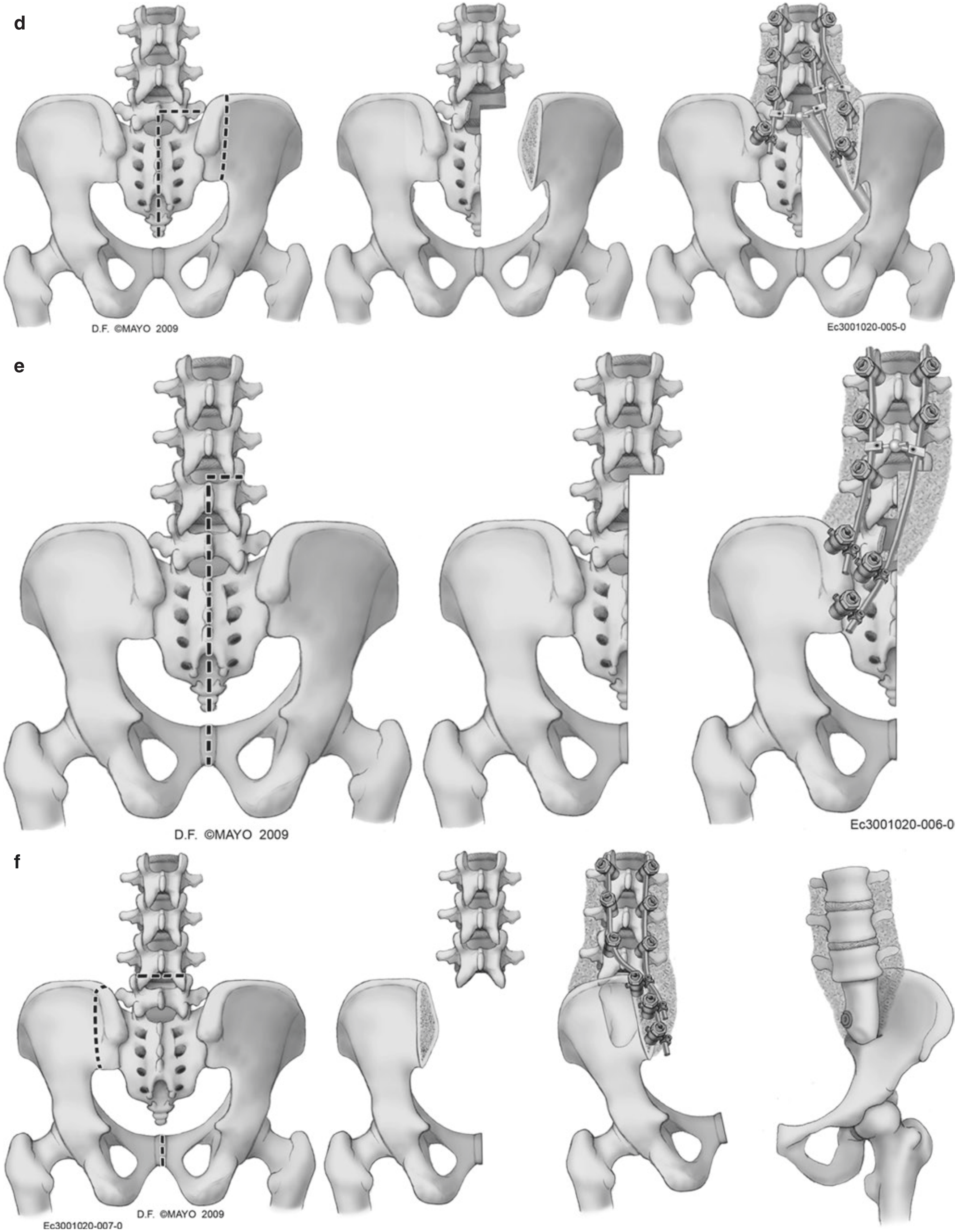

Fig. 30.2 (continued)

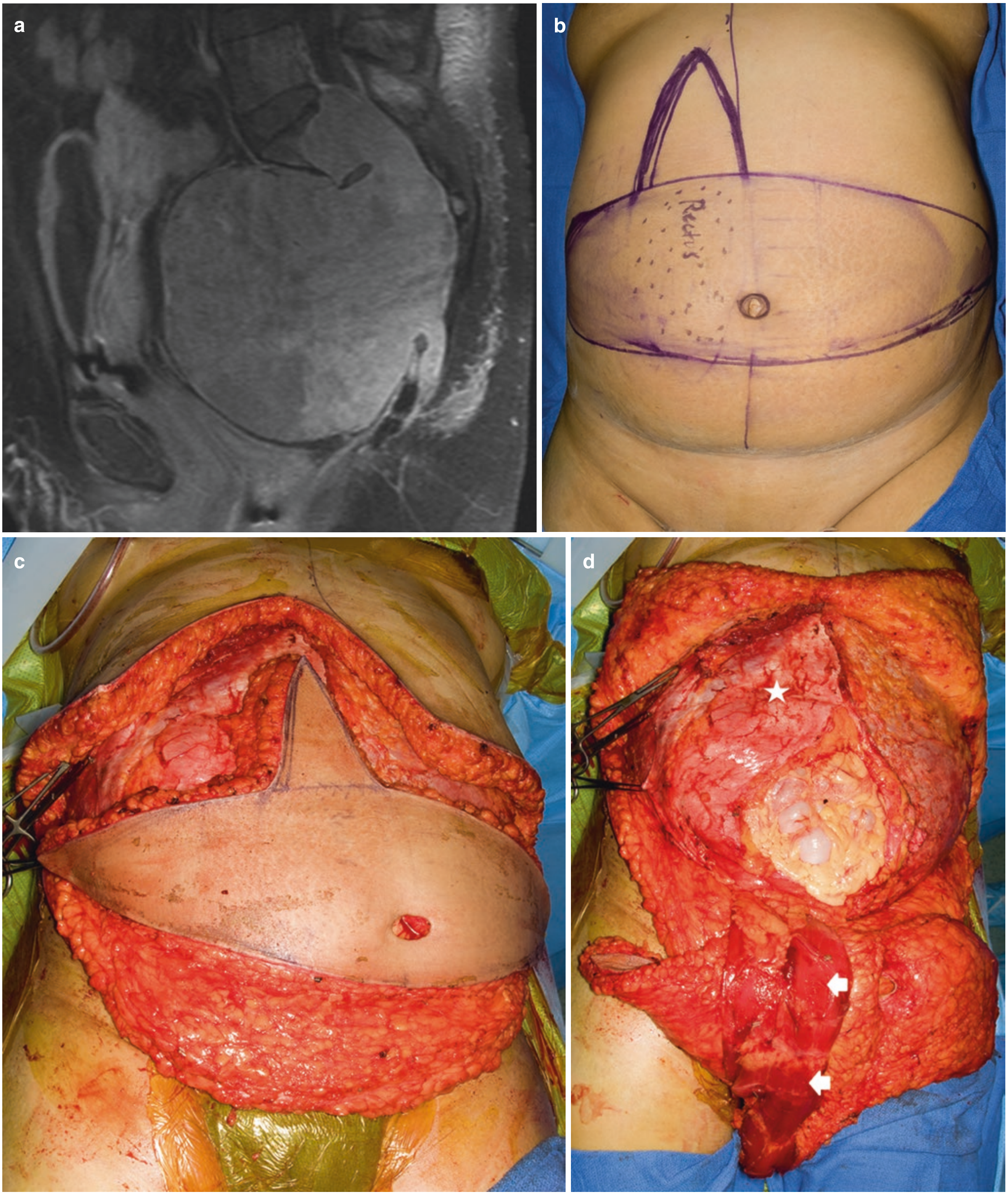

Fig. 30.3 Representative T1 MR imaging of a large sacral chordoma (**a**). Due to the size of the tumor, a combined approach was planned with the use of a TRAM flap to assist with wound closure (**b**). The flap was elevated prior to the extirpative surgery (**c**). The TRAM (**d**—arrows) was elevated off the abdominal wall (star) but left attached to the pubis distally

flap depending on the size of the defect. For patients with very large soft-tissue defects, we prefer to use a TRAM flap in order fill the defect (Fig. 30.3c). The flap is based on the epigastric arteries, both the superior and the deep inferior (DIEA); however, for sacrectomy defects, the pedicle is typically the DIEA. The DIEA arises superior to the inguinal ligament and passes in a layer that is superior to the peritoneum and deep to the transversalis fascia. At the level of the arcuate line, the DIEA enters the rectus sheath and divides into the medial and lateral branches. The flap is elevated off the abdominal wall with both these perforators and left attached the pubic ramus to prevent kinking of the pedicle (Fig. 30.3d). It is then placed in the abdomen with a stich to marker "superior" for orientation on in setting. The flap is then either placed in a plastic bag or wrapped in a moist laparotomy pad and tucked into the pelvis.

During the anterior approach, the viscera are mobilized and the posterior divisions of the internal iliacs, middle and lateral sacral vessels are ligated. An anterior discectomy is then performed at the upper extent of the resection. The involved sacral nerve roots are ligated and the L4/5 roots should be identified and protected. Receptacles for the distal insertion of the fibular grafts are created into the bilateral ilia where the iliopectineal and a plumb line from the center of the lowest involved vertebrae and the center of the hip meet (Fig. 30.4). In cases where this area has been resected, the ischium can be used as a docking site. A silastic sheet is then placed between the vessels and the sacrum in order to protect these structures during the posterior approach. The abdomen is then closed over drains.

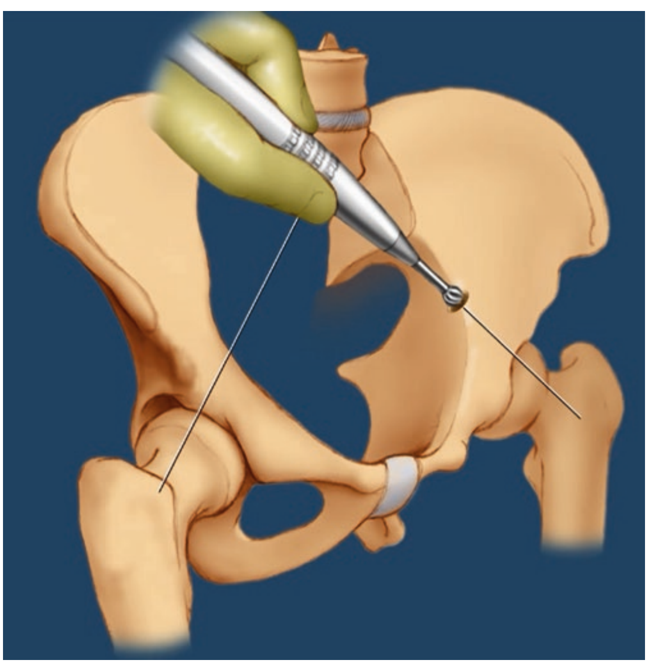

Fig. 30.4 During the anterior exposure of the tumor, receptacles for the strut grafts are made on the iliopectineal line at the crossing point from the center of the planned distal vertebrae and the center of the hip joint

Following this, a posterior approach to the sacrum is performed excising the previous biopsy site and soft-tissue in order to achieve a wide margin posterior typically 48 hours following the anterior approach. Lateral dissection is carried out and the sacrotuberous and sacrospinous ligaments are divided. The piriformis is then divided to expose the sciatic notch, and if it can be preserved, the sciatic nerve should be salvaged. Laminectomies are performed at the appropriate level in accordance with the tumor location in order to achieve a bone margin of 1–2 cm. The dural tube is then ligated and the sacrum and tumor are removed en bloc. Commonly the RAM flap is then temporary inset and the reconstruction is performed following resuscitation in the intensive care unit.

Reconstruction is started by the placement of bilateral pedicle screws in the two most distal remaining vertebrae. These are placed in a bicortical fashion in order to have the best bony purchase. In order to facilitate this and avoid damage to the vessels on the anterior surface of the vertebrae, a finger can be run up on the anterior surface of the vertebrae to ensure proper screw positioning. Bilateral double iliac screws are placed cephalad to the fibula docking sites and a burr is used to create a docking site for the fibulas in the caudal portion of the lowest remaining vertebrae. The fibulas are then fashioned in order to be inserted into the docking sites, creating a "cathedral" appearance (Fig. 30.5a). For cases of previous radiation, we advocate for the use of vascularized free-fibulas over allografts. Once the fibulas are "docked" with the vertebrae, a bilateral two-rod reconstruction with cross-links is performed and used to compress across the docking site (Fig. 30.5b). The RAM flap is then brought through the center of the cathedral pyramid with care to prevent twisting or compression of the pedicle (Fig. 30.6). To prevent posterior herniation and of the viscera and create a barrier between the intra-abdominal contents and the hardware a dermal replacement is used to recreate the posterior abdominal wall; with a caudal space to allow for the RAM pedicle. The RAM flap is then inset and the patient is kept on a specialized bed to allow for flap healing.

30.3.2 Unilateral Reconstruction

In addition to a bilateral reconstruction, in cases where only one SI joint has been resected, a unilateral cathedral technique can be performed (Fig. 30.7). Similar to the bilateral procedures, the technique is used to recreate the load-bearing properties of the SI joint. In order to accomplish this, pedicle

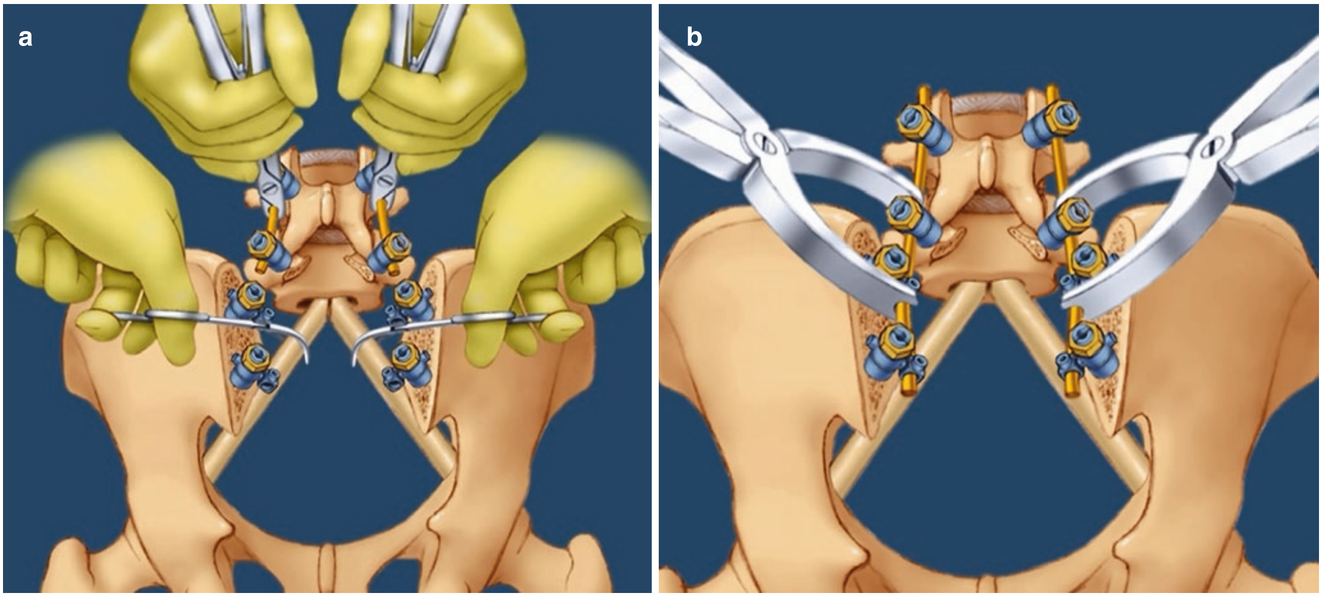

Fig. 30.5 During the posterior procedure, bicortical pedicle screws are placed in the remaining lumbar vertebrae and a docking site is made in the central portion of the most distal vertebrae. The strut grafts are then placed into the ilial docking sites and inserted into the distal vertebrae in a "cathedral" fashion (**a**). Following this, compression is placed across the docking site and the nuts of the pedicle screws are tightened (**b**)

and iliac screw reconstruction is performed and either an allograft or autograft strut is used to supplement the fixation. The spinopelvic fixation is then used to compress across the graft docking sites.

If the unilateral reconstruction is done from a posterior-only approach, our preferred technique is a V-Y gluteal advancement flap for soft-tissue coverage. This flap is based on both the gluteal vessels. The sliding of the flaps avoids the need to dissect the gluteus maximus, preserving some of its function; however, ambulatory patients may notice a decrease in strength. Instead of dissecting the muscle, the muscle and skin are elevated with a triangular skin flap and advanced from lateral to midline, allowing for coverage of the defect (Fig. 30.8).

30.3.3 Amputative Sacrectomy

When the sacrectomy is also associated with a hemipelvectomy, there are few options to restore spinopelvic continuity. In cases where a hemisacrectomy is performed which also involves removal of half of the lower lumbar vertebrae, although spinopelvic continuity is maintained with the remaining pelvis, it is often not enough to support the loads of the body. For these patients, the lumbar pedicle and iliac screw fixation is used to supplement spinopelvic continuity. Although there is no biomechanical data to support its use, depending on the patient size, pedicle screws are placed in two (smaller patients) or three (larger patients) remaining vertebra along with two iliac screws. Two rods are used to connect the most proximal of the iliac screw and the lumbar pedicle screws, while the more caudal iliac screw is connected to the ipsilateral sacral pedicle screw and the contralateral lumbar pedicle screws. An arthrodesis is performed in the remaining lumbar vertebrae and compression is placed through the rod/screw construct (Fig. 30.9).

For cases where a hemipelvectomy and total sacrectomy are performed, we recycle the amputated proximal femur for bone graft as long as there is no tumor located in this region (Fig. 30.10). Two or three lumbar vertebrae have bilateral pedicle screws placed and two iliac screws are placed in the remaining ilium. An intertrochanteric osteotomy is performed in the femoral autograft. The length of the femur should also restore the anatomic distance between the spine and pelvis prior to the resection in order to prevent stretching or kinking of the vessels and nerves. The distal femur is placed in contact with the most caudal vertebrae and the greater trochanter oriented to connect to the ilium with an osteotomy that allows the femur to be placed against the ilium, so it aligns with the long axis of the patient's body. Once this is inset, the pedicle/iliac screw construct is compressed over rods and an arthrodesis is performed in the remaining lumbar vertebrae.

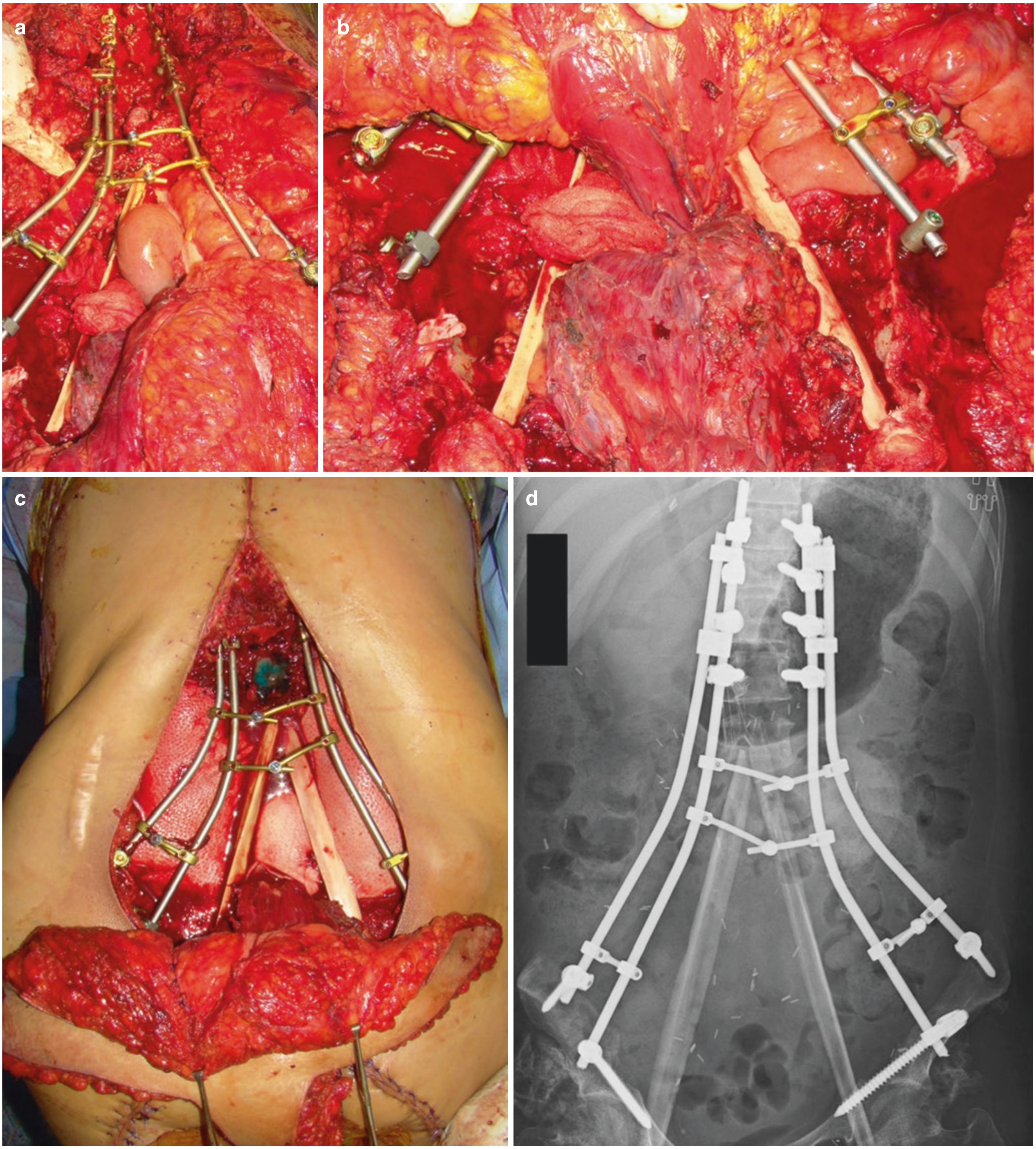

Fig. 30.6 Illustrative case of the cathedral technique showing the dual rod fixation with cross-linking and the underlying strut grafts (**a**). Once the reconstruction is completed, the RAM flap is pulled through the distal portion of the reconstruction with care taken not to kink the pedicle (**b**). A biologic matrix is then placed between the abdominal contents and the hardware in order to reconstruct the posterior abdominal wall (**c**). A slit is created in the distal portion of the matrix in order to allow for the pedicle of the RAM flap. Postoperative AP radiograph showing the completed reconstruction (**d**)

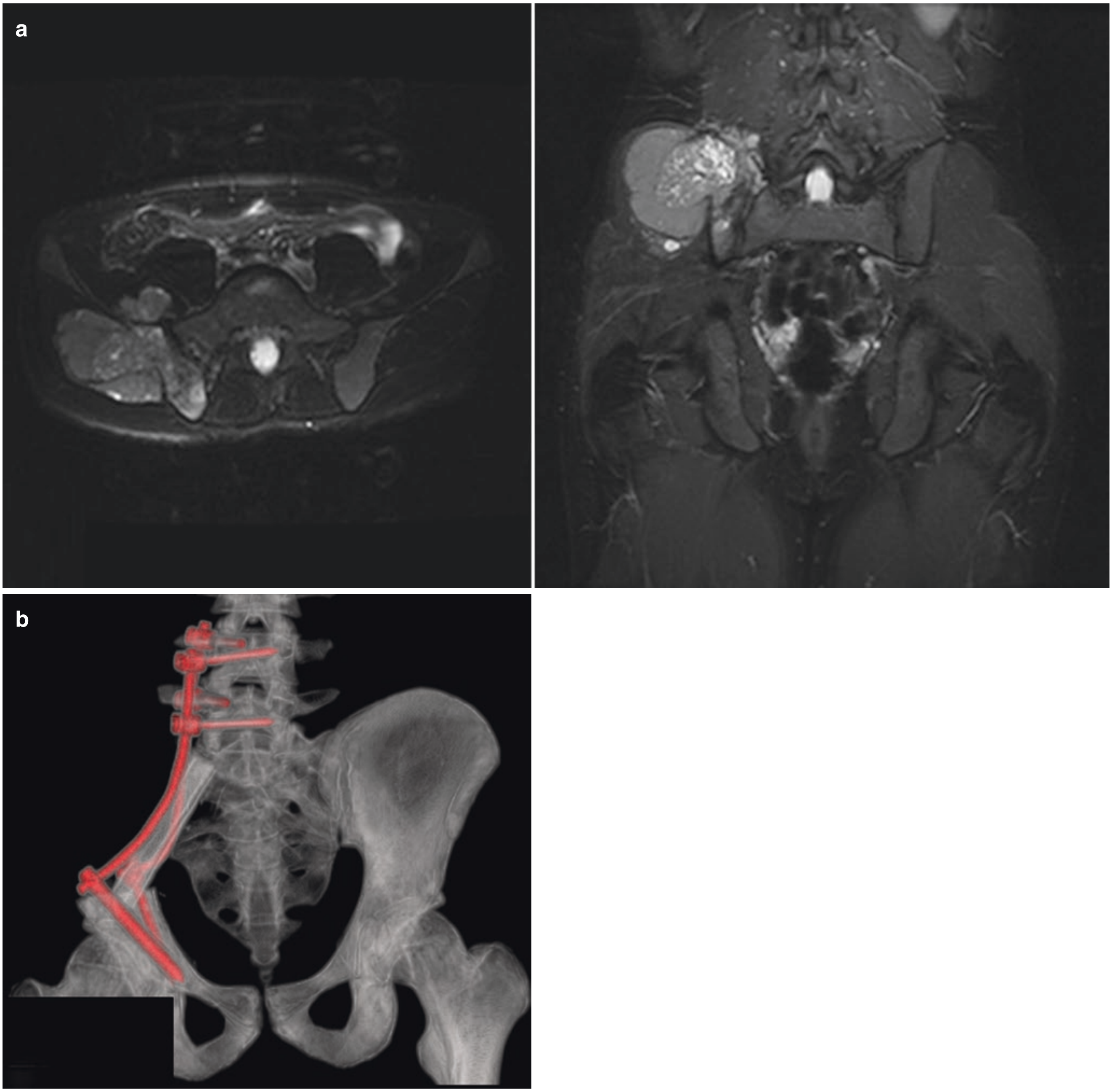

Fig. 30.7 Illustrative case of a patient with sacropelvic Ewing sarcoma involving only a unilateral SI joint (**a**). Following the resection, the patient underwent a unilateral reconstruction using a fibular strut graft as well as pedicle screw fixation in the pelvis as well as lower lumbar vertebrae (**b**)

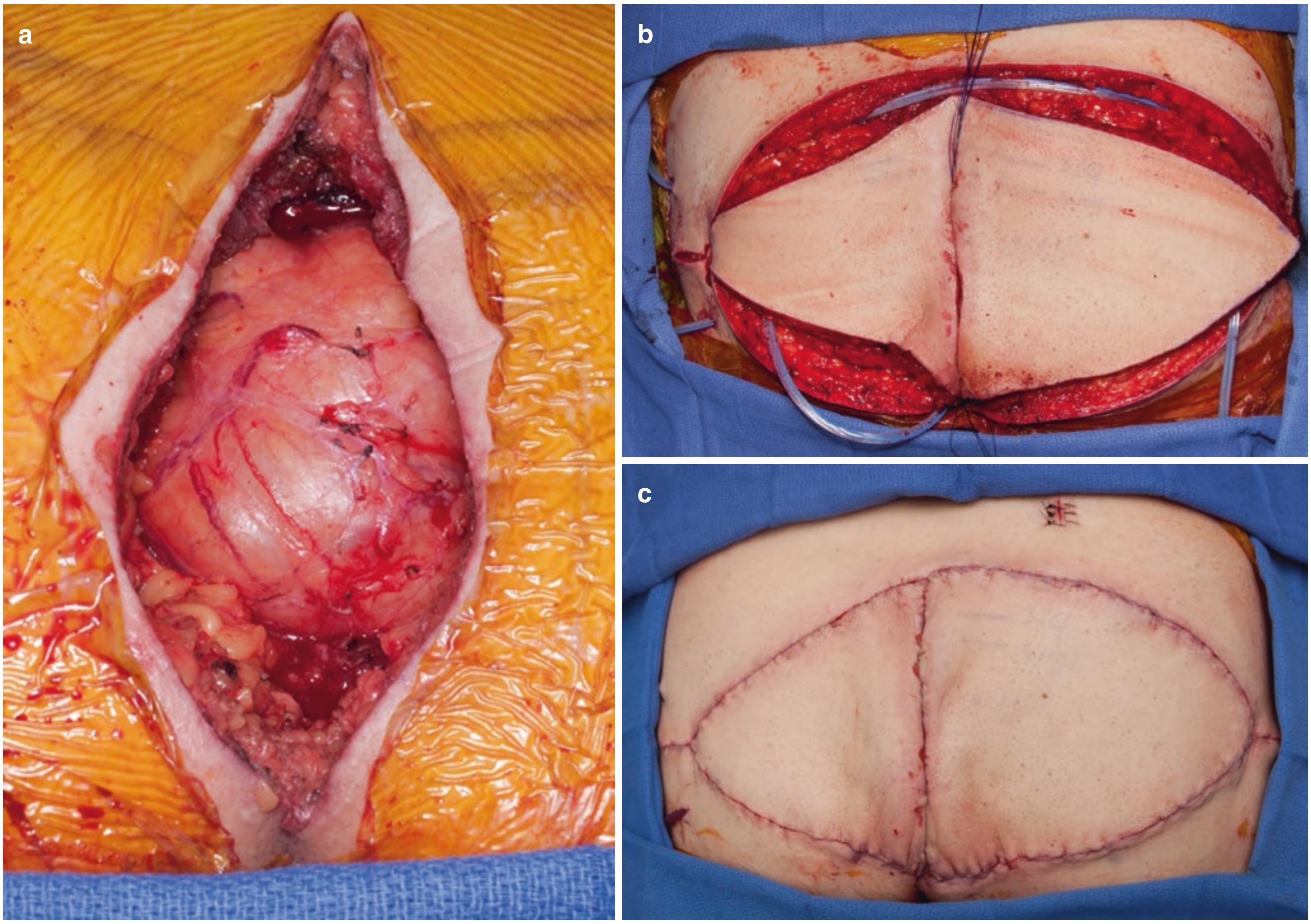

Fig. 30.8 Soft-tissue defect following a distal sacrectomy (**a**). V-Y gluteal flaps were elevated with a triangular skin flap and advanced to midline (**b**) and closed under minimal tension (**c**)

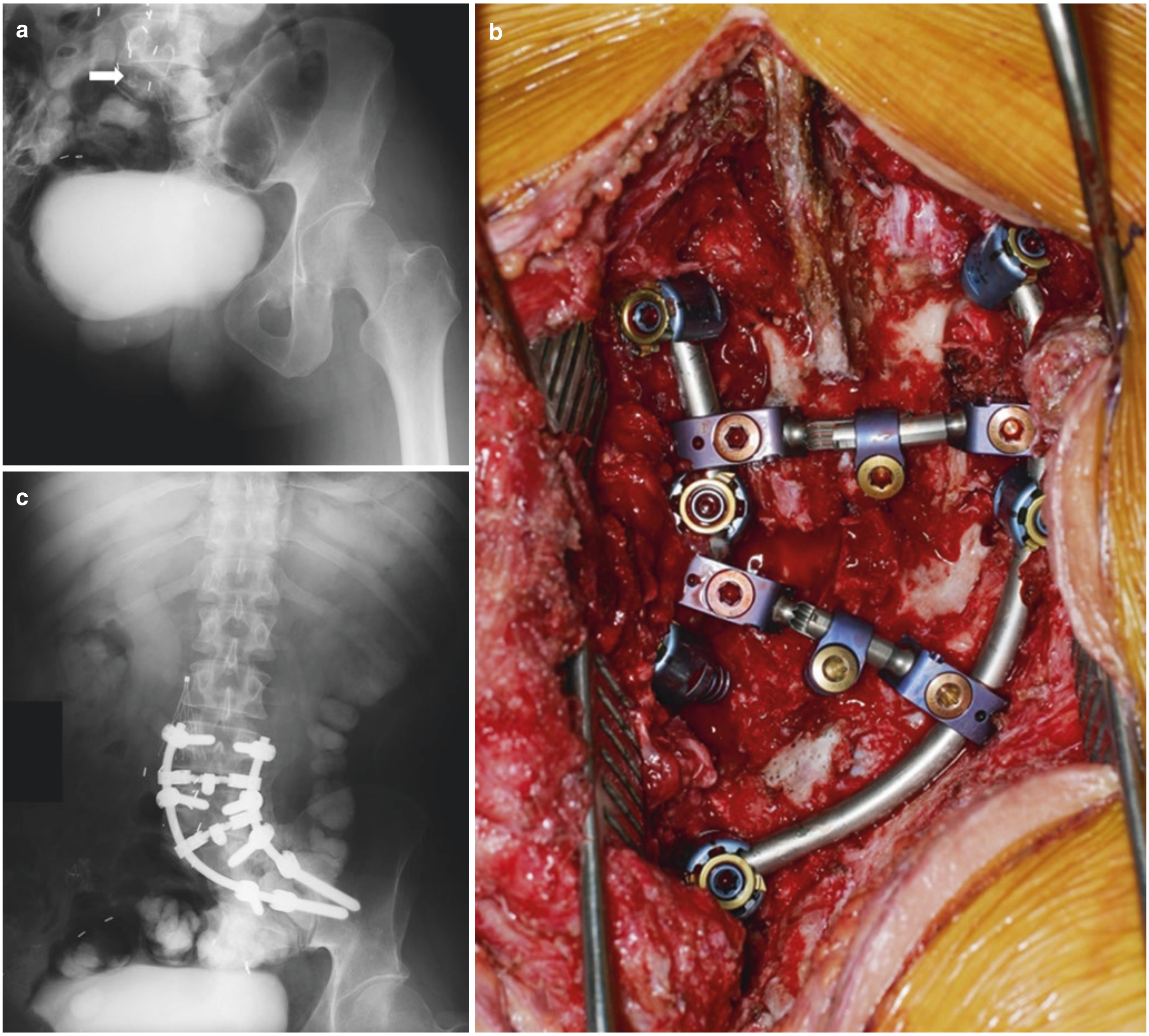

Fig. 30.9 Illustrative case of a patient who had previously undergone a hemipelvectomy and hemisacrectomy (Mayo Type 3). Following the procedure, they developed severe pain related to instability at the remaining lumbar vertebrae (**a**—arrow). They were reconstructed using pedicle screw and cross bar linking of the remaining sacrum and pelvis (**b**). Following the procedure, there was solid fusion of the vertebrae to the pelvis, with a substantial reduction in pain (**c**)

30.4 Conclusion

Sacrectomies are laborious procedures, which require a multidisciplinary team to undertake and reconstruct. The use of the cathedral technique at our institution has resulted in an 89% success rate [6]. In accordance with this, a recent meta-analysis has shown decreased instrumentation failure when the anterior column is supported, opposed to the posterior column-only support [11]. Currently, we advocate for the use of dual iliac screw fixation, double bar constructs with cross-linking. If the patient has a history of radiation to the area, we feel that the use of vascularized bone graft improves the healing potential and construct stability, although it increases the time of the procedure.

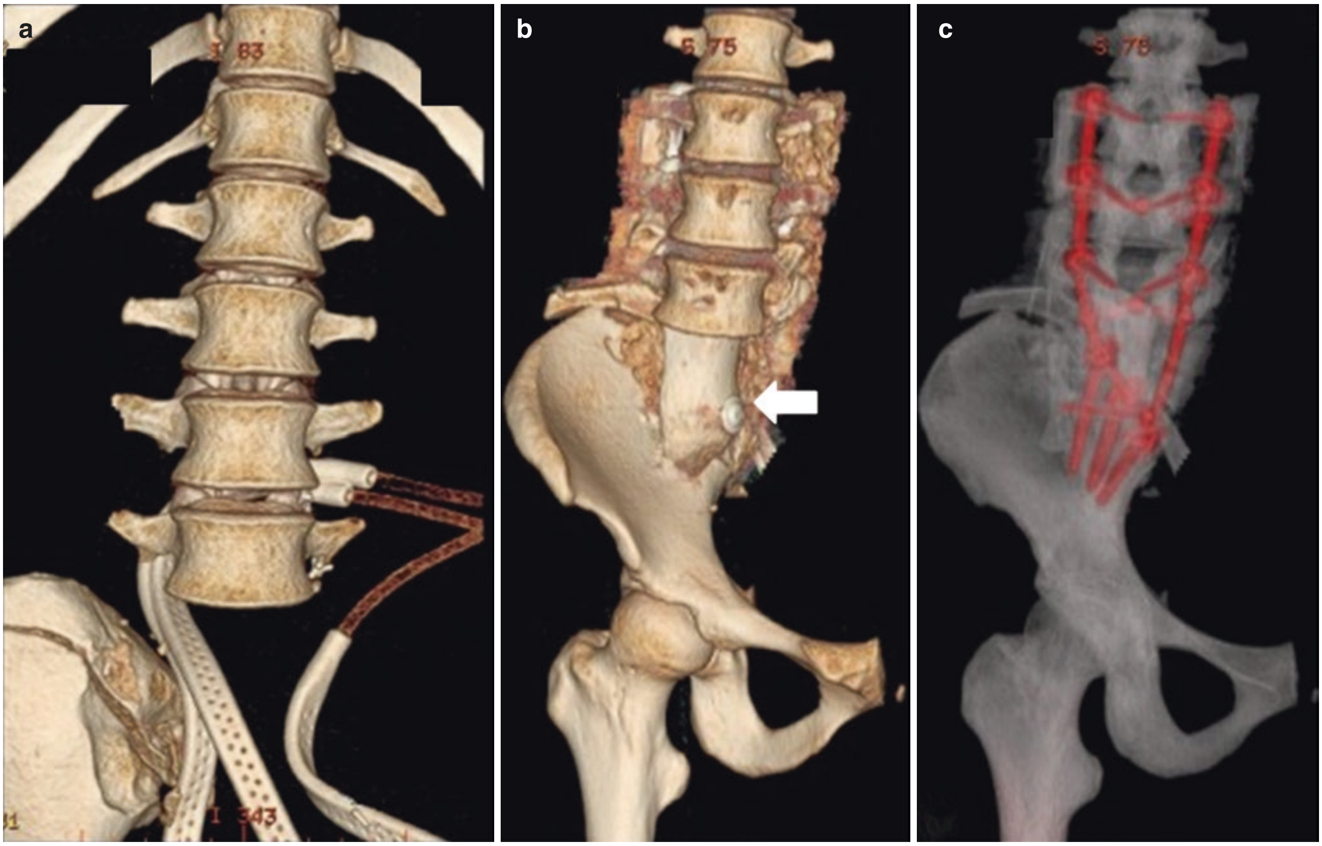

Fig. 30.10 Following a total sacrectomy and hemipelvectomy (Mayo Type 4), the reconstruction is staged and CT scan is obtained to plan for an osteotomy of the amputated proximal femur to provide autogenous bone graft (**a**). The proximal femur (**b**—arrow) is then used to restore the distance between the spine and pelvis and fixed with multiple pedicle screws and rods and compressed (**c**). Eight years following the procedure, they were ambulating with crutches without back pain and a solid arthrodesis

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31 Surgical Strategy for Sacral Giant Tumor

Wei Guo

Giant cell tumor of bone (GCTB) is an intermediate, locally aggressive but rarely metastasizing tumor, which accounts for approximately 4–5% of all primary bone tumors and 20% of benign bone tumors [1–4]. There might be some regional differences in its morbidity. In some Asian countries such as China and Japan, the incidence of GCTB may represent more than 15% of primary bone tumors [3, 4]. It typically affects adults between 20 and 40 years (61%) while in adolescents with an incidence of 10–15% and rarely occurs in juveniles [3, 4]. Approximately 1–4% of GCTB would develop pulmonary metastasis without histologically malignant transformation [5]. It is usually located in long bones but can also develop in unusual locations. 5% or so GCTB could occur in flat bones, mostly in pelvis, and the sacrum is the fourth most common site, accounting for between 1.7 and 8.2% of cases [6–9]. It is difficult to handle sacral giant cell tumor because of its local invasiveness, complex regional anatomy, and hypervascular biological characteristics. Massive blood loss during surgery is a severe complication, which may be life-threatening and make the surgery impossible to complete. Therefore although surgery is the main treatment method of this tumor located in sacrum, its operative procedure has not been well established.

Compared with the GCT of extremities, sacral GCT usually presents with huge volume since it is always asymptomatic until the tumor has grown for a long time. The main symptom may be a sore of the lumbosacral region with 1/3 patients combined with ischioneuralgia. Unlike chordoma of sacrum, it is seldom combined with difficulty in defecation and urination, because it usually occurs at high segmental sacral vertebrae [10]. At the same time, sacral giant cell tumors usually develop in an eccentric position but commonly extend to involve both sides of the midline and into the presacral space. There is usually a thin cortical rim, which the soft tissue can break through. In addition, these tumors have the propensity to cross the sacroilicac joints and intervertebral discs, which indicates that there are more Campanacci radiographic type III tumors than the GCT of extremities [11]. Due to anatomical difficulties such as potential impairment of iliac vessels, lumbosacral nerve roots, ureters, and rectums, surgery may be challenging and risky. Thus optimal treatment for these locations is still controversial. Treatment strategies include radiation, intra-lesional curettage, combined with intra-lesional curettage and radiation, and wide or marginal resection. The obvious advantage of radiation is that there is no incidence of surgical complication. However, the main disadvantage is the high recurrence rate without surgical removal and risk of radiation-induced secondary sarcoma. Intra-lesional curettage could also avoid the injury of nerve roots, bony support of the pelvic ring, and vascular structures; however, there is a substantial probability of recurrence. Total or partial en bloc sacrectomy can enhance local tumor control and overall patient survival, but it could notably increase potential complications and neurologic dysfunction [12, 13]. With decades of experience of operation at this location, we think that it is infeasible to carry out total en bloc sacrectomy of sacral giant cell tumor because of the sacral nerves and the intra-abdominal organs. Thus it is sometimes inevitable to have local recurrence after surgical treatment of sacral giant cell tumor. We are presenting our experience with diagnosis and especially surgical management of 141 cases of giant cell tumors of sacrum from July 2000 to December 2013 in Peking University People's Hospital Musculoskeletal Tumor Center, which accounted for 17.8% of all the primary tumors of sacrum. There were 69 males and 72 females, with an average age of 34.2 (range, 16–61 year).

31.1 The Choice of Surgical Approach for Sacral Giant Cell Tumor

All those patients had routinely taken X ray, CT, and/or MRI to define the local lesion. The imaging of giant cell tumor on MRI usually manifests as heterogeneous signal, which

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indicates intra-lesional hemorrhage or cystic degeneration. The tumor mostly involved the S1-2 vertebra, while sometimes it might also involve the entire sacrum after long-time growth. All medical charts were reviewed, and approximately 1/4 of the cases involved the sacroiliac joint. The tumor was locally aggressive and destructive, and it grew rapidly, destroying bone and spreading into surrounding soft tissues, which lead us not to deal with it as a general benign tumor. In spite of this, the standard treatment for a giant cell tumor nowadays is still aggressive curettage followed by adjuvant local therapy. We had only bare cases located at lower than S3 with tumor en bloc wide resection while most patients received intra-lesional or marginal resection or even combined curettage and resection. For those with remarkable sizes, the only option left was multiple vessels embolization and radiotherapy. We needed to use curettage combined with resection to deal with those located at high segmental sacral vertebrae. Especially for those with neoplastic boundary surpassing lower than S3 (S1-S4, S1-S5), we needed to marginally resect the part of the tumor at S3 or lower than S3 and curette the proximal part of the tumor, with which we could effectively reduce the recurrence with maximum avoidance of damage to the sacral nerves (Fig. 31.1). After these procedures, we then use a high-pressure flushing gun or "water jet cutter" to deal with remnant cavity as well as operative surface to thoroughly clean up neoplastic tissue.

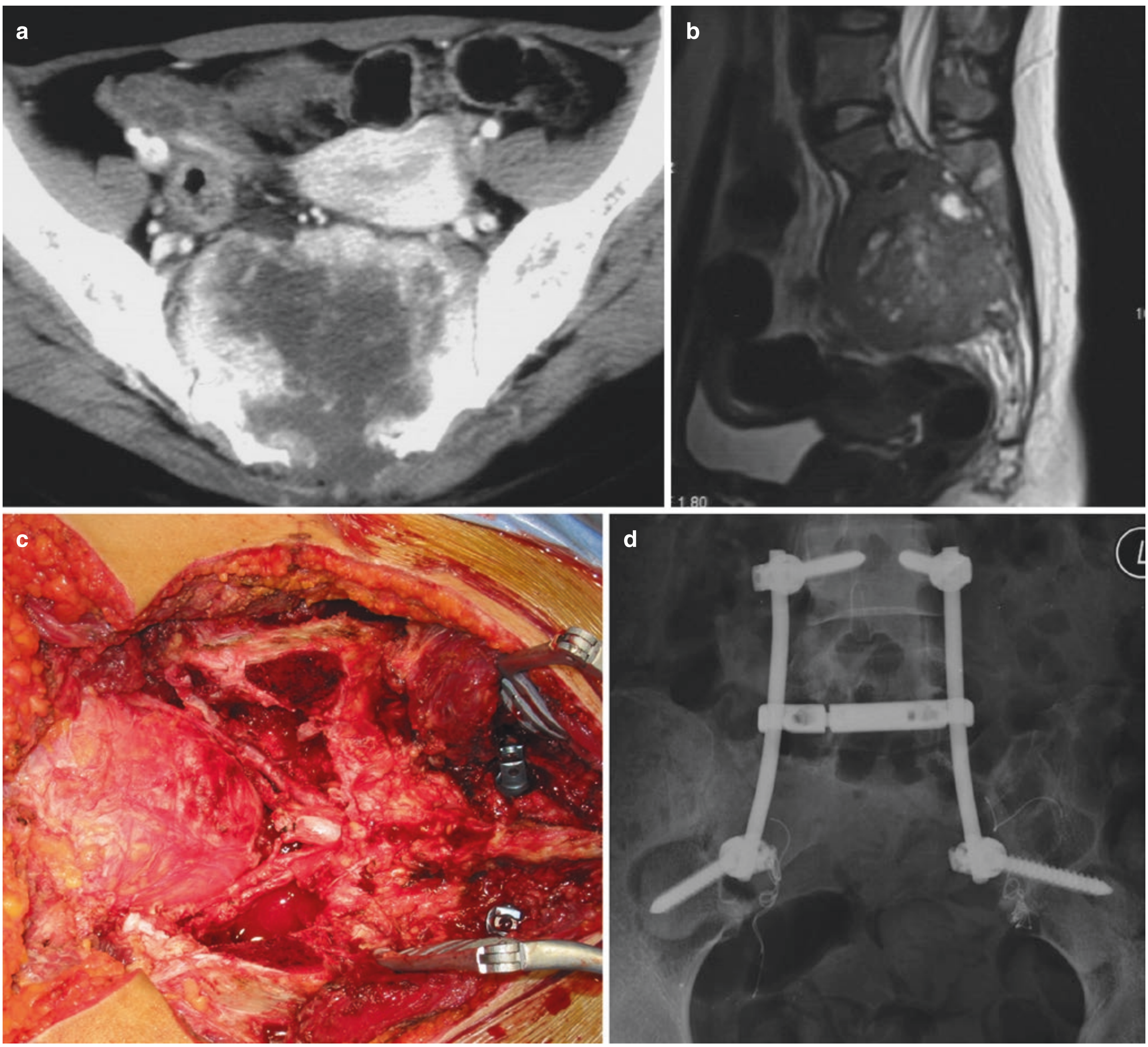

Fig. 31.1 Male, 25 years old, sacral giant cell tumor; (**a**) contrast CT manifested heterogeneous intensified tumor; (**b**) sagittal view of the T2 weighted phase showing that the tumor involves S1-S3; (**c**) Perioperative photograph showed that after marginal resection of the tumor, the bilateral nerve roots of S2 and S3 were intact. The dilatant organ was rectum; (**d**) the X ray after surgery showing the vertebral pedicle screws-rods fixation system for bony reconstruction

It is possible for surgeons to choose a single posterior approach to complete the surgery of giant cell tumor of the sacrum with satisfactory result. Some doctors thought it might be more convenient to use one-stage anterior and posterior combined approach when patients presented with larger tumor, while we thought it might not be necessary. With years of practice, the procedure with posterior-only approach brought us a local recurrence rate of 18.9% in our 130 cases of sacral giant cell tumors, far lower than the incidence of 47% reported in the literature [14], which, in our opinion, relied on wide resection of most part of tumors and effective control of intra-operative hemorrhage. Giant cell tumor is a hypervascular lesion [15]. Taking necessary measures to control blood loss during operation could precisely expose the surgical field, which is helpful for clearing the tumor and avoiding residual lesion. In all our cases, 99 patients had temporary blocking operation of abdominal aorta, of which 10 cases used tapes to temporary block it from anterior approach so as to separate abdominal aorta from retroperitoneal approach and 89 cases used preoperative abdominal aorta balloon to block the bloodstream during surgery (Fig. 31.2). At the same time, we had 36 patients with no blood flow blocking-up. Nevertheless we routinely performed preoperative highly selective arterial embolization of all those sacral giant cell tumors.

Among our cases, the average intraoperative level of blood loss was 2546.3 ml, which was done using an inflatable balloon temporarily blocking the abdominal aorta, while those who did not use balloon had a mean bleeding volume of 4727.8 ml. There was a significant difference between the groups that use abdominal aorta balloon and groups that do not in dependent-samples t test ($P < 0.001$), which was obviously below the blood loss volume of 6900 ml reported by Ozaki [12].

Leggon et al. [14] indicated that the local recurrence rate of giant cell tumor of the pelvis and sacrum seemed higher than any other location. In the study by Sanjay et al. [16], 6 of 19 pelvic tumors developed local recurrence. Due to the complicated anatomical structure and hypervascularity of the tumors, massive blood loss often occurs during surgical treatment procedures, which makes the local management of the tumor even poorer. Previous studies have reported survival to local recurrence after curettage, radiation, and embolization for sacral giant cell tumors ranging from 57 to 80% [6, 7, 14, 17–19]. Except for wide or marginal resection of the tumor, all the other treatments may result in higher rate of recurrence [6, 7, 14, 18]. The risk increases with time, so that the rate might be even higher with a longer follow-up.

The role of irradiation in the treatment of giant cell tumor is still a matter for discussion. It is used as a primary or adjuvant treatment, but is accompanied by a high risk of inducing secondary sarcomas. On reviewing the literature [14, 20], we did not find any statistical difference between radiation combined with intra-lesional curettage for large sacral GCT and any single-therapy method above. Furthermore, radiation combined with curettage was associated with substantial complications, including wound healing problems, infection, and vascular and visceral injuries. There is also no evidence that high dose radiotherapy could apparently lower the recurrence rate [20, 21]. In this series, 2 of 5 patients who received radiotherapy after surgeries had short-term local relapse, which indicated that it did not help local control. It is believed that wide or marginal resection of giant cell tumor located in sacrum could evidently reduce local recurrence; however, it could also increase surgical complications, such as injury to the sacral nerves and pelvic viscera. Cryotherapy as an adjunctive treatment to curettage when the size and stage of the tumor were increased has not been discussed much to have enough number of cases to conduct statistical analysis [22].

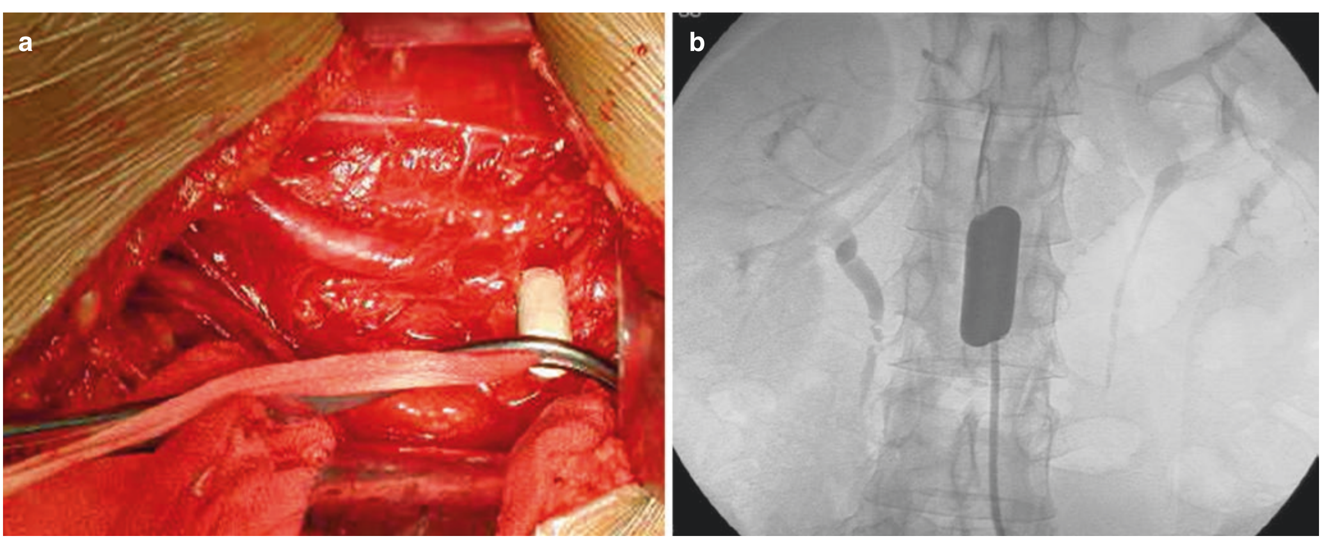

Fig. 31.2 The two methods of adjuvant vascular blockade to control hemorrhage during operation; (**a**) after the dissociation of abdominal aorta, indwell tape around the vascular with approximately 2 cm thoracic drainage catheter to repress and block the blood flow; (**b**) the application of abdominal aorta balloon for intraoperative blood control of sacral tumor. From the picture, we could see the balloon has been inflated

In our opinion, en bloc wide resection should be the best option for surgeons to deal with sacral giant cell tumor, which in actual fact during operation may even reduce so excessive hemorrhage as curettage and reduce local relapse. However sometimes doctors need to weigh the advantage and disadvantage according to the tumor size, location, or even the patients' wishes to make the most appropriate choice.

31.2 Influencing Factors for the Recurrence of Sacral Giant Cell Tumor

For sacral giant cell tumors, the difficulty of surgical procedure lies in the hypervascularity inside the tumors, which causes massive hemorrhage during surgery and high perioperative mortality rate. It is also because of the excessive blood loss during operation that makes the surgical field unclear, which leads to hasty manipulation during neoplastic curettage and incomplete tumorous clearance. Turcotte et al. [20] reported that the local relapse rate of sacral giant cell tumor curettage was as high as 33% while Leggon et al. [14] showed the local recurrence was 47% after reviewing the literature. Conventionally our patients were treated by preoperative selective arterial embolization the day before surgery to prevent excessive intraoperative bleeding. For some huge sacral giant cell tumors, we may even have them repeatedly embolized as many as six times before surgery, which reduced intraoperative hemorrhage obviously.

There were a total of 25 cases (18.9%) with local recurrence in our 141 sacral giant cell tumors. On comparing the two groups with and without temporary vascular blockade, we found that there were 13 recurrences (13.1%) with abdominal aorta temporary balloon blockade and 12 (33.3%) relapses without. By χ^2 test analysis we found that there was an obvious statistical difference ($P = 0.04$). It should be related to more clear exposure to surgical field and more radical elimination of tumor cells. Thus we think more effective control of the blood loss would lead to better control of the local recurrence. According to our clinical data, the average recurrence time after surgery was around 14 months. And different surgeries with different marigins might result in different outcomes. In our experience the recurrences of tumors had most relationship with residual tumors without complete clearance. Reducing intraoperative hemorrhage could help improve the degrees of thoroughness and decrease the recurrence.

There are mainly two ways of low-level vascular blockade to control hemorrhage (Fig. 31.2), of which one way is to ligate affected side internal iliac artery and use tape to temporarily block the abdominal aorta with anterior approach and the other way is to use preoperative angiography to selectively embolize unilateral or bilateral internal iliac arteries as well as tumor blood vessels. As far as we know, the effect of SAE is not satisfactory [10, 14, 16, 22]. When preoperative angiography and selective arterial embolism were in progress, we usually insert an air-free double-lumen balloon catheter into the femoral artery. When the tumor was exposed and the hemorrhage was excessive, during which the edge of tumor was difficult to identify, we gradually filled the balloon catheter with saline. By this way we could achieve the same effect as the temporary blocking of abdominal aorta with anterior approach. However this method could remarkably shorten the surgical time and avoid relevant complications with separating vessels through anterior approach, which improved the safety of the surgery. As a matter of fact, the application of a balloon dilation catheter (BDC) for intraoperative blood control of sacral tumor is an effective and safe way, which has been used widely in the recent years [23].

Besides, the high recurrence rate might also be connected with a high-level vertebral involvement of the tumors, which made surgeons use curettage more to avoid severe damage of bilateral S3 or above S3 sacral nerves. Our results indicated that true recurrence following en bloc excision was at the lower end of recurrence rate. Through the posterior approach, we used laminectomy and curettage with a curette and high-speed burr. The sacral nerve roots were identified and preserved as much as possible. The S1 and S2 nerve roots are large and easy to distinguish and preserve. According to Pietro Ruggieri [24], substantial and well-defined neurologic deficits from injury of the L5-S2 nerve roots were classified as major neurologic deficits. The S3 and S4 nerve roots are smaller and more difficult to identify, the damage of which would lead to pain and hypesthesia in the outer perianal region as well as the penis or labia, which are subtle and are classified as minor neurologic deficits. We usually ignored the minor neurologic deficits but did pay attention not to sacrifice all L5-S2 nerves, which in other words means that we could still keep unilateral high-level sacral nerves to preserve most of the functions of the perineum. Given the recurrence rate of our data, it is acceptable to do this procedure, which combined curettage of high-level sacrum and wide en bloc resection of lower-level sacrum to reach a satisfactory neoplastic margin.

31.3 Drug Therapy of Sacral Giant Tumor

With improved understanding of the pathogenesis of giant cell tumor, drug therapy for GCT has been investigated for decades. The discovery of the RANK/RANKL pathway has recently led to the development of the monoclonal antibody denosumab [25–27]. RANK–RANKL interaction and macrophage colony-stimulating factor (M-CSF) play important roles in osteoclastogenesis. Currently, denosumab has been approved by the United States Food and Drug Administration and in Europe by the European Medicine Agency for GCTB. Since June 2013, denosumab is indicated for the treatment of GCTB that is unresectable [28–30]. The safety and efficacy of denosumab for the treatment of GCTB were demonstrated in two phaseII, open-label studies, with all patients receiving 120 mg dose subcutaneously; $D_{1,8,15}$ for the first 28 days and then once every 4 weeks [31, 32].

This is especially encouraging for some sacral giant cell tumors whose locations are difficult to manage for safe margin during surgery or whose volume is too huge to handle. In our series of 25 giant cell tumors from February to December 2014, 14 cases with extremely huge volume located at sacrum or pelvis were considered as unresectable. With a median follow-up of 6 months (range, 4–12 months), a patient with sacral huge GCT got CR, 5 patients got PR, and 4 patients got SD. No PD had ever been noticed.

The rate of grade III tumors (15/19) of sacrum was higher than in patients with other locations [8, 33]. Diagnosis of tumors of this place may be delayed compared to tumors of the long bones. An external swelling is not visible until late, and radiographs are often misinterpreted. Moreover, the osteolytic lesion of the tumor may be confused with gas-filled intestines or may be covered by shielding of visceral organs, which may explain the high rate of soft tissue extension. As we have mentioned, approximately 1–4% GCTs develop pulmonary metastasis [15], lesions of which often have relatively indolent behavior. Malignant transformation has been described in less than 1% of all GCTs and may be either primary or more commonly secondary progression [32]. Besides, secondary aneurysmatic bone cysts are seen in 10–14% GCTs [6–8, 14, 15].

Other drugs, like antibody toward Epidermal Growth Factor Receptor (EGFR) signaling, is also under investigation [34]. EGFR expression was more frequent in recurrent and metastatic giant cell tumors, suggesting that it may also be a therapeutic target.

There were 3 of 141 sacral giant cell tumors in our series with repeated recurrence and mal-transformation, which was close to the incidence of 1% in the literature [2, 14, 24]. All 3 patients did not undergo radiotherapy, thus we think it may be related to the stimulation of surgical procedures and should still be defined as primary malignancy transformation. There were 5 cases with pulmonary metastasis

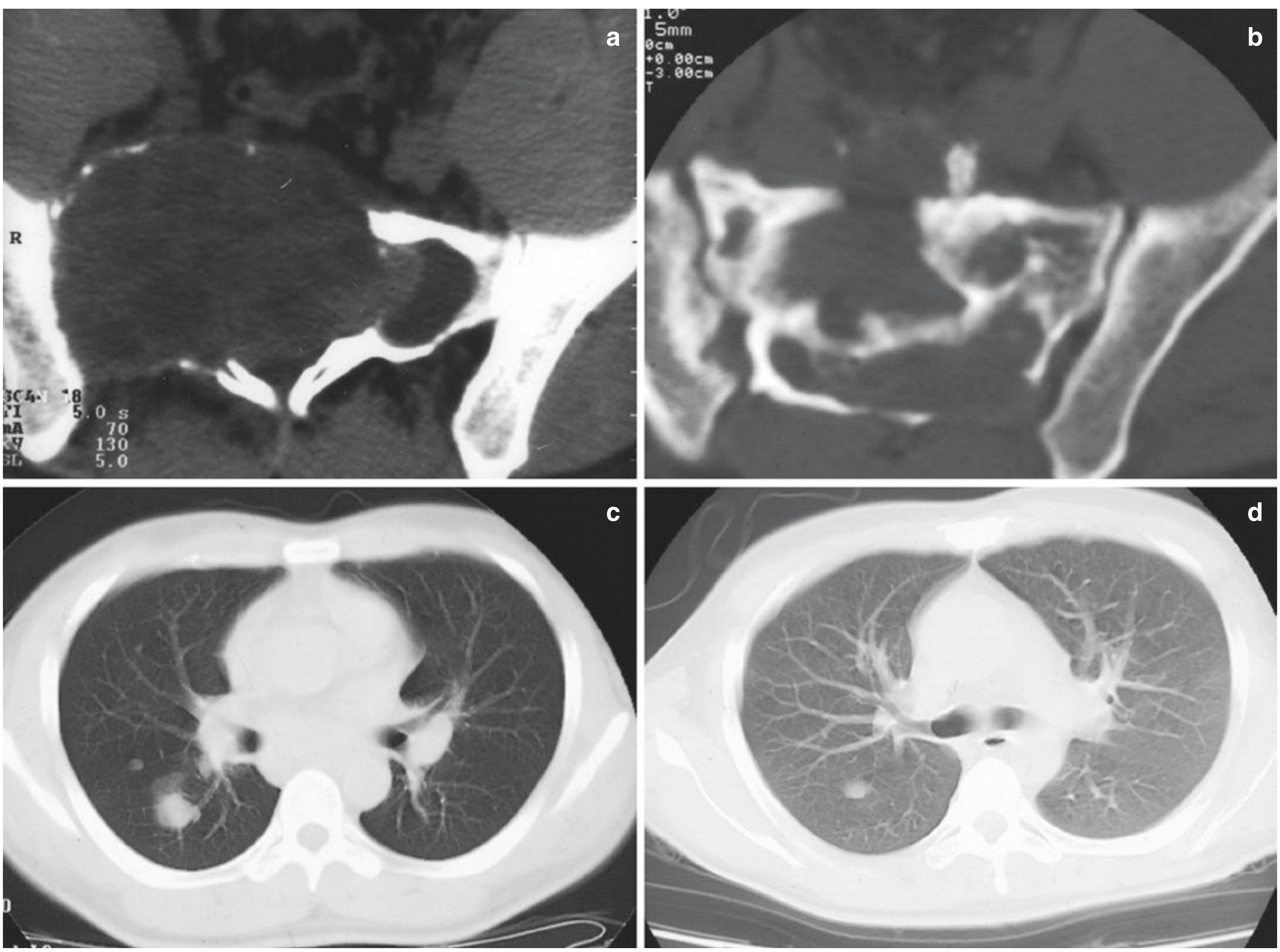

Fig. 31.3 Male, 33 years old, sacral giant cell tumor with pulmonary metastasis; (**a**) the pelvic CT at initial diagnosis showing that the tumor has grown eccentrically and involves sacroiliac joint; (**b**) 2 years after surgery the tumor has relapsed; (**c**) 7 years after the first surgery, pulmonary metastasis has appeared and chemotherapy was intended; (**d**) during routine reexamination 2 years after chemotherapy, the pulmonary lesion is seen to have shrunk

(Fig. 31.3), which was also approximate equal to the 2–9% in literature [11, 15, 20, 24, 35]. Usually in our experience pulmonary metastasis might develop in delayed diagnosis and tremendous bulking cases.

We need to reconstruct the bony stability of the sacro-iliac joint because most sacral giant cell tumors are located at high-level sacral vertebrae. At present the vertebral pedicle screws-rods fixation system is acceptable for an adoptive option [6]. Besides bone grafts could also be available for youngsters. For those involving ilium, bone cements padding should be considered.

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Surgical Strategy for Sacral Neurogenic Tumors

32

Wei Guo

Sacral neurogenic tumors are rare lesions, which include schwannomas, neurofibromas, malignant peripheral nerve sheath tumors (MPNSTs), and neurofibrosarcoma, and are estimated to represent approximately 10% of presacral tumors [1]. Sacral neurogenic tumors, originating from sacral nerves, grow through the neural foramens inward or outward from the sacral canal and are usually shaped like a dumbbell. Tumors growing inward are generally not in large volume due to the defined space of the sacral canal. However the outward growing tumors usually present with extremely large dimensions [2] because of the extensive presacral space, for which the diameters are usually above 10 cm; the largest reported was as big as 28 cm according to the literature [3]. As the incidence of sacral neurogenic tumors is low, it is difficult for a single department to have adequate number of cases for surgeons to be familiar with. Thus in this chapter, we focus on the surgical approach and procedures for this special subtype of sarcoma.

Benign sacral neurogenic tumors include schwannomas and neurofibromas, of which the latter has a higher incidence than the former. While malignant tumors include malignant schwannomas(also referred to as malignant peripheral nerve sheath tumors) and neurofibrosarcoma [1]. It is rare to see neurogenic tumors located at sacrum, which comprises about 7% of the sacral tumors [2]. Much of the literature related to neurogenic tumors were sporadic case reports [2, 4]. Presacral neurogenic tumors are frequently found to be a large mass due to delayed manifestations. Patients are asymptomatic until the mass compresses the adjacent organs and cause symptoms. Some patients have such huge mass that could even be palpated through the anterior abdominal wall. Incidental recognition of paravertebral or presacral tumors has multiplied due to the increased use of computed tomography scan or magnetic resonance to investigate chronic back pain or radiculopathy. This disease is easily found in females at their second and fifth decades of their lives [4]. Benign neurogenic tumor grows slowly with an integrated and thick capsule. The initial treatment of these tumors by complete resection gives them the best chance for an overall and disease-free survival. Giant sacral neurogenic tumors are characterized by an indolent growth pattern and nonspecific symptoms [3]. From plain film of sacrum, a distensible sacral neural foramen could be noticed; however, this characteristic could be blurred in malignant lesions. Most benign neurogenic tumors manifest as homogeneous high signals at T2-weighed phase of MRI, most of which are with cystic degeneration, while in malignant cases the manifestation turns out to be heterogeneous on MRI. Thus heterogeneity of the signals on MRI may indicate malignancy.

Previous studies on the tumors are limited and consist of a relatively small sample size [3]. We retrospectively investigated 128 cases who all had sacral neurogenic tumors removed at the same hospital in order to determine the optimal surgical approach for a specific tumor growth pattern. Between July 2000 and July 2014, 128 cases with sacral neurogenic tumors received surgical treatment at the Musculoskeletal Tumor Center of Peking University People's Hospital, of which there were 71 neurofibromas, 43 schwannomas, and 14 malignant peripheral nerve sheath tumors (MPNSTs). In actual fact, those sacral neurogenic tumors do not belong to primary sacral tumors. Most of our patients were asymptomatic while only less than 20% patients were diagnosed by imaging examination for sciatica, perineal numbness, constipation, or urinary abnormality. A small number of patients went to hospital for an abdominal protuberance and palpable painless mass. And some female patients even discovered sacral tumor because of dystocia. The typical radiographic manifestation of sacral neurogenic tumors was an enlarged sacral canal, outward growth of intrasacral tumor through intervertebral foramen, and a giant presacral soft tissue mass. MRI could detect most homogeneous signals of benign neu-

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rogenic tumors. However, some of those tumors could also present with cystic degeneration within the lesions. We usually could observe low signal on the T1-weighted phase and high signal on the T2-weighed phase images. The average tumor diameter was 12 cm, within which the longest one could reach as long as 31 cm. This tumor grew upward and reached L3/4 level. Because of the obvious characteristics of benign neurogenic tumors, most of them could be diagnosed before operation only by radiographic materials without biopsy. Nevertheless malignant neurogenic tumors were not so distinctive, thus a biopsy before surgery was necessary.

32.1 Classification System of Sacral Tumors

Based on the classification system proposed by Kim DH, et al. [1], we developed a revised classification system for sacral neurogenic tumors. The tumors are classified into four types with respect to tumor growth patterns. Type I tumors are confined to the intrasacral canal and manifest as an enlarged and swollen sacral canal (Fig. 32.1a). Type II tumors extend through the intervertebral foramens of sacrum into the presacral space, forming giant presacral mass (Fig. 32.1b). Type III tumors grow toward both the ventral and dorsal sacral

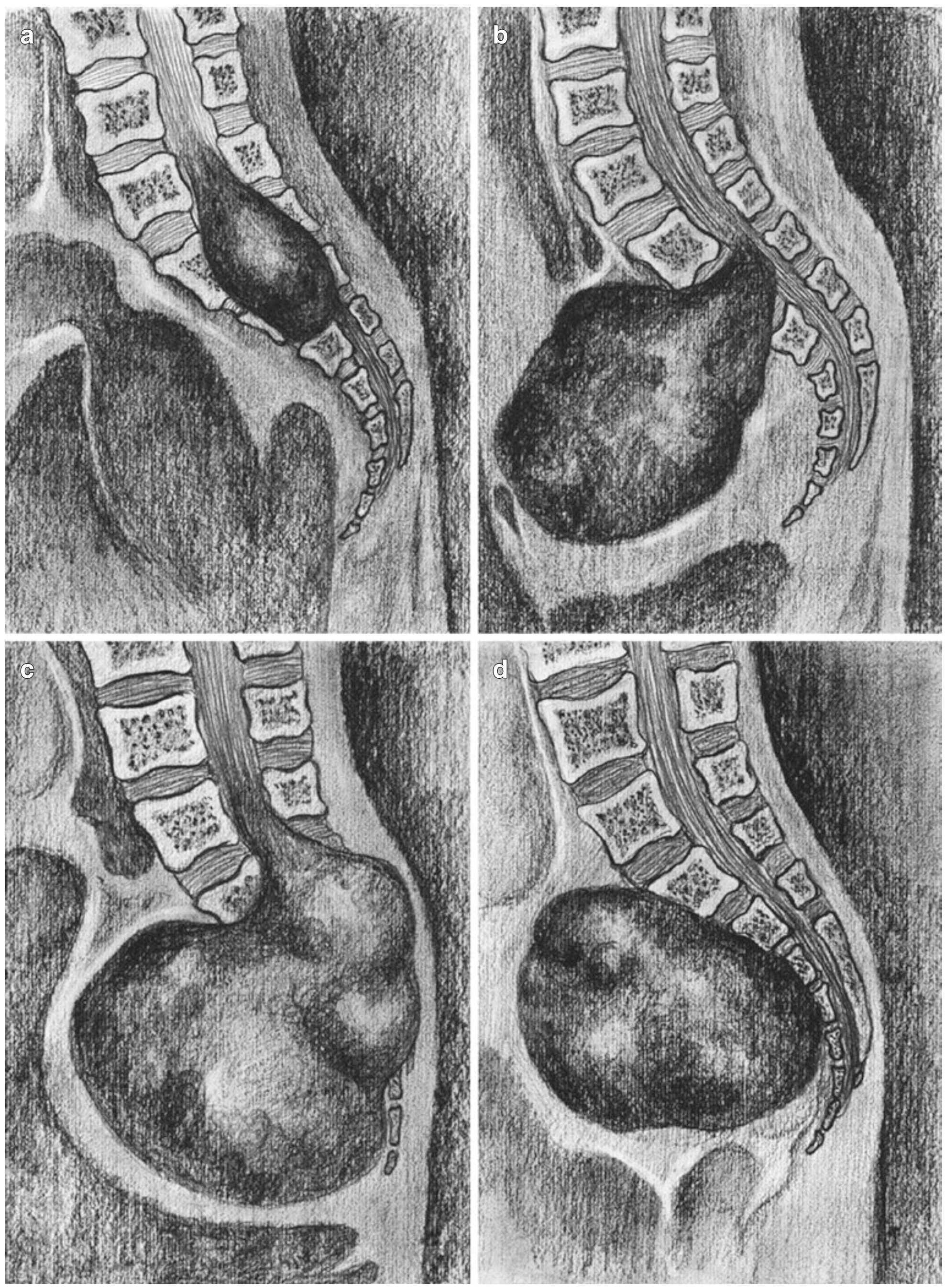

Fig. 32.1 According to its growth pattern, the neurogenic tumors arising from the sacrum can be classified into four types. Type I, the tumor growth is confined to the sacral canal along with the enlargement of the sacral canal (**a**); Type II, tumor grows forward out of sacral neural foramens, with the formation of a huge presacral lump (**b**); Type III, the tumor spreads both anteriorly and posteriorly with formation of lumps anterior and posterior to the sacrum (**c**); Type IV, the tumor growth is confined to the presacral space, with no tumor present in the sacral canal (**d**)

area, forming a mass that envelopes the sacrum (Fig. 32.1c). Type IV tumors are confined to the presacral space with no tumor observed within sacral canal (Fig. 32.1d). In our series, type II and III accounted for the majority [5].

32.2 The Characteristics of Tumor and Surgical Approach

Because of the benign property of sacral neurofibroma and schwannoma, as well as the tremendous volume of those tumors, it is difficult to do en bloc resection. Thus we usually recommend piecemeal resection. The blood supply is usually not so abundant and the tumor is often encapsulated with a relative thick coating. In our experience, the capsule itself is usually hypervascular. If we do the resection outside the capsule, the possibility of damage to the branches of the iliac vessels and injury to pelvic visceral organs such as rectum, uterus, and ureter might arise. Thus we suggest doing intra-capsule piecemeal resection. After getting rid of tumors, we can then separate the outside capsule by lifting and removing it as much as possible. We found only 9% relapse in our more than 100 cases, for which we used intra-capsule piecemeal resection in the majority of cases. For the type I cases and for some type II and III cases, in which tumors grew forward but were lower than S1 level, surgical resection of the tumors only required a simple posterior approach. For those type IV tumors involving high-level sacrum, we usually needed a combined anterior–posterior approach to complete the removal of presacral tumors. For those located at high-level sacrum (higher than the S1 level), with large volume (diameter longer than 10 cm), a combined anterior–posterior approach was required, by which the intrasacral tumors were resected and the intervertebral foramina were enlarged from the posterior side, while the major part of each tumor was resected from the anterior side (Fig. 32.2).

After the resection of the major part of the tumors through anterior approach, more attention should be paid to hemostasis. Because of the deficiency of the compression of presacral fascia, the blood exudation from lesional cavity could get into retroperitoneal space, which becomes difficult to control. The local wound surface often needed compression hemostasis for more than half an hour. The presacral space was entered via resection of the sacrotuberous ligament and coccyx, and the rectum was separated from the tumor. Gauze was packed into the space between the rectum and sacrum, which pushed the rectum forward and upward until the level of S1. From the lateral side, a part of the ilium was removed near the external margin of the tumor outside the sacroiliac joint. From the posterior approach, the vertebral lamina was removed, the sacral canal was revealed, and the sacral nerves were separated carefully. The neural foramina were enlarged by the tumor, and the tumor was resected together with the whole piece of partial sacra [6]. In order to reduce the intraoperative blood loss, a balloon dilation catheter (BDC) for temporarily occluding the abdominal aorta might be implanted for some huge tumors. Ligating ipsilateral internal iliac arteries and using tape to temporary block abdominal aorta could be employed by anterior approach or combined approach while a balloon dilation catheter (BDC) for temporary occluding the abdominal aorta could be used for posterior approach only.

For benign neurogenic tumors, marginal resection was used to the greatest extent possible. In general, those who had marginal resection could achieve a satisfactory prognosis and a low local recurrence rate. For those malignant tumors that had either an intact membrane or no membrane at all with a wide range of intraosseous damage, and which were mostly positioned at high-level sacrum, it was sometimes difficult to

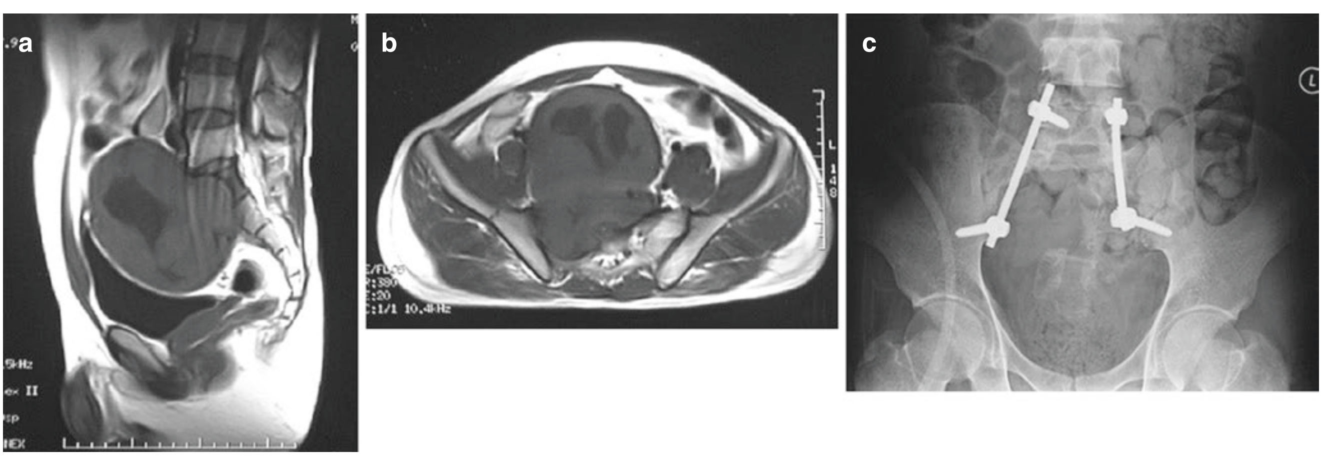

Fig. 32.2 The MRI showed a huge schwannoma (type II) located anterior to sacrum and involving the S1 vertebral body, with the upper limit reaching the upper margin of L5 (**a** and **b**). The radiograph plain (**c**) film shows the stability reconstruction of the lumbar-sacral region after resection

remove tumors thoroughly. Therefore these patients had a high rate of local recurrence. We recommended an extensive resection of tumors, which included the sacrifice of sacral nerves in order to reduce recurrence rate and improve the survival. Preoperative and postoperative chemotherapy can be administered for patients with malignant neurogenic tumors and radiotherapy can be added after surgery for adjuvant therapy. However we usually discovered malignant neurogenic tumors not so sensitive to systemic therapy. Thorough surgical resection appeared to be the best and main treatment method.

32.3 Nerve-Preservation and Stability Reconstruction After the Resection of Neurogenic Tumors of Sacrum

Sacral schwannomas and neurofibromas were both benign neurogenic tumors; therefore, sacral nerves should be preserved as much as possible. In most cases only the nerve roots of the involved side or the nerve roots lower than S4 would be resected. However, many sacral nerves should be sacrificed because of the blurry visual field during operation due to the tremendous size of tumor or excessive intraoperative hemorrhage.

In the past, the reconstruction of lumbar-sacral region was not performed on patients with total or subtotal resection of sacrum. As a result, patients needed longer bed rest time after surgery. More recently, due to the development of internal fixation devices for the spinal column, many physicians perform internal fixation operation after total or subtotal sacrectomy to reconstruct the stability of the lumbar-sacral region. This allows patients to have early-stage activities out of bed without delayed nerve-root symptoms caused by instability of the spinal column. However there still are many complications in internal fixation operations in the sacrum. Because large cavities exist after resection of the sacrum and there is only one fat layer covering the back without muscle, it is easy for effusion to appear locally, and sometimes infection occurs. If infection occurs locally, the internal fixation devices must be removed. Thus at present we think if the condition is permissible, internal fixation operations should be performed on patients with the resection of a single or bilateral iliosacral articulation so as to reconstruct the stability of the lumbar-sacral region. It is not necessary to reconstruct the sacrum on patients with S1 reservation or on patients with complete iliosacral articulation retention.

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33 Preoperative Embolization and Aortic Balloon

Xiaodong Tang and Wei Guo

33.1 Preoperative Preparation

The operation time for patients with sacral malignant tumors is long, often 4–6 h or more. The hemorrhage amount of this operation is usually large, and many patients need massive transfusion. The preoperative assessment of a patient's general condition, including the presence of metabolic disease, cardiovascular disease, and respiratory disease, is important. If the general condition of a patient before operation suggests inadequate tolerance to surgery and a high risk for it, other therapies such as radiotherapy are suggested first. Then, when the patient's general condition improves, surgery is considered. Inadequate preoperative preparations often result in elevated risk during and after surgery, even death. Autologous blood transfusion can be effectively used in rare cases, whereas large amounts of blood should be kept ready before surgery for intraoperative and postoperative blood transfusion in most cases. When the estimated blood loss exceeds 2000 mL, fresh frozen plasma(FFP) should be prepared to avoid coagulation dysfunction.

The risk factors for early infection are bacterial and fungal infections of the intestinal system and the urinary system, which must be cured before surgery. The most important part of the preoperative preparation is the preparation of the bowel, with the use of enema and laxative. If the preoperative bowel preparation is insufficient, the intestine may be damaged leading to deep wound infection. We usually use enema and laxative for bowel preparation 2 days before surgery. The patient starts liquid diet, and 24 h before surgery, the patient starts to take a laxative. In the morning of the surgery day, enema is performed on the patient. If bowel surgery is estimated to be performed during the whole operation, preoperative bowel preparation should be strict. Few days before surgery, intravenous nutrition support can be given, so that a clean intestine and the patient's general condition can be maintained. Giant retroperitoneal tumors often involve the ureter, colon, rectum, and other retroperitoneal organs. Mostly in such cases, urinary diversion surgery, nephrectomy, or colon resection surgery are needed first. If the tumor involves the ureter and separating the ureter during surgery becomes difficult, ureteral catheterization should be performed before surgery to avoid ureter injury during tumor separation. The amount of blood loss should be estimated and complications should be prevented considering the patient's individual condition. If colostomy or bladder shunt operation is to be performed during surgery, preoperative discussion with the general surgeons and urinary surgeons must be done to create a collaborative surgical strategy. Although not all cases require such operation, team work is a key factor to ensure successful removal of sacral tumors.

33.2 Evaluation of Large Amount of Blood Loss During Sacral Tumor Surgery

The sacrum can be affected by both primary and metastatic tumors. The most common primary sarcral tumors include neurofibroma, giant cell tumor, and chordoma [1]. Other benign or aggressive lesions, such as aneurysmal bone cysts and osteoblastomas, and primary malignancies, such as osteosarcomas, chondrosarcomas, and Ewing's sarcomas, also occur frequently in the sacrum [2].

Although chemotherapy and radiotherapy have been widely used, surgical resection is still a major treatment method for most primary sacral tumors and some metastatic diseases that resist radiation. En bloc resection with adequate margins is the only effective method to achieve long-term disease control or cure [3]. Surgical strategies are as follows. Routinely, marginal resection or intralesional curettage for primary benign and metastatic sacral tumors is performed through a posterior approach. Combined anterior and posterior approaches are needed for some neurofibromas and schwannomas with a large presacral mass. For primary malignant tumors caudad to the disc space between S2 and

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S3, the posterior approach is sufficient for en bloc subtotal sacrectomy. When tumors with limited volume involve S2 and above, the single posterior approach is still an effective choice. Combined anterior and posterior approaches are usually carried out only in patients with large soft tissue mass, which involves high levels of sacrum. Through an anterior approach, the visceral and vascular structures are dissected to prepare the ventral margin of the tumor. The distal portion of the abdominal aorta can be temporarily occluded by a nylon tape to reduce blood loss. Then, the tumor is removed from the posterior approach with a wide or marginal margin, and a spinal screw–rod system can be applied for sacral iliac joint reconstruction if the spinopelvic stability is disrupted. A balloon dilation catheter (BDC) is used to occlude the abdominal aorta in some patients undergoing the posterior approach to control hemorrhaging during surgery. Because of the complex anatomy of the sacral region and the typically large tumor size at presentation, aggressive resections are technically difficult. Total sacrectomy usually has complications including large volume of blood loss, cerebrospinal fluid leakage, superfacial wound problems, deep infections, neurologic deficits, pelvic instability, and internal fixation failure. Among these complications, intraoperative and postoperative extensive hemorrhage is a major concern and may threaten the life of the patient and jeopardize the outcome of surgery.

Blood volume loss during sacral tumor resections varies from case to case. We have carried out a retrospective study [4] that included 173 patients with sacral tumors, and the risk factors for severe hemorrhage were identified. In this study, the total blood volume loss of the surgery consisted of the estimated intraoperative blood loss and the drainage volume on the first day after surgery. The intraoperative blood loss volume was estimated by surgeons and anesthesiologists. It included the exact volume of suctioned blood and the estimated volume absorbed by gauze and dressings. The normal saline volume of lavage was subtracted from the suction volume. A volume greater than 3000 mL was defined as a large amount of blood loss. Obviously, a wide variety of risk factors influence perioperative blood volume loss. Age, gender, tumor blood supply, tumor location, recurrent tumor, preoperative radiation, tumor volume, aortic occlusion, surgical approach, type of resection, reconstruction, and operative time should all be considered.

In the univariate analysis, gender, tumor blood supply, tumor location, tumor volume, aortic occlusion, surgical approach, reconstruction, and operative time have an effect on blood loss. Combined surgical approaches, tumor volume greater than 200 cm^3, abundant tumor blood supply, reconstruction of the sacral iliac joint, operative time longer than 3 h, and male gender are associated with a large amount of blood loss. High-level tumors that involve the S2 body and above are more likely to have a large amount of blood loss than low-level tumors. Large amount of blood loss frequently occurs in patients receiving temporary aortic occlusion. Patients older than 40 years, recurrent tumors, and radiation treatment before operation have no effect on the amount of blood loss. No difference in blood loss exists between patients with an en bloc resection or piecemeal curettage. After multivariate analysis, we found that blood volume loss during sacral tumor resection is influenced independently by the location of the tumor, tumor volume, and tumor blood supply. Sacral tumors that invade cephalad to the S2–S3 disc space, with a volume greater than 200 cm^3, and with abundant blood supply tend to have a large amount of blood loss during operation. Temporary occulusion of abdominal aorta during surgery or other blood vessel control interventions should be considered for such patients. The combined anterior and posterior surgical approach does not independently predict blood loss. However, a combined approach requires complex surgical manipulation in the presacral area and more operative time than a posterior approach alone, both of which can lead to a large amount of blood loss.

Location of the tumor is associated with the greatest independent risk for a large amount of blood loss. The location of the sacral tumor is determined by the cephalad edge of the lesion. Sacral tumors involving the S2 body or cephalad to the S2 body are defined as high-level tumors, whereas tumors caudad to the disc space between S2 and S3 are low-level tumors. Because the upper part of sacrum is adjacent to great vessels, the tumors in this location always require a combined anterior and posterior surgical approach and a careful dissection. The S1 and S2 nerve roots in this area that have to be sacrificed are combined with the venous network that drains the epidural plexus. On the contrary, tumors situated on the lower part of sacrum can be removed easily using a single posterior approach without neurologic damage. Another reason for considerable greater blood loss in high-level sacral amputations is the exposure of a large area full of blood supply. Internal fixation for an osseous defect reconstruction, which is needed only after a high-level sacral tumor resection [5], also may result in increased bleeding, and although it is associated with a large amount of blood loss, it does not independently predict blood loss.

Large tumor volume is another important independent risk factor associated with great blood loss. Sacral tumors such as, neurofibroma and chordoma, are usually difficult to diagnose at an very early stage. The tumors may be far advanced at the time of presentation with a large soft tissue mass. Usually, the size of sacral tumors is often much greater in the sacrum than in other sites. The large sacral tumors always involve the upper portion of the sacrum and have a presacral mass adhered to or invading the pelvic visceral organs, both of which make surgical resection difficult and lead to increased bleeding.

Fierce bleeding easily occurs during the resection of tumor with excessive blood supply [6, 7]. Generally, malignant tumors are more likely to induce ingrowth of abnormal capillary vessels and arterioles than benign tumors. The notorious sacral tumors with an excessive blood supply include all the primary high-grade malignant tumors, multiple myelomas and metastatic hepatocarcinomas, renal carcinomas, thyroid carcinomas, and two benign tumors (giant cell tumors and aneurysmal bone cysts). The tumors with a reduced blood supply include other primary benign tumors and tumor-like lesions; they also include low-grade malignant and other metastatic tumors. In our study, the blood volume loss of giant cell tumors was similar to that of malignant tumors. Therefore, BDC or surgical cross-clamping of the aorta is now a standard procedure for sacral giant cell tumor resection at our institute.

33.3 Vascular Occlusion

The anatomic structure around the sacrum is complex, with important or irregular blood vessels densely covered. Thus, the blood supply is extremely rich. Surgeons usually face the problem of massive hemorrhage directly when performing sacral tumor resection and reconstruction surgery. Massive hemorrhage can affect the performance of the surgical program and directly affect patients' internal environment. In such a case, complications and adverse effects are expected.

33.3.1 Preoperative Digital Subtraction Angiography (DSA) and Selective Embolization of Blood Vessels

DSA is an important supplementary technique in sacral surgery. It can show the blood supply of the pelvis and the tumor clearly and block tumor blood supply, thus reducing intraoperative hemorrhage through selective vascular embolism, ensuring the successful completion of the surgery, and achieving a good result.

Internal iliac artery embolization is performed as follows. One day before the surgery, or on the surgery day preoperatively, the Seldinger method is used to perform femoral artery puncture. The catheter is inserted through the femoral artery to the proximal end retrogradely. After abdominal aortic angiography is performed, the catheter is inserted into the unilateral or bilateral internal iliac artery to observe the location, nature, range, and blood supply of the tumor. The bilateral or unilateral internal iliac artery (usually the side that the tumor involves more) is embolized, as well as other target arteries that can be embolized using embolic materials, such as gelfoam or coil (Fig. 33.1). The DSA machine is used. After embolization, a pressure bandage is applied at the puncture point. Sacral and pelvic surgery should be performed within 24 h.

The common DSA embolization method before complex pelvic surgery involves blocking of bilateral internal iliac arteries. There are two main shortcomings about this traditional method: first, after embolization, patients may suffer from abdominal discomfort, such as acute abdominal pain, from wound healing problems after surgery; second, the hemostatic effect of embolization is usually unsatisfactory.

Two main vascular networks exist in the human iliac region. One is the network between the bilateral internal iliac arteries, which has rich traffic branches and participation of other vessels from the abdominal and pelvic cavity. This network dominates the pelvic area, especially the gluteus muscle and the lateral pelvic wall. The superior gluteal artery springs from the internal iliac artery. Because of the rich traffic branches of this network, blocking one side or even two sides of internal iliac arteries completely is relatively safe. To date, most researchers pay attention to such characteristics of this network and regard bilateral iliac arterial embolization as a regular iliac vascular embolization method. The other is the network between the internal and external iliac arteries on the same side. This network is also important but does not cause enough attention. This network mainly comprises the iliolumbar artery, which springs from internal iliac artery,

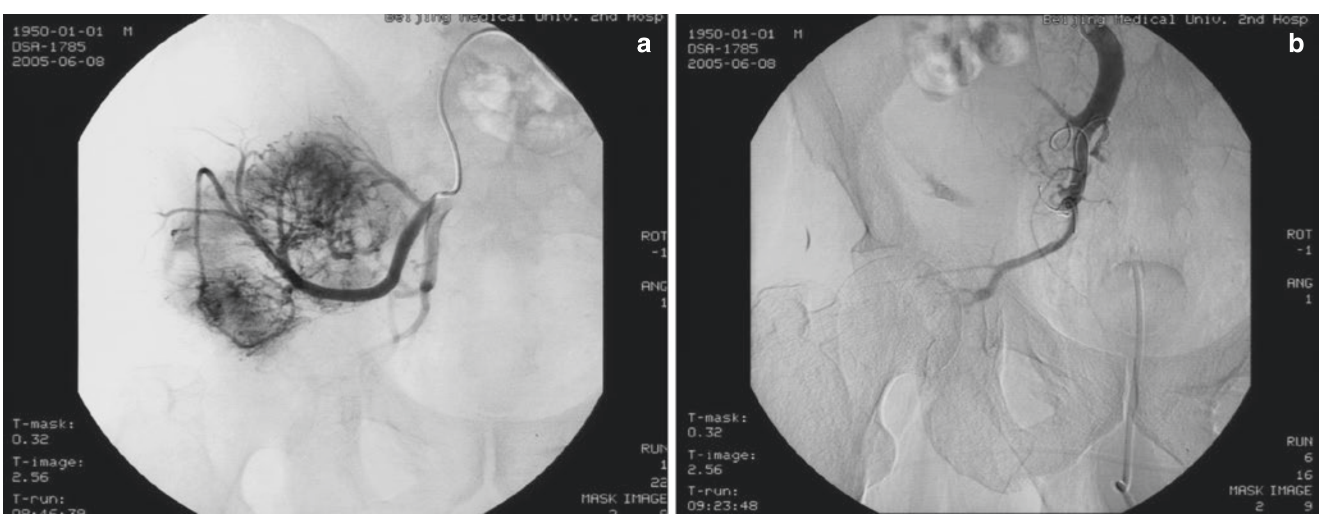

Fig. 33.1 Preoperative DSA to embolize the internal iliac artery in a patient with pelvic tumor. (**a**) The internal iliac artery dominates the tumor blood supply, and its blood supply is rich. (**b**) After internal iliac artery embolization, the tumor blood supply reduces dramatically

and the deep circumflex iliac artery, which springs from external iliac artery. It also involves the lateral circumflex femoral artery as well as some branches from lumbar artery. This network is mainly distributed in the pelvis surface and its surrounding muscles, which is the right location and the surgical area of most pelvic tumors, so the blood supply of the pelvic region is of vital importance.

In our cases, sometimes hemorrhage control during surgery is not achieved after the preoperative bilateral internal iliac artery embolization. For cases with huge sacral tumors, we use the anterior approach to ligate the unilateral internal iliac artery as well as temporarily block the abdominal aorta, so that hemorrhage during surgery can be effectively controlled. For cases with smaller sacral and pelvic tumors whose estimated blood loss is not obvious, anterior incision for vascular occlusion is not necessary. The aortic balloon occlusion technique developed recently is an effective method for hemorrhage control intraoperatively, controlling the blood flow of the abdominal aorta and reducing the amount blood loss during surgery. For suitable cases, we can purely implant an abdominal aorta balloon to achieve satisfactory hemorrhage control without blocking the bilateral internal iliac artery. Besides, after the surgery, the abdominal aortic balloon is removed to reduce the occurrence of postoperative wound complications and ensure the surgical safety.

33.3.2 Temporary Aortic Balloon Occlusion During Sacral Tumor Resection

Preoperative embolization and/or ligation of the internal iliac vessels may sufficiently control bleeding in some sacral tumor surgery. However, the abundant blood supply of sacral tumors could also come from branches of the middle sacral vessels, the internal, external, and common iliac vessels, and sometimes even from the abdominal aorta. A more effective bleeding control method is needed for patients with high risk of severe hemorrhage. Therefore, aortic balloon occlusion has been recently introduced into sacral tumor surgery. Temporary aortic occlusion by a BDC, which has been successfully used for cardiovascular disorders, has successfully reduced blood loss in a few studies on sacral tumor surgery. In our institute, before 2003, surgical cross-clamping of the aorta was performed in only a small number of patients in whom brisk bleeding was encountered during an anterior approach; most patients received no aortic control. Beginning in 2003, balloon occlusion took the place of cross-clamping of the aorta during sacral tumor resection in the hope of reducing blood loss. Because aortic balloon occlusion is only considered in patients with high risk of extensive bleeding, preoperative evaluation should be carried out thoroughly. Patients with a tumor that invades cephalad to the S2–S3 disc space, which has a volume of >200 cm^3 or has an abundant blood supply, are candidates for aortic balloon occlusion.

Abdominal aortic balloon occlusion is performed as follows. A BDC (Maxi LD; Cordis, Johnson & Johnson, Bridgewater, New Jersey) is inserted into the lower abdominal aorta through an 11F percutaneous introducer sheath (CROSSOVER; Cordis) inserted into the femoral artery 1 h before surgery. On inspection under an image intensifier, the balloon should be placed distal to the superior mesenteric artery and the renal artery. The balloon is then dilated using a diluted contrast medium until it is no longer moving with the impulse of the bloodstream, which means that the aorta has been occluded. After aortic blood flow has completely stopped and both renal arteries are clearly visualized, the contrast medium in the balloon is evacuated, and the volume needed for intraoperative aortic occlusion is recorded. During the operation, balloon dilation starts when the surgeons are ready to begin the dissection of the tumor mass or facing an unexpected hemorrhage; the dilation continues until the tumor is completely removed, a process during which brisk bleeding is always encountered. The occlusion time is usually no more than 90 min. When a longer occlusion period is needed, the balloon is deflated for 10–15 min after 90 min and again reinflated.

Monitoring of the abdominal aortic balloon catheter during operation is essential. In the whole procedure, we usually use the arterial pressure measurement as a monitor method. Once the artery puncture is finished, we connect the sheet to a pressure measuring device and compare the parameter achieved with the radial artery pressure so that we can indirectly observe whether the puncture is successful and monitor the general condition and basic parameters of the lower extremity vessels. When we block the abdominal aorta blood flow, the arterial waveform breaks into a straight line and the reading data maintains a low fixed value, which means the blood flow of lower extremity arteries is almost blocked. The pulse of the dorsalis pedis artery should also be monitored to ensure complete occlusion of the aorta after dilation of the balloon. By maintaining this level of occlusion, the tumor surgery can be safely performed. During the blood flow occlusion, we need to monitor the reading continuously to observe the blocking effect. Postoperative femoral arterial thrombosis can be effectively avoided by discontinuously pumping small amounts of heparin saline through the femoral artery sheath. The evacuated BDC is extracted along with the introducer sheath immediately after the operation. The puncture site should be compressed continuously for 24 h to avoid hematoma. If the patient's condition after surgery is not yet stable, the arterial sheath should be retained. We need to continue the observation of the lower extremity condition and maintain the pumping of small amounts of heparin saline. Whenever necessary, we can reperform angiography through the arterial sheath to observe the blood supply of the surgical region and reduce postoperative hemorrhage by using the embolization method. Oxygen saturation signals are monitored in the toes of both feet postoperatively for early detection of a possible thrombus.

In our retrospective study [8] including 120 patients with sacral tumor treated with balloon occlusion compared with 95 patients with sacral tumor with a high risk of brisk bleeding without using of aortic balloon occlusion, we found that the use of aortic balloon occlusion during sacral tumor surgery can decrease the total and intraoperative blood loss volumes but not the amount of postoperative blood loss. There was no significant difference between the two groups in terms of age, sex, grade of malignancy, tumor blood supply, location of the tumor, percentage of patients who had a recurrent tumor or previous surgery or radiation therapy, surgical approach, or type of resection. Patients treated with aortic balloon occlusion had a larger mean tumor volume, more frequently had a sacral reconstruction, and had a longer mean operative time than patients with no aortic control, and more of the primary malignant tumors had an adequate surgical margin in the group with aortic balloon occlusion. The total blood loss volume was 4337 mL in patients without aortic balloon occlusion compared with 2963 mL in those with occlusion. The mean blood loss volume during the operation in patients without aortic balloon occlusion was 3935 mL, whereas it was 2236 mL in patients treated with occlusion. In contrast, patients without aortic balloon occlusion had less postoperative blood loss (402 mL) than patients treated with occlusion (727 mL). A comparison of intraoperative and postoperative blood loss volumes between the two groups revealed that, although intraoperative bleeding can be reduced by aortic balloon occlusion, the postoperative blood loss increased compared with that in the control group. This finding might be attributed to the larger tumor volume, which led to a larger wound surface, in patients with aortic balloon occlusion.

Aortic balloon occlusion has some drawbacks. It has negative effects on circulation and metabolism. Metabolic acidosis and an increase in lactate concentration, particularly in blood drained from ischemic tissues below an aortic occlusion, have been demonstrated in animals and humans during cross-clamping (and to a greater extent on unclamping) of the infrarenal aorta [9]. After release of the aortic balloon, acidosis and increased potassium levels are frequently encountered. These metabolic changes would eventually cause intravascular fluid loss and more bleeding postoperatively [10]. To monitor the patient's metabolism, blood gas analysis is conducted twice, once before the dilation and once after the release of the balloon. Sodium bicarbonate is sometimes needed in patients with acidosis.

The rate of balloon-related complications is low. Balloon-related complications are defined as any vascular issue, such as injury, ischemia, embolism, hematoma, or infection, that involves the abdominal aorta, common iliac artery, external iliac artery, or femoral artery perioperatively. Cases of acute aortic thrombosis and rupture following aortic balloon dilation have been reported sporadically in patients with aortic disease, such as aneurysm, arteritis, and sclerosis [11, 12]. The most common complication related to aortic balloon occlusion in our patients is hematoma at the puncture site, which does not usually need treatment. The hematomas formed mainly because we used a large-diameter percutaneous introducer sheath, which facilitates insertion and extraction of the BDC. Femoral artery embolism is another serious complication which required immediate embolectomy. Doctors should pay more attention on the patient's blood circulation of the lower extremity. A good prognosis could be expected if early detection and treatment of arterial embolism are carried out.

Aortic balloon occlusion during sacral tumor surgery is suitable for patients in whom excessive blood loss can be anticipated; however, it should be done with caution in patients with aortic disease, such as aneurysm, arteritis, and sclerosis. If conditions permit preoperatively, we can observe in the enhancement scanning of the tumor imaging if obvious abnormalities of the vessel exist from the femoral artery to the lower segment of abdominal aorta. For instance, aortic aneurysm, aortic dissection, and other serious vascular diseases are contraindications for aortic balloon occlusion. For aortic plaques, the aortic balloon still can be applied, but we need to perform repeated angiography for observation and confirmation during the procedure and to determine the embolization effect. The balloon should be placed in the distal area of bilateral renal artery ostia. Normal saline is injected into the balloon and the right saline amount is ensured to completely occlude the abdominal aortic blood flow (Fig. 33.2).

Fig. 33.2 In a sacral tumor case, an abdominal aortic balloon was placed via the femoral artery sheath preoperatively to reduce the blood loss during surgery. After the balloon inflation, the contrast agent did pass through because the abdominal aortic blood flow was completely blocked

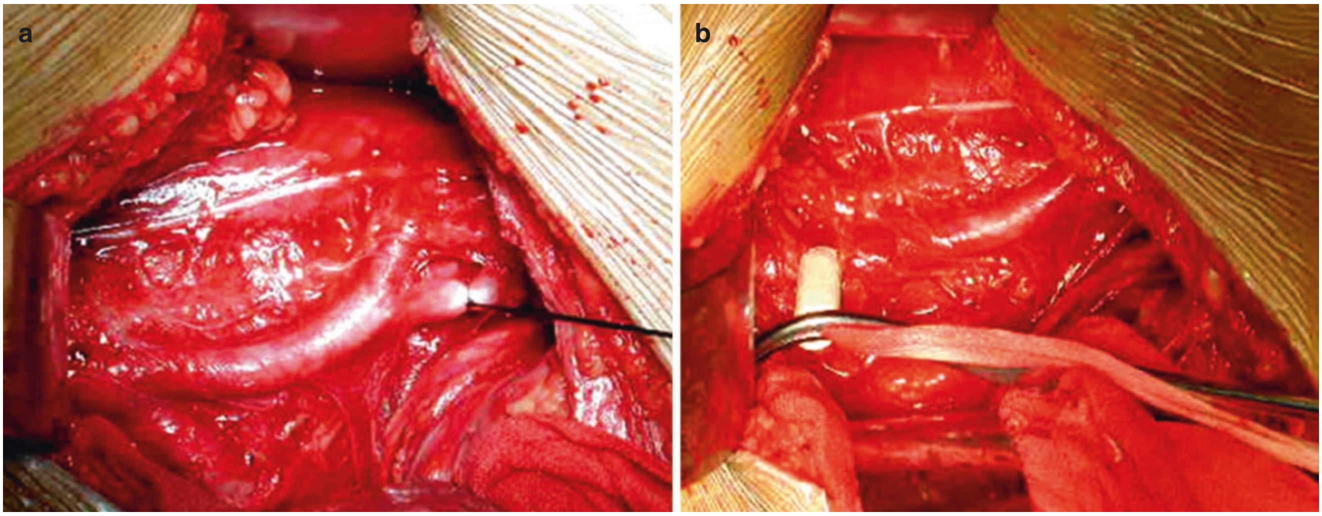

Fig. 33.3 Intraoperative views of sacral tumor surgery. (**a**) Ligation of internal iliac artery. (**b**) Temporary blocking of the abdominal aorta

33.3.3 Intraoperative Ligation of Internal Iliac Artery and Temporary Abdominal Aortic Occlusion

The patient is in a lateral position during the procedure, and a large anterior McBurney incision is made at the affected side. After the three layers of abdominal muscles are incised and the peritoneum is pushed to the unaffected side, the common iliac vessels and internal iliac artery as well as external iliac artery of the same side are exposed. Then, the ipsilateral internal iliac artery is identified, separated, and ligated. If necessary, the contralateral internal iliac artery is also ligated. The abdominal aorta is separated and exposes toward the proximal direction; gauze tapes with rubber tubes are used to block the blood flow of abdominal aorta at the point 1 cm above the bifurcation of the common iliac artery (Fig. 33.3).

33.4 Summary

The hemorrhage control effect of lower abdominal aortic blocking is evident. Blood supply and vascular traffic branches of the pelvis and sacrum are very rich. The amount of blood loss during tumor resection surgery in this site can reach 2000–10,000 mL. For the purpose of blood loss reduction, methods are used clinically, such as internal iliac artery ligation, vascular embolization via intraarterial intervention, and temporary abdominal aortic balloon occlusion. By applying these techniques, blood loss is reduced dramatically and the surgical site is clearly exposed, finishing the surgery successfully, which was almost impossible earlier. Low blood loss volume during tumor resection surgery provides the surgeon more time to perform the operation carefully, which ensures the quality of tumor resection. In addition, incidence of hemorrhagic shock is reduced because short-time massive hemorrhage will not happen during surgery.

The effect of abdominal aortic occlusion on systemic hemodynamics and visceral ischemia–reperfusion injury changes with the occlusion level. Lower-level abdominal aortic occlusion practiced clinically is relatively safe. Occlusion of the abdominal aorta above the bifurcation of the common iliac artery does not block the blood supply of ischemia-sensitive organs, such as the liver, kidney, and spinal cord. In addition, although the ovary and testis located in lower abdomen are also sensitive to ischemia, the ovarian or testicular artery that supplies them dominantly springs slightly lower than the renal artery ostia level. Hence, its blood flow is also not blocked, and the blood supply of ovary or testis is not affected by abdominal aortic occlusion during surgery. The single blocking time of the abdominal aorta should be no longer than 90 min; repeated blocking can be performed if necessary. The temporary occlusion of the lower-level abdominal aorta practiced clinically has been shown to be safe.

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34 Complications After Sacrectomy and Pelvic Tumor Surgery

Dasen Li and Wei Guo

For most primary and some metastatic tumors of the bony pelvis, sacrectomy or pelvic resection remains the main therapeutic modality. Sacral or pelvic tumor resection is a challenging and time-consuming procedure due to the complicated regional anatomy. The incidence of wound complications, including surgical site infection (SSI), and wound dehiscence remains high. These events will markedly influence the postoperative course of patients [1–3]. *As sacral tumors often involve sacral nerve roots and even the cauda equina in the sacral canal, because of the unique regional anatomy of the sacrum, damaging or sacrificing sacral nerves yields permanent motor/sensory deficits or loss of bowel, bladder, or sexual function. These wound and neurological complications should be considered carefully and discussed with the patient, because they are closely related to the postoperative quality of life of the patients.*

34.1 Wound Complications

SSI was defined by Horan et al. Wound dehiscence is defined as a wound showing breakdown in the absence of clinical signs meeting the SSI diagnostic standard [4].

34.1.1 Wound Care After Sacrectomy

34.1.1.1 Causes of Wound Complications

Sacral tumors often have a large mass at diagnosis. Resection of these tumors leaves a very large dead cavity. Postoperative computed tomography and magnetic resonance imaging show that this large dead cavity fills with hematoma in the short term and is eventually filled by scar tissue and the expanded rectum.

The distal end of the posterior sacrococcygeal incision is adjacent to the anus. Many patients may suffer from bowel incontinence, and bacteria from the GI tract may contaminate the wound.

Preoperative arterial embolism, wide soft tissue stripping during tumor resection, and ligation of the internal iliac artery and/or gluteal artery can lead to poor blood supply to the skin flap. The dorsal ramus of the sacral nerves is sacrificed during the operation, with the resulting neural atrophy having a negative impact on wound healing.

In addition, the sacrum connects the mobile spine with the pelvis and plays an important role in the mechanical transmission; thus, internal fixation is often necessary during spine–pelvic continuity reconstruction. Reconstruction prolongs the operation and the metallic components that are implanted in the wound act as foreign bodies.

Cerebrospinal fluid (CSF) leakage is another important issue in sacral wound care. The distal end of the dural sac at sacral level 2 and the sacral segment of the dural sac bear the highest static water pressure when the patient stands or sits upright. The sacral dural sac ought to be closed after tumor resection, via ligation or direct suturing. However, a huge tissue defect would be left around the sacral segment of the dural sac, and it is not easy to pad muscle or fat tissue tightly around the impaired or repaired dural sac. Postoperative cerebrospinal fluid leakage can cause delayed wound healing and, eventually, SSI and even central nervous system infection.

Based on these facts, historically, sacral procedures have had higher rates of infection compared with other spinal procedures [5, 6]. Their postoperative infection rates were 8.3–39.1%. The wound complication rate reported by Gabra et al. in 2006 was 60.6%. In a study reported by us in 2013, among 387 patients with a primary sacral tumor, 274 wounds healed uneventfully, whereas 113 wounds broke down because of infection or dehiscence, giving an SSI rate of 13.2% and a dehiscence rate of 16.0%. The total wound complication rate was 29.2% [2].

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34.1.1.2 Prevention and Treatment of SSI

Perform preoperative bowel preparation. Use prophylactic antibiotics perioperatively and administer additional doses of antibiotics during operations that last more than two half-life periods of the antibiotics or with a blood loss of more than 2000 mL. The use of prophylactic antibiotic therapy has significantly decreased the rate of SSI.

Although a study of 678 patients undergoing surgery for a spinal tumor (excluding sacral tumors) reported by Omeis et al., found that *Staphylococcus aureus* was the most commonly isolated organism ($n = 22$ of 65; 33%). We reported that Gram-negative organisms were cultured from 91.8% of the infected sacral wounds, with most being *E. coli*. Because *E. coli* are believed to grow in the gastrointestinal tract, the proximity to the perineum may be one reason for the high SSI rate of sacral wounds. Gram-negative bacteria should be well covered by the antibiotics administered perioperatively.

Colostomy is an operation during which bacteria from the bowel may contaminate the wound, which may result in SSI and failure of the orthopedic reconstruction. If colostomy is planned, it may be performed first, with sacrectomy being performed at a second stage.

Meticulous adherence to the aseptic technique is the single most important component of SSI prevention. Close the deep fascia tightly to waterproof level. Select silicon drainage tubes with a diameter around Fr 24, for wound drainage, and keep them until the daily drainage volume is <30 mL. Systemic antibiotics can be used until the removal of the drainage tubes. This may keep the incision dry when CSF leakage occurs. Pay attention to the characteristics of the fluid that is drained from the wound. If the wound drainage fluid becomes cloudy or the patient has a fever, perform organism culture of the drainage fluid and/or blood.

Protect the wound from contamination with feces when dressing the wound. For patients with postoperative anal sphincter dysfunction, this is not easy, because the intestinal content can leak out and contaminate the nearby sacrococcygeal wound at any time and senselessly.

To avoid sacrococcygeal pressure sores, all possible means, including the use of an anti-bedsore cushion, should be applied, and the presence of a team of professional nurses is inevitable.

For postoperative SSIs, perform wound debridement and apply sufficient drainage immediately. In some cases, more than one debridement procedure, and even vacuum sealing drainage, are needed. To control the infection, in some cases, implants have to be removed during debridement. Use regimens of culture-sensitive intravenous antibiotics for at least 3 weeks, and long-term suppressive oral antibiotics may be necessary in rare cases.

The sacrum is a unique structure that connects the lumbar spine and the pelvis; once postoperative SSI occurs, the decision about removing the instrumentation represents a dilemma. According to our experience, the instrumentation may be preserved during debridement in a large proportion of patients. The condition, type, and volume of the implants; the timing and quality of the debridement; and the characteristics of the infecting organism are factors that should be considered and balanced carefully. Early debridement is associated with a good result.

Based on our study, previous radiation, rectum rupture, longer duration of surgery, and CSF leakage are significantly associated with an increased likelihood of developing an SSI. These risk factors should be considered when planning the operation and managing sacral wounds. A rectus abdominis musculocutaneous flap is an option for patients who have a poor local soft tissue condition. For large sacral and perineal defects after sacrectomy with or without radiation, one option is to use a rectus abdominis myocutaneous flap. This flap has two advantages; the first is the wide arc of rotation allowed, based on the deep inferior epigastric vessels, and the second is the large bulk of tissue afforded, which will fill the large soft tissue defect in the sacrococcygeal region [7, 8].

34.1.1.3 Prevention and Treatment of Wound Dehiscence

Wound dehiscence is often caused by aseptic fat liquefaction and/or the skin and subcutaneous necrosis caused by ischemia.

Aseptic fat liquefaction is the necrosis of adipose tissue without infection and is the main cause of prolonged healing of aseptic postsurgical incision. It often occurs in overweight patients with diabetes or malnutrition.

Flap ischemia can be caused by many factors. An improper body position may lead to a pressure sore in the sacrococcygeal area or to flap ischemia (Fig. 34.1). We observed that

Fig. 34.1 Skin flap ischemia. Preoperative Internal iliac artery embolization was performed in this case

patients who underwent total sacrectomy rarely had flap ischemia. This phenomenon can be explained by the fact that the total loss of the sacrum reduces the pressure in the sacrococcygeal area. Embolization of the internal iliac artery and its branches may lead to flap ischemia in the sacrococcygeal region, and highly selective embolization of the tumor vasculature may reduce the incidence of flap ischemia strikingly.

Wound dehiscence is an aseptic situation in its early stage, but tends to develop to SSI soon after, because of the lack of normal skin tissue, which acts as an anatomical and immunological barrier against pathogens. Because there is a dead cavity and sometimes an implant in the wound, early surgical debridement is necessary in some cases, and a gluteus maximus myocutaneous flap or other flaps can be considered for repairing soft tissue defects after excision of the necrotic tissue.

34.1.1.4 Prevention and Treatment of CSF Leakage

A cerebrospinal fluid (CSF) leak caused by dural tear or resection is a common complication of surgery for sacral tumors. When the sacral nerves or part of the dural sac have to be resected with the tumor, ligate the proximal end of the sacral nerves or create a water-tight dural closure. As mentioned above, always close the deep fascia tightly. Place one or two silicon drainage tubes (Fr 24 or Fr 28) for wound drainage and keep them until the daily drainage volume is <30 mL. Thus, the skin wound remains dry and will heal even in the presence of a CSF leak. Patients with CSF leakage should remain in a Trendelenburg position after the operation. If the drainage volume is >30 mL until the 14th postoperative day, it could be blocked temporarily. In the absence of fever, CSF leakage from the wound, and signs of meningitis for >24 h, remove the drainage tube and dress the wound with pressure. Most patients with a CSF leak can be healed using a conservative approach. Lumbar drainage is needed only in rare cases.

CSF leakage may lead to SSI. Debridement should be performed as soon as possible once SSI occurs.

34.1.2 Wound Care After Pelvic Resection

When pelvic region III or I is resected alone, the resulting wound is relatively small, the skin flaps have good blood supply, and the probability of the development of wound complications is low. It is easy to deal with SSIs that occur in these two areas. However, this is not the case for periacetabular tumors. The incidence of wound complications in both external hemipelvectomy and internal hemipelvectomy is high.

34.1.2.1 External Hemipelvectomy

When external hemipelvectomy is considered for pelvic tumor, the incision should be planned individually. The factors that must be considered during the design of the incision include the scope of skin invasion by the tumor, incisions used in previous operations, and the lack of vitality in the area caused by radiation. In fact, standard incisions for external hemipelvectomy, including posterior flaps and front flaps, can be applied only in a small number of cases.

Among the 68 cases of external hemipelvectomy reported by Apffelstaedt in 1996, 11 cases (16%) had skin flap necrosis and 24 cases (35%) had a wound infection; i.e., about 50% of the patients who underwent amputation suffered from wound complications [9]. Between July 2004 and June 2014, we performed 70 external hemipelvectomies. Posterior flaps, front flaps, and anteromedial flaps were used in 66 cases, 3 cases, and 1 case, respectively. Two patients died perioperatively, 10 patients had SSI, and 10 patients had wound dehiscence caused by skin flap necrosis or fat liquefaction.

Maintaining the blood supply to the skin flap is key for reducing wound complications. In most patients, a posterior flap incision is the first choice, and gluteal arteries should be preserved when possible. When the gluteus maximus muscle and the deep fascia have to be resected for oncological reasons, the skin flap may not receive a blood supply that is sufficient for avoiding ischemic necrosis.

When the external iliac artery and the femoral artery can be preserved, an anterior flap incision, i.e., a quadriceps myocutaneous flap, is a practical option. For older patients and patients with arterial disease, the ipsilateral femoral artery should be assessed preoperatively, to exclude arteriosclerosis and stenosis.

34.1.2.2 Internal Hemipelvectomy

Internal hemipelvectomy is a time-consuming and very technical procedure. There are two options after this procedure: reconstruction or absence of reconstruction. Hemipelvic endoprosthesis has been used widely to reconstruct large bone-articular defects, with a much better postoperative function. However, surgeons must be cautious when using hemipelvic endoprosthesis or other similar implants, because wound complications, especially SSI, may lead to devastating consequences. For patients in whom the condition of the soft tissue does not allow complicated bone-articular reconstruction, i.e., only a radiated wound or skin flap is left, no reconstruction (a flail hip) is also a choice. The absence of implants, although associated with a poor functional prognosis, may reduce the incidence of wound complications and greatly facilitate debridement, if necessary.

In 2012, we reviewed retrospectively the charts of 100 patients who underwent modular hemipelvic endoprosthetic reconstruction for pelvic tumors from June 2001 to March 2010. The most frequent complication was wound healing disturbance, which occurred in 18 patients (11 cases of superficial skin infection and seven cases of skin necrosis), 15 of whom underwent debridement. Another 15 patients had deep infection, 3 had acute infection, and the remaining 12 cases had late deep infections at a mean time of 15.6 months after surgery. Recurrent discharge or fistula was a common manifestation. Twenty-seven debridement surgeries were performed on these 15 patients. The wound healed in four patients after debridement, irrigation, and the administration of systemic antibiotics. The endoprosthesis was removed in six patients. The remaining five patients had hemipelvectomy because of intractable infection. Three of the five patients who had received radiotherapy before modular hemipelvic endoprosthetic reconstruction developed a deep infection [10].

To reduce the probability of wound complications, a limb salvage surgery should be considered only in patients with a relatively good soft tissue condition. If a hemipelvic endoprosthesis or other large implants are used, enclose them with muscles. Choose antibiotic-impregnated cement when necessary. The tip of a retrograde flap located just below and lateral to the anterior superior iliac spine is vulnerable to ischemia and necrosis. Do not free this skin flap from the deep fascia and try to reduce its tension during wound closing. The measures that are used to deal with wound complications after sacrectomy should also be applied to pelvic wounds. As pelvic wounds are adjacent to the perineum, prophylactic systemic antibiotics must cover both Gram-negative and -positive bacteria. Additional intraoperative antibiotics should be administered according to the duration of the operation and blood loss. Identify skin-edge necrosis early, perform debridement as soon as possible, and use a skin or muscular flap to close the wound.

Techniques and tips used to diagnose SSI after sacral tumor resection could be also be applied here. In the early postoperative period (within 3 weeks), peri-incisional erythema, tenderness to palpation, and body fever or chills are particularly alarming. The white blood cell count, the erythrocyte sedimentation rate (ESR), and C-reactive protein (CRP) levels have been shown to be indicators of the presence of SSI. Cultures from the drainage and B ultrasound-guided wound aspiration are good options to confirm SSI.

Irrigation and debridement may be considered for early postoperative SSI, and the artificial internal hemipelvic prosthesis may be left in place. Alternatively, it should be removed and a flail hip joint procedure without reconstruction is preferred after debridement. In a small number of patients, external hemipelvectomy (external hemipelvectomy) remains a reasonable and reliable alternative.

34.2 Neurological Complications of Sacrectomy

In the 1970s, Gunterberg et al. published a series of studies concerning neurological disorders in patients who had undergone sacrectomies. Those authors reported that the unilateral sacrifice of S1–S5 roots does not affect bladder and bowel function significantly, and that the preservation of both S3 roots is sufficient for normal function [11]. Larger and well-documented studies were carried out subsequently and identified the importance of S3 nerve roots in preserving postoperative function. The sacrifice of sacral nerves during operation depends on the extent of the resection margin.

To date, there is no effective method to recover the function of the sacrificed sacral nerves. In a very small number of cases, sacral malignances may originate from an ala sacralis and only involve the ipsilateral sacral nerves. If the contralateral sacral nerves are not affected by the tumor and are located beyond the resection line, en bloc resection of the hemisacrum vertically may be performed. Thus, a much better postoperative function can be expected.

Unless the rectum is affected by the tumor and must be removed, no colostomies are performed at our center. During the follow-up process, the vast majority of patients who have anal sphincter dysfunction learn to control their bowels through a diet-based mild constipation, and increased abdominal pressure helps defecation. In our experience, very few patients request a colostomy postoperatively.

34.2.1 Motor Function of the Lower Limbs

The sciatic nerve is formed by the lumbosacral trunk and nerve fibers from the S1 to the S3 nerve roots. All of these nerve fibers unite to form the sciatic nerve in front of the piriformis muscle. Then, the nerve passes beneath the piriformis muscle and through the greater sciatic foramen, exiting the pelvis.

In our experience, although slight gait abnormalities or mild weakness in the lower limbs may be observed, and intermittent use of external support, such as a cane or crutch, was found in some patients, walking function is not remarkably impaired after sacrectomy, unless the ipsilateral lumbosacral trunk is damaged. The sacrifice of the sacral nerves, including the bilateral S1 nerves, does not affect the patient's gait obviously, because gravity compensates for the loss of ankle plantar flexion strength. The lumbosacral trunk comprises the whole of the anterior division of the fifth lumbar nerve, and part of the anterior division of the fourth lumbar nerve, and runs downward over the pelvic brim, to join the first sacral nerve just in front of the S-I joint. The lumbosacral trunk may be adhered or even infiltrated by the sacral tumor. As the lumbosacral trunk is responsible for important lower-limb functions, such as the ankle dorsal flexion, claudication may occur when it is damaged.

34.2.2 Bowel, Bladder, or Sexual Dysfunction

34.2.2.1 Neuroanatomy of the Pelvic Organs

Automatic and somatic nerves control defecation and urination. The automatic nerves include sympathetic and parasympathetic nerves. Sympathetic outflow occurs from the first and second lumbar segments of the spinal cord. Sympathetic nerves innervate the bladder neck, trigone, and urethral area. These nerves also inhibit the contraction of the detrusor muscle of the bladder wall and stimulate the closure of the sphincter vesicae. Parasympathetic outflow occurs from sacral segments 2–4 of the spinal cord. Parasympathetic nerves innervate the detrusor muscles of the bladder wall and inhibit the action of the sphincter vesicae.

Somatic outflow encompasses the first and second sacral spinal segments of the spinal cord, running in the S2 and S3 nerve roots, which comprise somatic motor nerve fibers and somatic sensory nerve fibers. Somatic motor fibers form the pudendal nerve, which innervates the external urethral sphincter, the external anal sphincter, and other skeletal muscles of the pelvic floor. These nerve fibers also join the inferior gluteal nerve and sciatic nerve to innervate the skeletal muscles of the lower limbs. Somatic sensory fibers arise from the S2–S4 dorsal root ganglia, run mainly in the pudendal nerve, and conduct pelvic skin sensation and sexual arousal. In a handful of cases, segments up to the S1, or as low as the S5 or C0 (tail), may also be involved in the innervation of pelvic organs.

The integrity of the complicated innervation network is the premise of normal sphincter function and sexual function.

34.2.2.2 Selective Innervation of the Bladder, Bowel, and Sexual Organs by Different Sacral Nerve Roots

Innervation of the bladder detrusor muscle and urethral sphincter. By intraoperative electrical stimulation of each individual S2–S4 root, Zhang Shi-min et al. observed the frequency of differential sacral root innervation of the bladder detrusor muscle in 10 patients with complete supraconal spinal cord injury. They found that the parasympathetic innervation of the bladder detrusor muscle in Asians is mainly provided by the S3 and S4 roots. The S3 root was the most frequent (100%) and the most efficacious (52.2%) contributor. S4 was the second most frequent (90%) and a still significantly efficacious (44.9%) contributor, while S2 was the least frequent (25%) and the least efficacious (2.9%) contributor [12]. Juenemann et al. dissected three male human cadavers and demonstrated that the extrinsic urethral sphincter, which is formed by the rhabdosphincter around the membranous urethra, the levator ani muscle, and the pelvic floor (especially the transversus perineal muscle), is innervated by somatic nerve fibers that emanate primarily from the sacral roots S2 and S3. In five patients with neurogenic lower urinary tract dysfunction, electrostimulation of the sacral root and pudendal nerve markedly increased the intraurethral closure pressures. Almost 70% of the closure pressure of the external urethral sphincter was induced by stimulation of the S3 ventral root, while the remaining 30% was derived from S2 and S4 neuronal impulses [13].

MacDonagh et al. reported clinical and manometric data collected from 12 consecutive patients with spinal injuries who had undergone implantation of sacral anterior root stimulators. They found that stimulation of the S2 root produced low-pressure colorectal activity, whereas stimulation of the S3 root and, to a lesser extent, the S4 root, produced colorectal contraction. Stimulation of S4 gave the maximum pressure response in the anal canal, almost certainly because of its action on the external anal sphincter [14].

S2 roots carry a significant number of fibers, which are involved in genital sensation. Dorsal root action potentials were recorded intraoperatively to map the distribution of pudendal afferent fibers in S1–S3 roots bilaterally before the performance of rhizotomies in 114 children with a diagnosis of cerebral palsy and debilitating spasticity (Huang et al.). Those authors reported an asymmetrical distribution in the majority of these patients (56%), with S1 roots contributing to 4%, S2 roots contributing to 60.5%, and S3 roots contributing to 35.5% of the overall pudendal afferent activity. The pudendal afferent distribution was often confined to a single level (18% of the patients), or even to a single root (7.6% of the patients) [15].

In general, S3 seems to be the dominant nerve for detrusor and colorectal contraction. S2 is the dominant nerve of urethra or anal sphincter and it also plays an important role in sexual function.

34.2.3 Scoring System for the Evaluation of Sacral Nerve Function After Sacrectomy

In the 1970s, Gunterberg et al. published a series of studies concerning neurological disorders in patients who had undergone sacrectomies. Those authors reported that the unilateral sacrifice of S1–S5 roots does not affect bladder and bowel function significantly, and that the preservation of both S3 roots is sufficient for normal function [11]. This was deemed as the first systematic research study in this field. In 2002, Todd et al. reported a study of postoperative bowel and bladder function in 53 patients, revealing the preservation of at least one S3 root and the preservation of bowel and bladder function in the majority of patients after sacral resection. Among the patients who had unilateral sacrectomy and in whom the contralateral sacral nerves were preserved, normal bowel and bladder function was retained in 87% and 89% of the patients, respectively. All the patients in whom the bilateral S2–S5 nerve roots were sacrificed had abnormal bowel and bladder function. Among patients who had bilateral S3–S5 resection, normal bowel and bladder function was retained in 40% and 25% of the patients, respectively. Among patients who had bilateral S4–S5 resection with preservation of the S3 nerves bilaterally, normal bowel and bladder function was retained in 100% and 69% of the patients, respectively. Among patients who had asymmetric sacral resections with preservation of at least one S3 nerve root, normal bowel and bladder function was retained in 67% and 60% of the patients, respectively. Additional studies were published subsequently that supported these findings. We found that the integrity of the bilateral S3 roots was essential to prevent the incidence of urine leakage. As reported by Juenemann et al. in 1988, S3 nerve roots play a major role in controlling the external urethral sphincter. Contraction of the sphincter may be greatly compromised when at least one S3 is injured, which will result in reduced urethral pressure and an increased incidence of urine leakage [16].

A recent study of 115 patients with sacral chordoma performed by Ji et al. proposed a newly designed score method that was adopted to evaluate critically the functional outcome after resection of the sacrum [17]. By reviewing the literature and summarizing the clinical features of patients who had undergone sacral surgeries at our center, we proposed a scoring system (Table 34.1) that guarantees the accurate description of lower limb, bladder, and bowel function; the system contains nine items and allots 0, 1, 2, or 3 points to each item, according to the degree of functional deficits, thus providing a maximum score of 27 points. These nine items are assigned to three categories: motor function and sensation of lower limbs (M), urination and uriesthesia (U), and defecation and rectal sensation (D) [18].

Cross-sectional data were collected by reviewing all patients who received sacral resections for primary tumors at our center from December 2010 to September 2013. The criteria used for sample collection were as follows: (1) a known primary lesion localized in the sacrum; (2) no previous history of motor system/lower gastrointestinal tract/urogenital disease or other neurological deficits of the

Table 34.1 Scoring system

		3	2	1	0
Motor function and sensation of lower limbs (M)	Pain	None	Mild pain not requiring the use of analgesics	Moderate pain can be controlled by analgesics	Intractable pain/drug dependence
	Motor	Normal	Mild deficit not requiring the help of external support for walking	Deficits requiring the help of external support (cane, crutch, *etc.*) for walking	Deficits requiring the help of wheelchair for motion/ Bedridden
	Perineal sensation	Normal	Mild numbness or hyperesthesia not affecting daily life	Numbness or hyperesthesia affecting daily life	Complete loss of perineal sensation
Urination and uriesthesia (U)	Dysuria	Normal	Mild dysuria not requiring manual exertion of abdominal pressure or medical interventions	Urinary retention requiring manual exertion of abdominal pressure or occasional catheterization (less than once a week)	Urinary retention requiring indwelling catheterization or regular intermittent catheterization/total incontinence/cystostomy
	Bladder incontinence	Normal	Occasional urine leakage not requiring the regular use of diapers	Frequent urine leakage requiring regular use of diapers.	Total incontinence/indwelling catheterization/cystostomy
	Abnormal bladder sensation	Normal	Slightly changed but still existed bladder feelings when micturating	Losing the feelings of stimulus to micturate at times	Total loss of the feelings of stimulus to micturate. Indwelling catheterization/ cystostomy
Defecation and rectal sensation (D)	Constipation	Normal	Mild defecation difficulties not requiring medical interventions	Moderate defecation difficulties requiring regular use of enemas or laxatives	Defecation difficulties requiring manual assistance to evacuate the feces/total incontinence/colostomy
	Bowel incontinence	Normal	Occasional incontinence not requiring the regular use of diapers	Frequent incontinence requiring regular use of diapers	Total incontinence/colostomy
	Abnormal bowel sensation	Normal	Blunted but still existed rectal sensation when voiding	Losing the feelings of stimulus to defecate at times	Total loss of the feelings of stimulus to defecate/colostomy

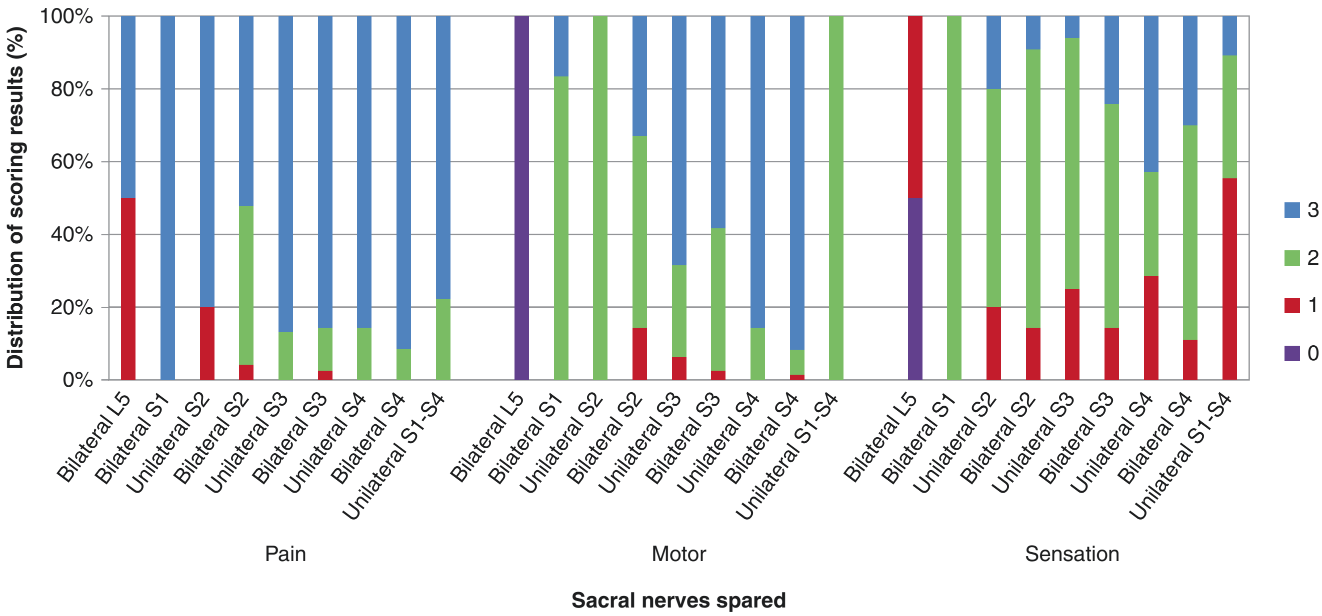

Fig. 34.2 Distribution of the results of scoring of lower limb function at different spared levels (%) [18]

sacral nerves not associated with the lesion; (3) availability of well-documented medical records; (4) availability of detailed records of the sacrificed nerve roots; and (5) surviving patients with no clinical relapse at the time of the latest follow-up. Ethical approval was obtained from the ethics committee board of the participating hospital regarding this study's methods and conduct. Between December 2010 and September 2013, 283 consecutive patients with primary sacral neoplasms underwent 314 sacral resections at our center. A total of 170 patients met the grouping criteria and served as the study population. The scoring results that were obtained using the system described are shown in Fig. 34.2.

34.2.4 Rehabilitation of Sacral Nerve Function After Sacrectomy

When some sacral nerve roots are resected, the function of the remaining ones would be significantly enhanced. With proper behavior management, the impaired sacral nerve function may be recovered completely or partly. In most patients, the recovery period of bowel and bladder sphincter is within 3–6 months. Generally, evaluation of the neural function needs to be at least 6 months after surgery. It usually needs 1 year for sexual dysfunction following operation to resolve gradually, and thus the minimum follow-up interval for determining sexual function should be 1 year.

For patients who undergo total sacrectomy, all sacral nerves are sacrificed, and they have to learn how to deal with bowel and bladder sphincter incontinence in their daily life. Surprisingly, only a very few patients underwent colostomy for fecal incontinence. The following methods may help some patients to recover from postoperative bowel and bladder dysfunction.

34.2.4.1 Behavior Management

An individual schedule for eating, drinking, defecation, and urination should be prepared, and patient compliance should be strict.

Pelvic-floor exercises are aimed at squeezing the muscles that surround the anal canal without contracting the abdominal wall. Encourage the patients to perform these exercises many times per day soon after the operation. During each session, patients should repetitively contract (for 5–10 s) and then relax the pelvic-floor muscles. This exercise strengthens the function of the bowel and bladder sphincters and is helpful in dealing with fecal and/or urinary incontinence.

Defecation is assisted by taking a deep breath and trying to expel this air against a closed glottis (Valsalva maneuver). Contraction of the expiratory chest muscles, diaphragm, abdominal wall muscles, and pelvic diaphragm exerts pressure on the digestive tract. The patient can further facilitate the defecation process by pressing the sacrococcygeal area with one hand. This maneuver may somewhat compensate for the bony loss of the sacrum.

In all patients who undergo sacrectomy, a Foley catheter is left in place for more than 1–2 weeks. In patients in whom some sacral nerves are spared, the bladder dysfunction improves gradually. Usually, the dysfunction is still present at the time of discharge from the hospital, and some patients have permanent bladder dysfunction. In this situation, instructions should be given for intermittent self-catheterization. The cornerstone of bladder management is clean intermittent self-catheterization; however, other treatments are often needed to achieve full continence, to reduce infections and stone formation, to protect the upper urinary tract from excessive bladder pressure, and to prevent chronic renal failure [19]. The Valsalva method or the Crede method (placement of the thumbs on the anterior superior iliac spine and of the remaining eight fingers on the suprapubic area, followed by massaging and pressing of the lower abdomen) help with urination. These exercises might reestablish the urination and defecation reflex arc.

34.2.4.2 Drug Intervention

Drugs such as vitamins B1 and B2 promote nerve cell and muscle cell metabolism and the function of neuromuscular joints. Enema or oral laxatives can relieve persistent constipation. In patients suffering from fecal incontinence, the challenge lies in reducing constipation while avoiding diarrhea. Glycerinum in enema stimulates the bowel wall directly and promotes colorectal peristalsis through a nerve reflex, thus causing bowel movement. Moreover, this nerve reflex also excites the pelvic nerves, leading to bladder detrusor contraction and bladder sphincter relaxation, thus promoting urination.

34.2.4.3 Diet Management

Diet management must be tailored to the patient's clinical manifestations. For persistent constipation, high-fiber foods such as bananas and celery should be eaten. The management of patients with fecal incontinence should be the opposite. Modification of the bowel habits is often the cornerstone of an effective management of bowel sphincter dysfunction.

34.2.4.4 Surgical Treatment

Colostomy is preferred only when the rectum or the sphincter muscle is affected by the tumor. Very few patients need a colostomy, even after total sacrectomy.

Although sacral nerve stimulation is primarily used for urinary and fecal incontinence, sometimes it is used in the setting of constipation. However, this has been used rarely after sacrectomy. Dynamic graciloplasty is another option that is used rarely; it involves continuous electrical stimulation of the gracilis muscle, which is surgically transposed around the anal canal.

Placement of a penile prosthesis offers high patient satisfaction and a low rate of complications in patients with prostate cancer or bladder cancer, and it should be an option for well-selected patients. However, no experience with this procedure has been reported in patients with sacral tumors.

Acknowledgment Figure 34.1 was from the database of our center.

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Rehabilitation After Sacrectomy and Pelvic Resection

35

Jie Xu and Wei Guo

Sacrectomy and pelvic resection are the surgical procedures for treatment of tumors located in sacrum and pelvic ring. Some symptoms can always be found after surgery, such as neurologic dysfunction and pain, which result from instability of the pelvic girdle. This procedure could be associated with severe disability because of the nature of the tumor, surrounding structure, and aggressive surgical intervention. Previous studies have reported neurologic dysfunctions such as sensory abnormalities, bowel, bladder, and sexual dysfunction, and lower limb motor weakness. The type of dysfunction mostly depends on the level and extent of spinal nerve severance. Despite the common agreement on the purposes of rehabilitation after surgery, the actual guidelines for rehabilitation that patients should follow vary among different centers [1]. Moreover, rehabilitation following different anatomical locations, resections, and reconstruction methods have different goals. However, only general description of gait training and exercises about joint and muscles have been reported in the published literature [2, 3]. In this chapter we will discuss the rehabilitation protocol in the postoperative management that addresses different time and anatomical locations.

35.1 General Rehabilitation in Hospital Phase

35.1.1 Immobilization and Pain Management

The etiology of the pain after surgery is multifactorial: increased instability including spine and iliac bone movement, vertical weight-bearing on the surgical area, and neurogenic pain caused by irritation to the residual nerve root [4]. From the biomechanical viewpoint, stability of the pelvic ring is majorly supported by the posterior osseous structures of the ilium, sacrum, and the posterior non-osseous sacroiliac ligaments. Surgery in pelvis and sacrum results in partial or complete dissociation of the spine from the pelvic ring. Pain in the lumbar and pelvic girdle may occur early after surgery because of mechanical instability.

A lumbar-sacral corset with posterior metal stays custom fabricated for patient when sacrum or sacroiliac joint is involved, and then can be used to fix the joint and manage the pain [5]. Previous research shows a 30% decrease of the range of spine motion with the utility of lumbar-sacral corset [6]. Additionally, the corsets also support the abdomen and reduce the stress on the lumbosacral spine. By doing this, it can provide a straighter and more comfortable lower back. This corset is recommended to be used in the daytime, especially during sitting and ambulation to decrease the tension of lumbar and pelvic girdle.

External hip joint stabilizer (Fig. 35.1) and corrective shoes (Fig. 35.2) are recommended after surgery, if acetabulum is involved. Biomechanically, hip abductor is respon-

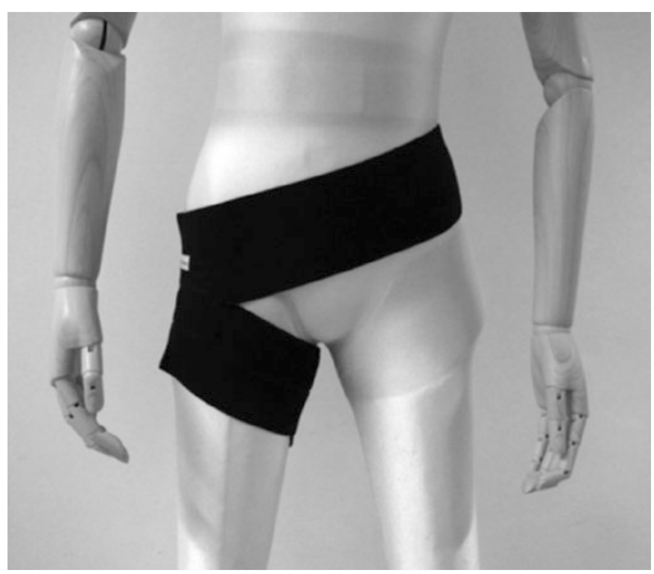

Fig. 35.1 A photograph of the hip stabilizer

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sible for lateral pelvic stability [7]. A careful and proper reconstruction of abductor after proximal femur resection could provide hip joint stability, and is well recognized as a key point to achieve better functional outcomes [8]. Unlike the reconstruction of proximal femur site of abductor muscles, the pelvic site of the bony attachment is typically resected without any kind of reconstruction. In that way, external hip stabilizers are recommended for additional support to hip abductors. The utility of hip stabilizers is not limited in patients following proximal femur resection, but also in those after pelvic resection to achieve better stability and functional outcomes [9]. Evidence from gait analysis showed that both temporal and spatial gait parameters are improved with the help of hip stabilizers. Abductor support elastic band is most commonly used as a simple and effective aid to improve gait function in patients.

Fig. 35.2 A photograph of corrective shoes

35.1.2 Ankle Pump

Ankle pump is recommended in all patients after surgery. Ankle pump, as the most commonly applied exercise for acute phase after surgery, will utilize every muscle in the lower leg. The movements will help to avoid ankle stiffness. On the other hand, the pump function of muscles could also help to squeeze fluid up and out of the leg, and relieve swelling in the lower legs as a result (Fig. 35.3).

35.2 Neurogenic Rehabilitation After Sacrectomy

Neurologic deficit is one of the most common complications after sacral resection and has been mentioned in the most literature about this procedure. In 1970s, Gunterberg et al. published a series of studies concerning neurologic disorders in patients who have undergone sacrectomies, reporting that unilateral sacrifice of S1–S5 does not affect much of the bladder and bowel function and that preservation of both S3s is sufficient for normal function [10, 11]. According to articles published recently, bladder and bowel functions are dependent on the preservation of the third sacral nerve root [12, 13]. There are a wide range of treatment options available to manage neurogenic bladder and bowel dysfunction.

Fig. 35.3 Ankle pumps exercise. Patient is required to lie down supine with both knees straight and pump the feet up and down as far as possible

35.2.1 Neurogenic Bladder Rehabilitation

Neurogenic bladder (NB) or the so-called neurogenic lower urinary tract dysfunction (NLUTD) is a major global medical and social problem. By definition, it refers to the dysfunction of bladder or urethra, which is caused by the damage of nervous system, either the central nervous system or the peripheral nerves. Assessment of the degree of NB is important before rehabilitation. Urodynamic investigations and post-void residual are two important tests after surgery. Both the filling and voiding phases of bladder function are studied in urodynamic investigation, as well as several other parameters, such as compliance of the detrusor, duration of detrusor contraction, and the detrusor pressure [14, 15]. The amount of leftover urine in the bladder after voiding (known as post-avoid residual) can be measured either by catheterization or by a transabdominal ultrasound scan. A bladder ultrasound scan provides a noninvasive method of measuring residual urine, thus avoiding catheterization. A post-void residual of more than 200 ml indicates incomplete voiding problems [16, 17]. If the patient has a partner, it may be appropriate to involve them in the discussions, with the patient's consent.

The aim of NB rehabilitation after surgery is to provide the most acceptable and effective option for the patient, prevent upper renal tract damage, and prevent incontinence. There are many options available for the rehabilitation.

35.2.1.1 Bladder Training

Bladder training is the most important noninvasive strategy comprising three components: education, scheduled voiding, and positive reinforcement [18]. Bladder training involves progressively increasing the time between voiding, and the patient may need to be taught techniques to suppress unstable bladder contractions; for example, pelvic floor contraction to activate the detrusor–sphincter inhibitory reflex [19]. Patients are taught to void every 2 h at first, and then the time may increase to 3 h. Some patients have successfully used a hand massage over the abdominal area to improve bladder emptying. Patients can attempt to remove the ureter catheter if they have a feel of distension in their bladder, successfully pull out urine though the margin of catheter, and demonstrate a post-void residual of less than 200 ml.

35.2.1.2 Intermittent Self-Catheterization

Intermittent self-catheterization (ISC) (Fig. 35.4) has developed over the past 20 years as a method of managing neurogenic bladder disorders. It is appropriate for patients who have a post-voiding residual urine greater than 30% of bladder capacity [20]. Patients in hospital will usually be intermittently catheterized by health-care staff using a sterile technique, but, in the community, they will be taught to use clean intermittent catheterization. However, there is some evidence to suggest that using clean intermittent catheterization does not expose patients to greater risk of urinary tract infection [21]. Patients need to be able to physically prepare for and perform the procedure and have the psychological functioning to understand the technique and the motivation to carry it out up to six times a day.

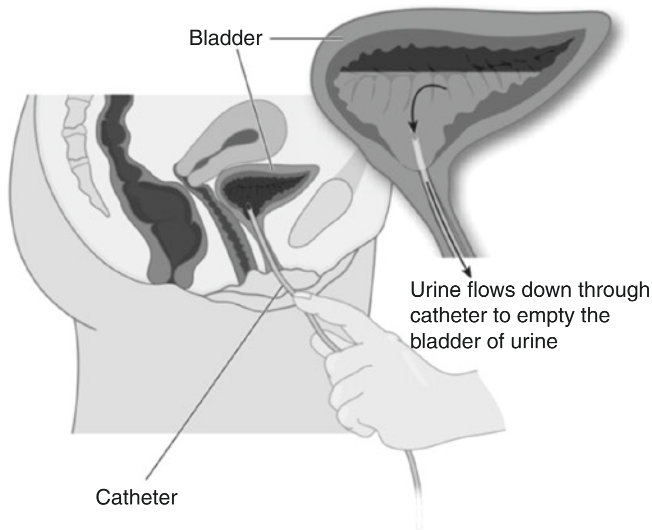

Fig. 35.4 Intermittent self-catheterization

35.2.2 Neurogenic Bowel Rehabilitation

Surgery of sacrum may lead to neurogenic bowel when sacral nerve injury occurs. It will result in fecal incontinence, constipation, or, more often, both. No definitive strategy of rehabilitation has been identified to manage neurogenic bowel dysfunction; developing an acceptable and effective management program relies on individualized assessment and a process of trial and error to develop personalized solutions for patients.

35.2.2.1 Conservative Interventions

Although no robust evidence has been noticed for any dietary intervention, we recommend patients to manipulate their diet to assist their bowel management, especially for those with constipation. Fruits, vegetables, oil, and whole grain foods can help the patients with constipation. First of all, health-care providers should assess the stool consistency of each individual patient, and then give suggestions for diet adjustment accordingly to maintain an appropriate stool consistency.

For patients with constipation, various oral laxatives are recommended to be taken regularly, to modulate stool form and maintain a predictable consistency. Laxatives with different mechanisms could be utilized in this circumstance, which include softeners (e.g., Dioctyl), bulkers (e.g., ispaghula husk), and osmotics (e.g., polyethylene glycol, lactulose). On the other hand, laxatives working as stimulants (e.g., senna, bisacodyl) should only be taken prior to planned evacuation, since it could increase bowel activity, which will result in the movement of stool into the sigmoid colon and rectum.

A more convenient method for patients to control the time of evacuation is rectal stimulants. At present, the most commonly used local stimulants are glycerol suppositories, which are mild and easy use. Enema could also be utilized as a more aggressive method, by delivering a certain volume of fluid, with or without a drug, to the rectum. A tap water enema of 500 ml combined with fecal softeners and bulking agents is the most effective method of bowel management [22].

Another method to help with constipation is abdominal massage. On a theoretical basis, a noticeable response has been found in the rectum and anus in most recent physiological studies [23], where massage is applied to the abdomen following the supposed location of the colon in a clockwise direction. The effective rate of massage varies from 22 to 30% according to previous reports [24].

The most commonly used intervention is digital evacuation of feces, where patients themselves or their health-care providers may insert a gloved, lubricated finger into the rectum, in order to break up or remove stool. This manual evacuation is suggested to remove stool from rectum in a short period of time after surgery, in order to avoid over-distension with consequent damage to later reflex rectal function. A shorter duration of bowel care and fewer episodes of fecal incontinence is reported with the help of manual evacuation. For patients with long-term bowel evacuation problem, it is also a reasonable choice to maintain normal bowel habit.

35.2.2.2 Surgical Intervention

The last resort to deal with neurogenic bowel dysfunction is percutaneous colostomy. It can also be used as a salvage treatment after the failure of bowel rehabilitation. As colostomy significantly brings down the time spent and increases patients' independence on bowel management, it is reported to improve the quality of life for certain individuals [25, 26]. As most surgical procedures, although colostomy itself is considered safe and leads to positive outcome in most cases, stomas have several complications, such as postoperative intestinal obstruction, hernia, paralytic ileus, and skin rashes around the stoma, which may lead to failure of stool collection. Besides, after colostomy, the effusion of mucous from the remaining rectum or anus can also be problematic, calling for the additional use of pads. Apart from the surgical procedure itself, education of postsurgical stoma care for patients and their families is important to minimize or avoid these complications [25, 27].

35.3 Rehabilitation After Pelvic Resection

For patients with primary malignant tumors of pelvic region, surgical resection is the most commonly used local treatment, including the traditional hemipelvectomy and pelvic resection followed by functional reconstruction in the last few decades. Limb-salvage procedure refers to pelvic resection followed by functional reconstruction. According to the most recent literature, the oncological outcome of limb-salvage procedure is not inferior to hemipelvectomy with proper patient selection, in both long-term survival rate and local recurrence-free rate [28, 29]. More than 90% patients with primary malignant disease are evaluated as proper candidates for limb-salvage surgery [29]. Given the complicated structure of pelvic ring and the extensive resection of pelvic tumor to get a clean surgical margin, loss of osseous or ligament connection, instability of bony structure, and incomplete pelvic ring often occur as a result of wide resection. Reconstruction after resection is required to avoid severe disability in these cases [30]. However, guidelines for the rehabilitation after complicated resection and reconstruction have yet to be formally established.

For rehabilitation after pelvic surgery, there are two major goals. One is the healing of both abdominal and pelvic muscles, repair with minimal tension, and stability of the pelvic girdle. The other is to maintain or re-achieve normal joint functions, such as knee and ankle functions, to make up and minimize the loss of function of hip joint. Thus, rehabilitation procedures should be distinguished in different anatomic locations.

35.3.1 Type I Resection of Ilium

In type I pelvic resection, only part of the iliac bone is resected. Active abductor muscles strengthening, as well as muscle strengthening and ROM of knee and ankle could be initiated three days after surgery. Before abductor strength is regained, patients can use a cane to help with walking. Because the integrity of pelvic ring is reserved after type I resection, we recommended early ground exercise (usually 3–5 days after surgery) with full weight-bearing. A lumbarsacral corset should be considered in the first 6 weeks, if a large part of the iliac is resected and screw-rod system is used (Fig. 35.5), especially during sitting and ambulation to decrease the tension of lumbar and pelvic girdle.

35.3.2 Type II Resection of Acetabulum with or Without Pubis

Type II pelvic resection is the resection of the acetabulum, often followed by endoprosthetic or allograft reconstruction

Fig. 35.5 Screw-rod system after type I resection

Fig. 35.6 Endoprosthetic reconstruction after type II resection

(Fig. 35.6). In most cases, abductor muscles are reconstructed during surgery. In the first 6 weeks, keeping the ipsilateral limb suspended in flexion (30°) and abduction (30°) using corrective shoes are vitally important. Squat, tailor sitting, or sitting on a low stool should always be forbidden, no matter how long after surgery to prevent hip dislocation.

Quadriceps sets and straight leg rising in supine position can be taken 3 days after surgery. Voluntary knee and ankle motion exercises of the ipsilateral limb are encouraged in lateral position, with the immobilization of hip joint. Commonly speaking, abductors maintain the pelvis level during the stance phase, and then prevent titling of the contralateral hip during the following swing phase. Based on this, progressive hip abductor strengthening is suggested from the beginning of the second week. This issue could be initialed by concentric hip abduction while patients keep supine position, and later through isometric hip abduction to go against resistance. As the formation of obvious scar adhesion usually takes 6 weeks, active strengthening exercises of the abductors and flexors related to the hip joint can be started 6 weeks after surgery.

Toe touch weight-bearing (known as TTWB) or partial weight-bearing (named PWB) as tolerated can be attempted 2 weeks after surgery. Non-weight-bearing (NWB) itself may actually call for greater muscle forces in order to maintain the normal pelvic position and finally give more pressures over the hip joint. Considering this, TTWB is better than NWB as initial weight-bearing exercise. However, in patients with femoral head autograft or fibular autograft, the time of ground exercise should be postponed to as long as 6 weeks. External hip joint stabilizer is needed in ground exercise.

Various assistive devices could be utilized to provide support and balance as well as to unload the operated joint in acute phase. Generally speaking, walkers are the first choice, which will unload the affected pelvis and provide stability. Since both the hands are required to use walkers and lots of daily activities will be challenging as a result, most patients will easily evolve from walkers to crutches or canes. Compared to walkers and canes, axillary and forearm crutches allow faster gait and yield better energy efficiency; however, they offer the worst stability and are not suitable for older or more fragile patients with weak muscle strengths who are undergoing full weight-bearing.

A good quality of life and a better functional outcome is our goal for rehabilitation. The utilization and strict adherence to a properly designed, anatomically specific, individualized rehabilitation program can improve the functional result as well as quality of life after surgery. For malignant tumors, oncologic principles often dictate sacrifice of healthy tissues. However, we should always keep in mind that a well-designed, detailed resection and reconstruction to preserve nerve structure, osseous support, and soft-tissue protection as much as possible is the most important thing to achieve the best functional result. A great rehabilitation program could not be considered as a substitute for proper preservation.

It should be noted that in the procedure of rehabilitation, additional challenges could be found in patients who underwent other surgeries (e.g., lung metastasectomy) at the same time or following surgical complications. Lack of compliance can also be found in patients who suffer from fatigue brought by concurrent adjuvant chemotherapy or radiotherapy.

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