Eur Spine J (2015) 24:1778–1785
DOI 10.1007/s00586-015-3869-2

ORIGINAL ARTICLE

Surgical treatment options for aggressive osteoblastoma in the mobile spine

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Received: 15 September 2014 / Revised: 11 February 2015 / Accepted: 8 March 2015 / Published online: 7 April 2015

Abstract
Purpose Osteoblastoma (OBL) is a benign bone tumor with considerable recurrence potential. Resection is the mainstay for the treatment of Enneking stage 3 (st. 3) OBL. This retrospective study aimed to verify the appropriate surgical strategy for st. 3 lesions in the mobile spine.
Methods 19 cases of st. 3 OBL was diagnosed between 2001 and 2011. Clinical, radiological, surgical, and follow-up data were analyzed. This series included 14 men and 5 women and the mean age at diagnosis was 30.4 years. The lesions were located in the cervical spine in ten cases, in the thoracic spine in eight, and in the lumbar spine in one. Fourteen patients were surgically treated for the first time (intact cases), and five were referred to us after previous unsuccessful treatments (non-intact cases).
Results Before 2008, 11 patients underwent curettage followed by radiotherapy, including 5 non-intact cases and 6 intact cases. Tumor recurrence was observed in all five non-intact cases and four of the six intact cases. After 2008, eight intact patients underwent intralesional vertebrectomy, including six who underwent piecemeal total vertebrectomy and two who underwent intralesional en bloc vertebrectomy. All the eight patients had embolization before surgery. 18 patients had an average 67.3 months (range 36–148 months) of follow-up. Recurrence was not observed in any of these eight cases with intralesional vertebrectomy.
Conclusion Intralesional total vertebrectomy might be an appropriate choice for intact st. 3 OBL spine lesions. More cases with longer follow-up periods should be recruited in the future to better understand the treatment options available for this disease.

Keywords Osteoblastoma · Spine · Resection · Treatment

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Introduction

Osteoblastoma (OBL) is a benign bone tumor with considerable recurrence potential. By definition, the lesion size at diagnosis is above 2 cm [1]. Enneking stage 3 (st. 3) tumors are classified as rapidly growing lesions, with invasive extracapsular or extracompartmental extensions and rarely with metastases [2].

Resection is the standard treatment option for OBL, but its clinical outcome varies according to the surgical intervention. Local recurrence after surgery, especially subtotal resection, has been reported to be approximately 50 % in st. 3 spine lesions [3]. However, regression after subtotal removal has also been reported by Lucas et al. (3/59, from Mayo Clinic) [4] and by Boriani et al. (1/22, from Rizzoli Orthopedic Institute) [3]. Malignant transformation of OBL is rare and usually occurs after surgery. In the Mayo Clinic's report, only 2 of 306 cases demonstrated malignant transformation [4].

Boriani et al. were the first to systematically use the Enneking grade system to evaluate OBL lesions in the spine, and reported 40 st. 3 cases that were surgically treated between 1984 and 2010. They suggested wide/marginal en bloc resection for treatment of these st. 3 lesions [3], although other authors have reported good results with intralesional excision [5–7]. Wide/marginal en bloc resection could minimize the recurrence rate, but it is quite aggressive, with a mortality rate up to 7.7 % [8, 9]. Moreover, for cervical OBL, the decision of sacrificing the vertebral artery to achieve wide en bloc resection is controversial.

The Global Spine Tumor Study Group (GSTSG) described three different techniques for aggressive spine tumor resection (total vertebrectomy, Fig. 1) [10], namely, piecemeal total vertebrectomy, intralesional en bloc total vertebrectomy (e.g., Tomita's technique [11]), and wide en bloc total vertebrectomy (e.g., Boriani's technique [12]). Among them, wide en bloc total vertebrectomy is the most challenging.

It is accepted that OBL involving the central neuraxis is usually associated with greater recurrence rate [4]; however, according to data from the Massachusetts General Hospital, the recurrence rate was 11 % for vertebral OBL lesions, which was lower than that reported for extremity lesions [13].

We retrospectively reviewed 19 cases of OBL in the mobile spine diagnosed over the past 11 years in our hospital, focusing mainly on the treatment choices for st. 3 OBL.

Materials and methods

Nineteen consecutive cases with st. 3 OBL in the mobile spine were surgically treated in our department between 2001 and 2011 (Table 1). Most patients diagnosed before

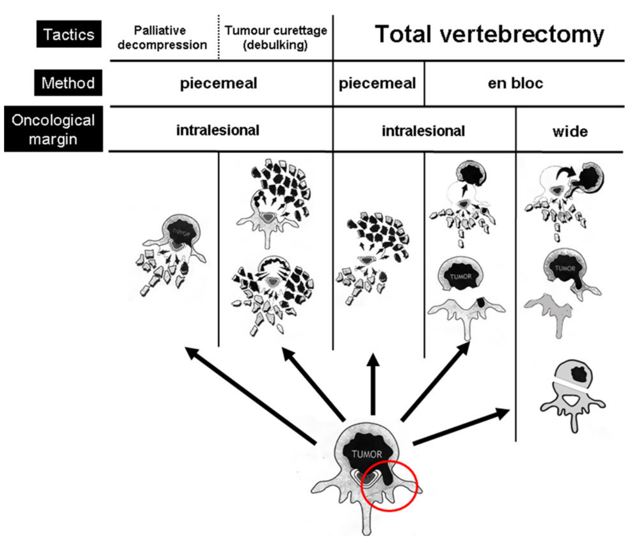

Fig. 1 Classification of surgical strategies, as determined by GSTSG. Total vertebrectomy includes three techniques, namely, piecemeal vertebrectomy, intralesional en bloc total vertebrectomy, and wide en bloc total vertebrectomy (with permission from Mr. David Choi, National Hospital for Neurology and Neurosurgery, London, UK)

2001 were lost to follow-up because of frequent changes in their addresses and telephone numbers following major city renovations in China. Our hospital's institutional review board approved this study.

A retrospective review of hospital charts, operating room reports, office charts, and radiographs was performed. The data collected included patient age, gender, radiological features, pathology, treatment method, outcome, and complications of biopsy and treatment.

Imaging and biopsy

We routinely performed posteroanterior and lateral spinal radiography, computed tomography (CT), and magnetic resonance imaging (MRI). Our radiologists carried out percutaneous CT-guided trocar biopsy for suspected malignant cases.

St. 3 OBLs are fully lytic lesions, eroding part of the cortex and extending into the canal and/or the surrounding soft tissues [3] (Figs. 2, 3). These tumors demonstrate bony shells with ill-defined margins on CT, confirming their aggressiveness. The clinical course is usually more rapid, with early manifestation of possible neurological symptoms [3].

Treatment protocol

Before 2008, extensive curettage through the tumor lesion was our standard treatment (Fig. 3). After 2008, owing to the high recurrence rate observed after curettage, we adopted total vertebrectomy. For cervical lesions, piecemeal total vertebrectomy was used to preserve the vertebral artery when possible, by combined posterior and anterior procedure for lesions extending into the vertebral body (Fig. 4). Total En bloc vertebrectomy was indicated for thoracolumbar lesions; however, we performed intralesional total vertebrectomy for those lesions that involved the pedicle(s) (Fig. 5, the Tomita's technique, and total en bloc spondylectomy [11]). Internal fixation and fusion were used to reconstruct spinal stability if needed.

Technical note

Traditional curettage is difficult to perform in cases of aggressive spine OBL lesions [14]. In the presence of cortical destruction, the reactive bony shell is too thin to observe on CT and to visualize during the operation. The reactive fibrotic tissue around the lesions is usually quite stiff, making its intraoperative distinction from the thin bony shell of the tumor difficult. Therefore, simple intralesional curettage might become either a debulking procedure or subtotal resection, which often leads to recurrence. During piecemeal total vertebrectomy, the most

Table 1 Treatment and outcomes of 19 patients with Enneking stage 3 (st. 3) osteoblastoma of the mobile spine

No.	Gender	Age	Prior surgery	Location	Fracture	Frankel scale	Surgery	Follow-up months	Last status
1	M	16	No	C4–5 PE	No	D	Curettage + Rt	148	NED
2	M	24	No	T5–6 PE	No	D	Curettage + Rt	131	SWD
3	M	17	No	T10 PE	No	E	Curettage + Rt	121	NED
4	M	48	No	C4 PE + VB	Yes	D	Curettage + Rt	110	SWD
5	M	27	Yes	T6–8 PE + VB	No	A	Curettage + Rt	–	Lost
6	F	36	Yes	T5–6 PE	No	E	Curettage + Rt	47	DOD
7	M	47	No	C3 PE	No	E	Curettage + Rt	75	DOD
8	M	16	Yes	C4–6 PE + VB	Yes	E	Curettage + Rt	75	SWD
9	M	39	Yes	T3–4 PE + VB	No	E	Curettage + Rt	17	DOD
10	M	16	No	C2 PE + VB	Yes	E	Curettage + Rt	63	SWD
11*	F	26	Yes	T7–11 PE + VB	No	A	Curettage + Rt	14	DOD, MT
12	M	38	No	T2–3 PE + VB	No	E	PV	61	NED
13	M	24	No	C2–3 PE	No	E	PV	60	NED
14	M	30	No	C7 PE	No	E	PV	56	NED
15*	F	55	No	T5 PE + VB	Yes	E	IEV	52	NED
16	F	47	No	C4-5 PE	No	E	PV	48	NED
17	F	34	No	L1 PE + VB	No	E	IEV	40	NED
18	M	23	No	C3 PE	No	E	PV	36	NED
19	M	14	No	C7 PE + VB	No	E	PV	36	NED

M male, *F* female, *PE* posterior element, *VB* vertebral body, *PV* piecemeal vertebrectomy, *IER* intralesional en bloc resection, *NED* no evidence of disease, *SWD* survival with disease, *DOD* dead of disease, *MT* malignant transformation

* This patient did not have regular follow-up as suggested

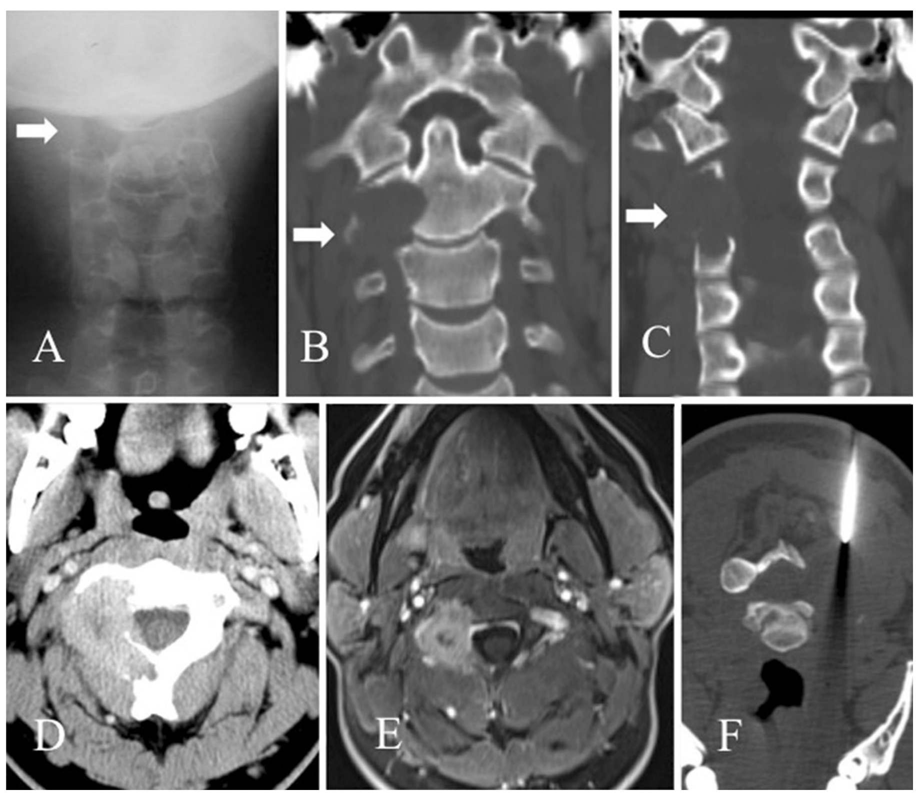

Fig. 2 Case 13. A 24-year-old male patient who developed a C2/3 lesion and experienced neck pain for 3 months. **a**–**d** Posteroanterior radiograph and sagittal and axial CT scans showing a fully lytic lesion eroding the cortex of the C2 vertebral body on the *right* and the *right* lateral C2/3 mass. A bony shell, atypical radiological feature of OBL, could not be identified. **e** Axial MRI with contrast. **f** Percutaneous trocar biopsy was carried out under CT guidance for this suspected malignant lesion, which was confirmed as OBL

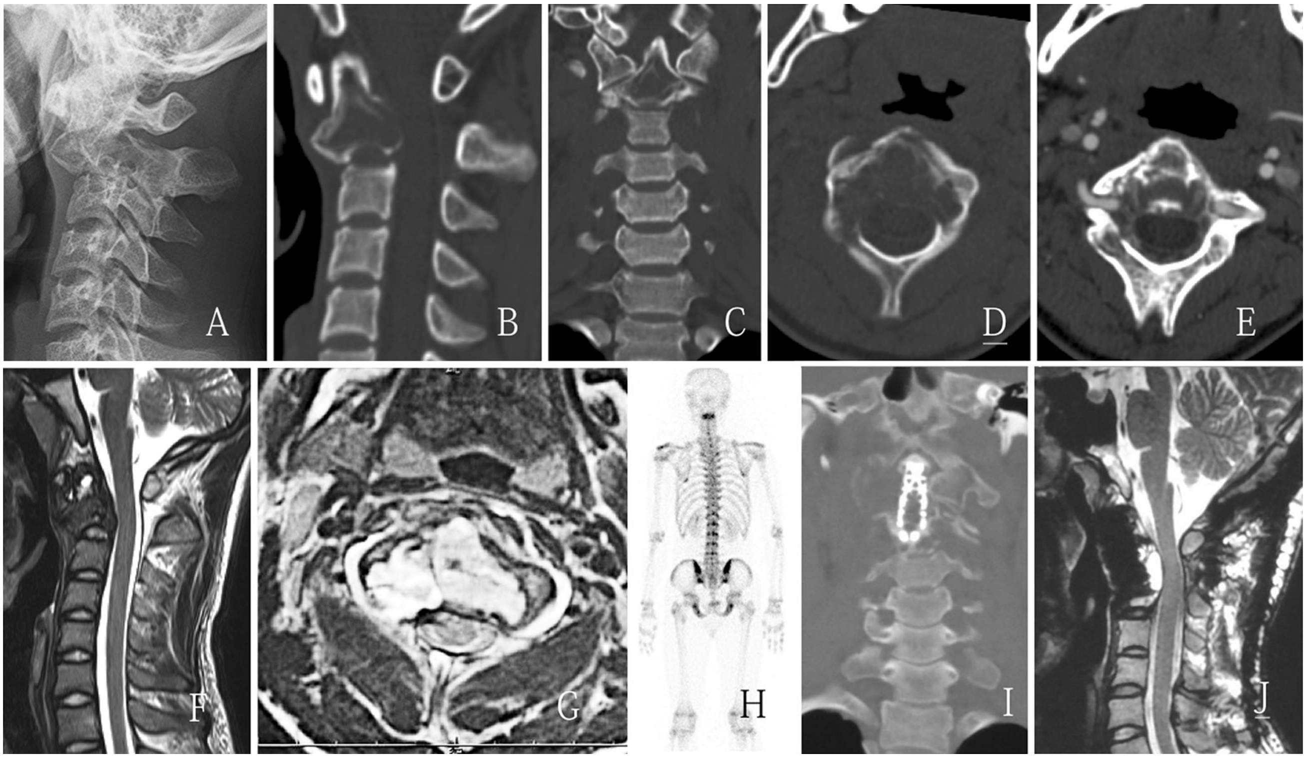

Fig. 3 Case 10. A 16-year-old female patient experienced severe neck pain for 1.5 months. **a** Lateral radiograph showing a pathological C2 fracture. **b–d** Sagittal, coronal, and axial CT scans showing a lytic lesion eroding the cortex of the vertebral body and C2 pedicles. **e** Contrast CT scan showing bilateral vertebral arteries. **f**, **g** Sagittal and axial T2-weighted MRI scan showing hybrid signal intensity. **h** Bone scan. **i**, **j** Coronal CT and sagittal MRI scans obtained 18 months after transoral curettage followed by radiotherapy showing recurrence around the cage

Fig. 4 Case 14. A 30-year-old male patient experienced severe neck pain and radiculopathy for 4 months. **a** Posterioanterior radiography. **b**, **c** Coronal and axial CT scans showing a lytic lesion of the right lateral mass, pedicle, and transverse process of C7, along with cortical expansion and extension into the spinal canal and paravertebral soft tissue. **d** Fat-suppressed axial MRI scans showing marked reactive tissue surrounding the lesion. The patient underwent combined posterior and anterior approaches with intralesional sagittal vertebrectomy. **e**, **f** Radiograph and axial CT at 4 years follow-up

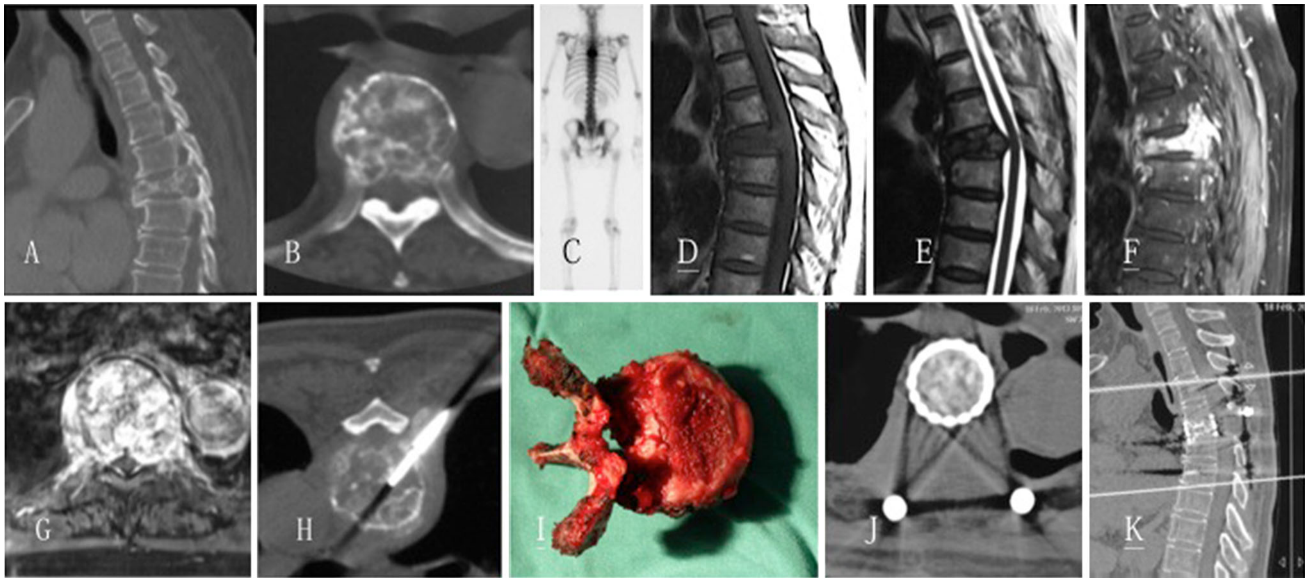

Fig. 5 Case 15. A 55-year-old female patient experienced severe back pain for 4 months. **a**, **b** Sagittal and axial CT scans showing a lytic lesion eroding the cortex of the T5 vertebral body and pedicles with paravertebral extension and pathological fracture. **c** Bone scan. **d**, **f** Sagittal T1, T2, and fat-suppressed MRI scan. **g** Axial MRI scan showing a lesion that extended into the canal and paravertebral space. **h** CT-guided biopsy. The patient underwent intralesional en bloc vertebrectomy and reconstruction (Tomita's technique). **i** Photograph of the specimen. **j**, **k** Axial and sagittal CT scan at 3 years follow-up

important step is the thorough exposure of the lesion around the reactive fibrotic tissue (Fig. 4), followed by tumor removal using the piecemeal technique. After this procedure, only the vital structures (dural mater and/or vertebral artery) and the healthy muscles are left. Thus, the incidence subtotal tumor resection could be minimized.

After 2008, embolization was routinely carried out 24 h prior to the scheduled operation to minimize blood loss. Adjuvant radiotherapy is usually suggested after curettage, except for patients who have undergone intralesional total vertebrectomy (piecemeal or en bloc). All these cases were done by our two senior surgeons (ZJL and XGL).

Follow-up

We obtained roentgenograms, CT, and MRI scans every 3 months in the first 2 years, and then every 6 months for the following 3 years, and annually thereafter. If the patient displayed symptoms indicative of tumor recurrence, immediate CT and MRI scans were requested. If suspected recurrence could be neither confirmed nor ruled out, bone scintigraphy and CT scans with contrast were used.

Results

This series included 14 men and 5 women (male to female ratio, 2.8:1). The mean age at diagnosis was 30.4 years (range 14–55 years), with 11 patients aged (57.9 %) under 30 years (Table 1). The lesions were located in the cervical spine in ten cases, in the thoracic spine in eight, and in the lumbar spine in one. 14 patients were surgically treated for the first time (intact cases) and 5 were referred to us after previous unsuccessful treatments (non-intact cases), of which 2 previously underwent one surgery, 1 underwent two (Case 8), 1 underwent four (Case 5), and 1 (Case 11) underwent five.

The mean duration of symptoms before presentation was 13.4 months (range 1–60 months). All 19 patients presented with back pain and 7 of them experienced night pain. Mean visual analog scale score at admission was 4.5 ± 2.8 (range 2–9). Nine patients received non-steroidal anti-inflammatory drugs, and four had partial pain relief. Neurological deficit was documented in ten cases: five with radiculopathy and five with myelopathy [including two cases with Frankel grade A (Case 5 and 11) and three with Frankel grade D]. No apparent scoliosis and/or torticollis (Cobb's angle $> 10°$) was documented. A local palpable mass with slight tenderness was observed in one patient.

Three lesions were mainly located in the vertebral body and extended to the pedicel(s); posterior arch involvement was observed in 16 cases with lesion extension into the vertebral body. Unilateral and bilateral pedicle involvement was observed in 14 (Figs. 2, 4, 6) and 5 cases (Figs. 3, 5), respectively. In the ten patients with cervical lesions, unilateral and bilateral vertebral arteries were affected in seven and three patients, respectively. The vertebral lesions extended into the spinal canal in 15 cases and/or into the paravertebral space in 12 cases, with ten tumors involving both the spinal canal and paravertebral space (Figs. 2, 3, 4, 5, 6). Four patients presented with pathological fracture (Figs. 3, 5).

All the 13 intact cases underwent CT-guided needle biopsy. The correct pathological diagnosis was achieved in 11 patients (84.6 %), whereas 1 was misdiagnosed as a cartilage tumor and in the other only necrotic tissue and osteoclast-like giant cells were identified. All the postoperative pathology diagnoses were OBL.

Before 2008, 11 patients underwent extensive curettage with/without postoperative radiotherapy (40–45 Gy), including 5 patients with non-intact lesions. After 2008, eight patients with intact cases underwent vertebrectomy, and six and two of them underwent piecemeal sagittal vertebrectomy (Figs. 4, 6) and intralesional en bloc vertebrectomy (Fig. 5), respectively.

One patient with recurrence after four previous T6–8 curettages was treated with intralesional excision. Owing to the fact that embolizations were not available in our hospital at that time, this surgery had to be aborted due to extensive bleeding (12,000 ml, Case 5). For the other 18 cases, the average operative time was 371 min (148–525 min) and the average intraoperative blood loss was 3076 ml (200–4000 ml).

After surgery, one patient had cerebrospinal fluid leakage and two experienced pleural effusion, but none of these complications required surgical intervention. No instrument failure was observed at the last follow-up. Three patients showed neurological improvement from preoperative Frankel grade D to postoperative Grade E, while the other two remained at Frankel grade A.

Eighteen patients (94.7 %) had an average 67.3 months (range 36–148 months) of follow-up. The only exception was the patient affected by extensive bleeding. At the last follow-up, 14 patients were alive, including 10 with no evidence of disease and 4 surviving with tumors.

Among the ten patients treated with intralesional curettage and radiotherapy, four patients with non-intact lesions developed recurrence, and four of the six patients with intact lesions also showed recurrence. One had malignant transformation after five surgeries and died due to extensive metastatic disease (Case 11). Three others had massive recurrent upper cervical/thoracic tumor masses, but refused to undergo further treatment and died due to respiratory failure. None of the eight patients who underwent vertebrectomy developed recurrences.

Unfortunately, some patients did not have appropriate insurance to afford the frequent MRI and CT examinations

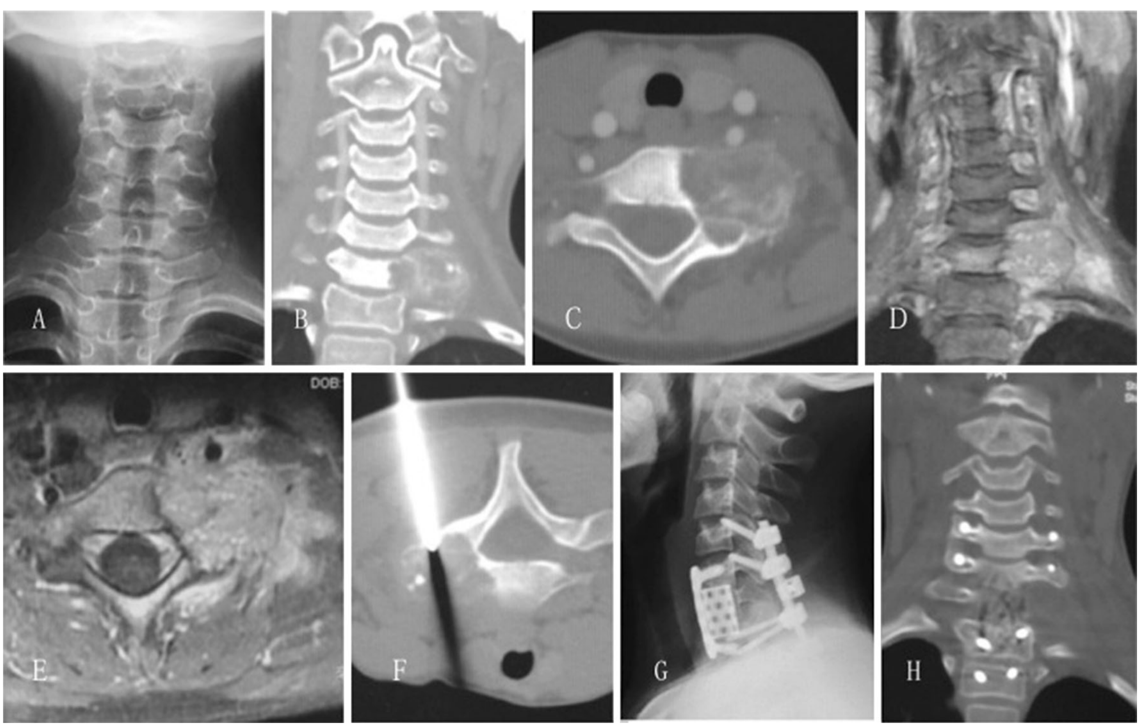

Fig. 6 Case 19. A 14-year-old male patient experienced neck and night pain for 6 months. **a** Posterioanterior radiography. **b**, **c** Coronal and axial contrast CT scans showing a lytic lesion of the left lateral mass, pedicle, and transverse process of C7. **d**, **e** Coronal and axial MRI scans showing marked reactive tissue surrounding the lesion. **f** CT-guided biopsy. The patient underwent a combined posterior and anterior approach with piecemeal sagittal vertebrectomy. **g**, **h** Radiograph and coronal CT at 2 years follow-up

during follow-up; therefore, the accurate recurrence time could not be clearly documented.

Discussion

The treatment of aggressive OBL in the mobile spine is still controversial and limited data are present in the literature. This is attributable to several reasons. First, OBLs are relatively uncommon; therefore, most reports are case reports. Second, the large series analyzed so far usually included mixed types of lesions [7], which were not appropriately documented according to the Enneking staging system. In 1990, Nemoto et al. [15] reviewed 75 spinal OBL cases, although they mainly focused their analysis on the clinical and radiological presentations. In 1994, Lucas et al. [3] reported 306 OBL cases, including 75 treated in the Mayo Clinic. Tumors located in the spine accounted for 32 % (66/306) of the lesions, and the overall recurrence rate was 16 %. In 2008, Berry et al. [13] studied 99 cases from a 30-year database of the Massachusetts General Hospital (MGH) and reported a recurrence rate of 11 % for 28 spine lesions. Unfortunately, these two large OBL studies did not provide details of staging and treatment choices for the spine lesions.

Radiological presentation of OBL lesions can vary. Typically, a dense shell of bone surrounds st. 2 OBL lesions. In st. 3 lesions, the bony shell tends to be very thin with expansion into the adjacent soft issues [13, 16, 17]. As reported by Lucas et al. [4], cortical expansion and destruction were common radiographic findings (39 %), and 12 % of cases had features suggestive of malignancy.

Grossly, the area surrounding the primary tumor can be very dense (st. 2) or, alternatively, quite lytic (st. 3) [18]. In typical st. 2 cases, OBL is well circumscribed and surrounded by a clear reactive bone shell with mineralization; whereas st. 3 lesions usually do not have a clear bony shell with surrounding reactive stiff fibrous tissue [13]. In these circumstances, incomplete removal might occur during intralesional curettage.

Local recurrence rate varied among different anatomic sites. According to the MGH database, OBLs recurred in 11 % of patients with spine lesions, 23 % with tibial tumors, 33 % with humeral tumors, and 43 % with femoral tumors [13].

Recurrence of OBL after total resection is rare, but can vary between 10 and 23 % after incomplete removal [13]. Compared to en bloc resection, curettage has lower morbidity, but might leave behind microscopic disease, which can cause recurrence. In the 99 OBL cases reported by the MGH, 23 % of patients who were treated with curettage required further surgery for recurrence, compared to 14 % of patients who underwent en bloc resection [13]. Data from the Mayo Clinic showed a recurrence rate of 19 % (10/52) after intralesional curettage, compared to the 8.7 % (2/23) reported after en bloc resection with marginal and wide margins [4].

Boriani and his colleagues reviewed 40 st. 3 cases that were treated over 26 years between 1984 and 2010. For the intact cases, the incidence of recurrence was 0 % (0/10) after wide/marginal en bloc resection and 7 % (2/28) after intralesional excision with or without radiotherapy. For the non-intact group, the recurrence rate was much higher: 67 % (2/3) after en bloc resection and 75 % (3/4) after intralesional excision [3].

For the recurrent OBL cases, en bloc resection is usually recommended [4], although Boriani et al. [3] successfully treated six patients with recurrent lesions with further intralesional excision.

We observed high recurrence rates with curettage, as reported by Boriani et al. The main reason for high recurrence is subtotal removal. As mentioned above, the margin is usually not clear in st. 3 OBL lesions, unlike st. 3 giant cell tumors that demonstrate clear pseudo-capsules [14]. MRI may be useful in the evaluation of extraosseous inflammatory reactions associated with st. 3 OBL.

With intralesional excision, we achieved good results in eight cases with more than 2 years follow-up. Other authors have also reported similar results [5–7]. In our hospital, preoperative embolization is suggested to reduce intraoperative bleeding and to achieve ideal visualization. Moreover, to minimize the risk of microscopic tumor residues, our procedure involves exposure of the whole lesion through the normal tissue and removal of the lesion and the reactive tissue by piecemeal vertebrectomy or intralesional en bloc vertebrectomy.

Wide/marginal en bloc vertebrectomy could definitely reduce the recurrence rate, but is more technically demanding and associated with higher morbidity and mortality [8, 9]. OBL itself has neither malignant nor metastatic potential. Histologically, the lesion has benign features, even when, radiographically, it appears aggressive [19]. On the basis of our data and those from others, we suggest intralesional total vertebrectomy for st. 3 OBL lesions for patients with no prior surgical history and wide/marginal en bloc vertebrectomy for recurrent OBL cases.

Our study has some intrinsic shortcomings. It is a retrospective study and only included 19 cases treated in a short period of time. More cases should be recruited with longer follow-up in the future to better understand the treatment choices for this disease. It is important to note that the terminology for both curettage and vertebrectomy in spine tumor surgery is still not standardized. Choi et al. [10] used “vertebrectomy”, while Boriani et al. [3] preferred “resection” in slightly different contexts. For some orthopedic oncologists, "intralesional total vertebrectomy" is still a kind of "curettage." The "modified curettage" used for aggressive benign bone lesions, such as giant cell tumors of bones located in the extremities, is performed through a wide cortical window followed by high-speed burring with or without adjuvant treatments [14].

In conclusion, intralesional total vertebrectomy might be an appropriate choice for intact st. 3 OBL spine lesions; however, to validate these findings, more cases should be recruited with longer follow-up in the future.

Conflict of interest The authors declare that there is no actual or potential conflict of interest in relation to this article.

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