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Skeletal Radiology

Case report 566

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Clinical information

This 15-year-old male presented with a 6 month history of a limp and a one month history of urinary retention with overflow incontinence. Physical examination revealed generalized muscle weakness in the right lower extremity and sensory loss involving the 2nd, 3rd and 4th sacral nerves on the right side. Cystography showed a hypotonic neurogenic bladder with overflow incontinence and significant post-void residual urine (1500 cc). EMG findings were consistent with right lumbosacral plexopathy.

Radiographs of the pelvis revealed a well-defined, round, lytic lesion in the inferior aspect of the right sacral ala, which lacked sclerotic margins and expanded the 3rd and 4th sacral neural foraminae (Fig. 1 A). The lateral view of the sacrum showed that the destructive process also involved the middle and posterior aspects of the sacrum (Fig. 1 B). Metallic clips from an attempted biopsy of the lesion at an outside hospital were noted anterior to the right sacral ala. That procedure had been terminated because of profuse hemorrhage and no diagnostic tissue was obtained.

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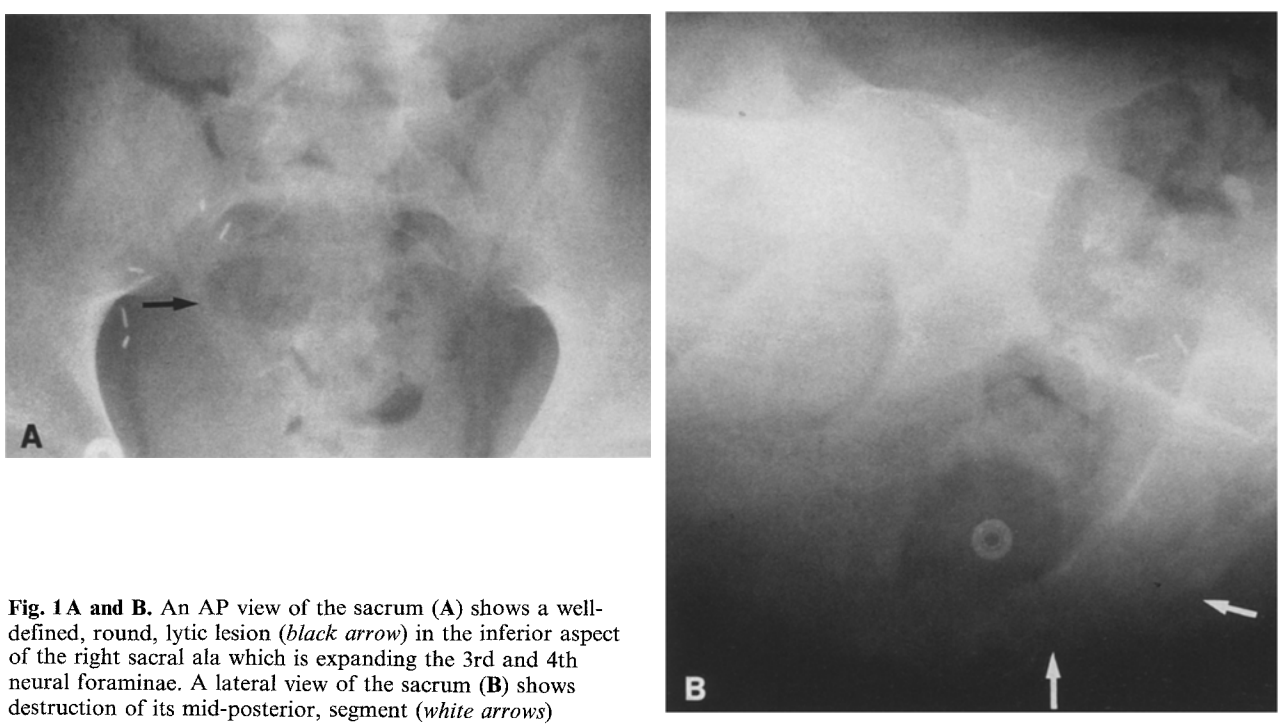

Fig. 1 A and B. An AP view of the sacrum (**A**) shows a well-defined, round, lytic lesion (*black arrow*) in the inferior aspect of the right sacral ala which is expanding the 3rd and 4th neural foraminae. A lateral view of the sacrum (**B**) shows destruction of its mid-posterior, segment (*white arrows*)

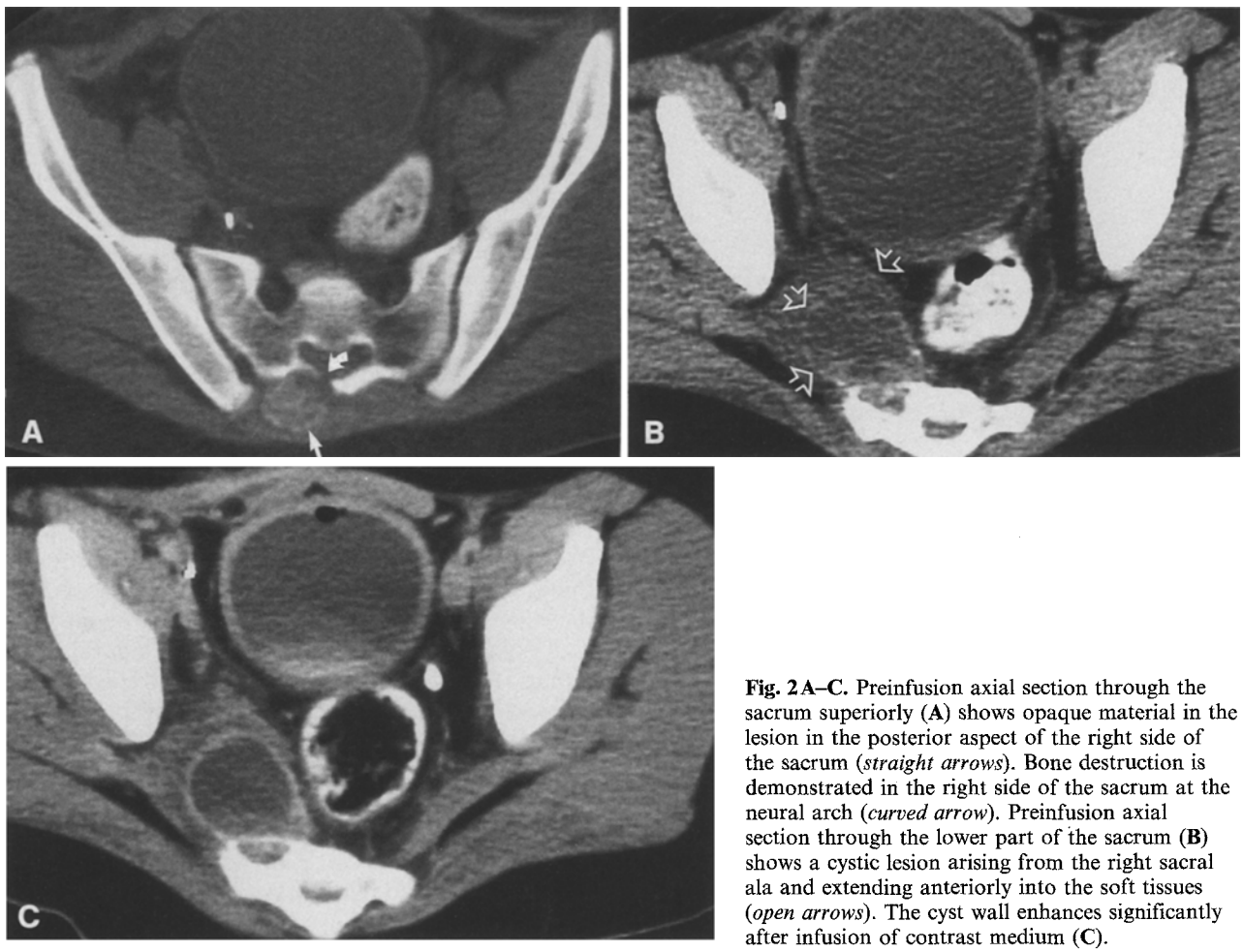

Fig. 2A–C. Preinfusion axial section through the sacrum superiorly (**A**) shows opaque material in the lesion in the posterior aspect of the right side of the sacrum (*straight arrows*). Bone destruction is demonstrated in the right side of the sacrum at the neural arch (*curved arrow*). Preinfusion axial section through the lower part of the sacrum (**B**) shows a cystic lesion arising from the right sacral ala and extending anteriorly into the soft tissues (*open arrows*). The cyst wall enhances significantly after infusion of contrast medium (**C**).

Transverse CT of the pelvis prior to contrast infusion demonstrated a destructive process in the right sacral ala which extended across the sacrum anteroposteriorly, crossed the midline to the left, and involved the sacroiliac notch inferiorly. Posteriorly and superiorly the lesion showed areas of high attenuation suggestive of bone formation (Fig. 2A). Anteriorly and inferiorly the lesion appeared cystic and contained a fluid-fluid level (Fig. 2B). The cystic portion of the lesion was expansile – anteriorly into the presacral soft tissues. CT scans obtained with contrast infusion revealed abnormal enhancement only around the cyst wall (Fig. 2C).

MRI of the pelvis was performed with a 1.5 T unit, using a spin-echo sequence with a TR of 2000 ms and TE of 20 ms and 70 ms. MR images of the lesion in the posterior and superior aspect of the sacrum demonstrate new bone formation (Fig. 3A) which was seen with CT (Fig. 2A). The cystic component of the lesion in the anterior part of the right of the sacrum had a fluid-fluid level with increased signal intensity of the supernatant fluid on both the first and second echo images (Fig. 3B and C). In addition, multiple, similar, but smaller, fluid-fluid levels were seen in the posterior aspect of the sacral lesion (Fig. 3C).

Based on the imaging findings, surgical biopsy was carried out, using a posterior approach to encounter the more solid portion of the tumor.

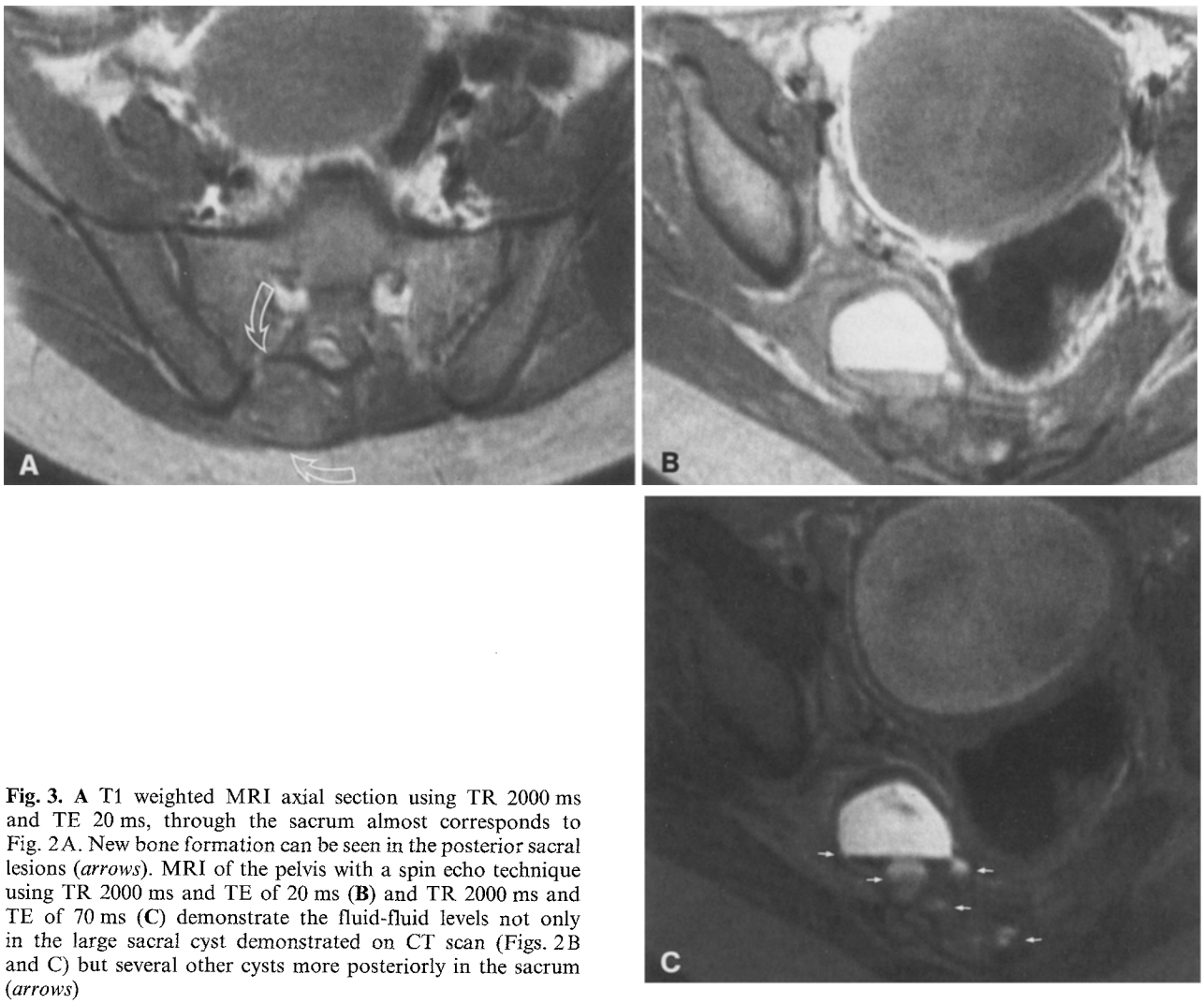

Fig. 3. A T1 weighted MRI axial section using TR 2000 ms and TE 20 ms, through the sacrum almost corresponds to Fig. 2A. New bone formation can be seen in the posterior sacral lesions (*arrows*). MRI of the pelvis with a spin echo technique using TR 2000 ms and TE of 20 ms (**B**) and TR 2000 ms and TE of 70 ms (**C**) demonstrate the fluid-fluid levels not only in the large sacral cyst demonstrated on CT scan (Figs. 2B and C) but several other cysts more posteriorly in the sacrum (*arrows*)

Diagnosis: Osteoblastoma of sacrum with secondary aneurysmal bone cyst

Friable hemorrhagic tissue was found overlying the sacrum and similar tissue was curretted from the right sacral ala. Histological studies revealed that some portions of the tumor consisted of multiple cystic spaces containing blood. The walls of the cysts were composed of proliferating granulation tissue and multinucleated giant cells, but did not have any lining (Fig. 4A). Other areas of the lesion showed cellular tumor (Fig. 4B). The cells were plump, ovoid to spindle-shaped, and had uniform vesicular nuclei. Abundant osteoid tissue was lined by osteoblasts. The supporting stroma was vascular. No pleomorphism or mitotic activity was noted (Fig. 4C). The histopathological diagnosis was benign osteoblastoma with secondary aneurysmal bone cyst.

Discussion

Aneurysmal bone cysts (ABC) may arise de novo in bone or may occur in various lesions such as giant cell tumor, chondroblastoma, chondromyxoid fibroma, fibrous dysplasia, chondrosarcoma and telangiectatic osteosarcoma [1].

Primary bone tumors of the spine are rare in children with the exception of ABC and benign osteoblastoma (BO) or osteoid osteoma [2]. BO comprises 1% of primary bone tumors and occurs in the posterior vertebral column, long bones and calvarium [3]. BO shows a predeliction for males between 3–30 years of age. Osteoblastomas are expanding masses that may sometimes resemble ABC. They are classified as cortical, spongious, periosteal, or multifocal, according to their sites of origin [4]. Cortical osteoblastoma, also referred to as osteoid osteoma is located in the compacta, where it evokes a striking perifocal sclerosis and rarely exceeds 1 cm in diameter. Spongious osteoblastomas are characteristically expanding and well circumscribed and evoke a variable amount of sclerosis. Periosteal osteoblastoma arises beneath the periosteum, but has not been described in the spine. In the entire spine, spongious osteoblastomas are seen twice as often as cortical osteoblastomas, but the two types are equal in frequency in the lumbar spine. BO involves the posterior elements of the spine. When the neural arch, rather than the posterior spinous processes alone is involved, spinal cord or nerve root compression may result. Epidural extension occurs in 49% of vertebral BO, and 54% of vertebral BO present with neurological symptoms [5]. Extension of epidural tumor must be sought in patients with radicular pain but may not always be seen on lumbar myelography [5]. BO shows a mixed sclerotic and osteolytic pattern more commonly than a purely lytic pattern, although the lytic component usually predominates. An extraosseous soft tissue mass outlined by a rim of calcification may be present. The lesion is usually well circumscribed, but the radiographic features may not be sufficiently distinctive to permit accurate identification. The differential diagnosis of BO in the spine in children includes ABC, Ewing's tumor, giant cell tumor and eosinophilic granuloma [6]. Osteoblastoma is usually progressive in size and in causing pain. Although relief of pain is achieved in 93% of patients with complete resection of BO, partial resection does not relieve pain [5]. Recurrence of tumor has occured as late as 12 years after resection. Patients with osteoblastoma should therefore be followed closely with attention to clinical behavior until it is believed that either local recurrence or cure [7, 8] has occurred.

Pathologically, the typical osteoblastoma is homogeneously friable and granular, varying in color from red to purple. All are well demarcated from surrounding sclerotic and normal bone. Microscopically, a prominent layer of osteoblasts is usually visible at the trabecular bone surfaces. Osteoclasts are present in fibrous tissue, sometimes eroding and remodeling the bone as evidenced by the presence of cement lines [9]. Although the histological material is of crucial diagnostic importance, the tissue must be studied thoroughly because osteoblastoma can be mistaken for telangiectatic osteosarcoma or ABC at low power microscopy [2, 6].

Aneurysmal bone cyst is a non-neoplastic lesion occurring in the first two decades of life and not infrequently arising in the spinal posterior elements. The lesion may regress even after incomplete resection. Rates of recurrence range from 10–20%. The treatment of choice is surgical resection of the entire lesion or as much of it as possible. Grossly, ABC consists of spaces filled with unclotted blood, old blood, or serous fluid. Microscopically, cavernous spaces with fibrous walls that lack normal features of blood vessels such as endothelium and elastic laminae of muscles are seen [10]. Areas of osteogenesis are also noted in ABC [10]. Adequate biopsy is therefore needed to avoid misinterpreting an area that resembles an ABC as a neoplasm [11].

A characteristic feature of ABC noted on CT and MRI scans is the presence of fluid-fluid levels [12, 13]. Giant cell tumor, chondroblastoma and telangiectatic osteosarcoma have been described as

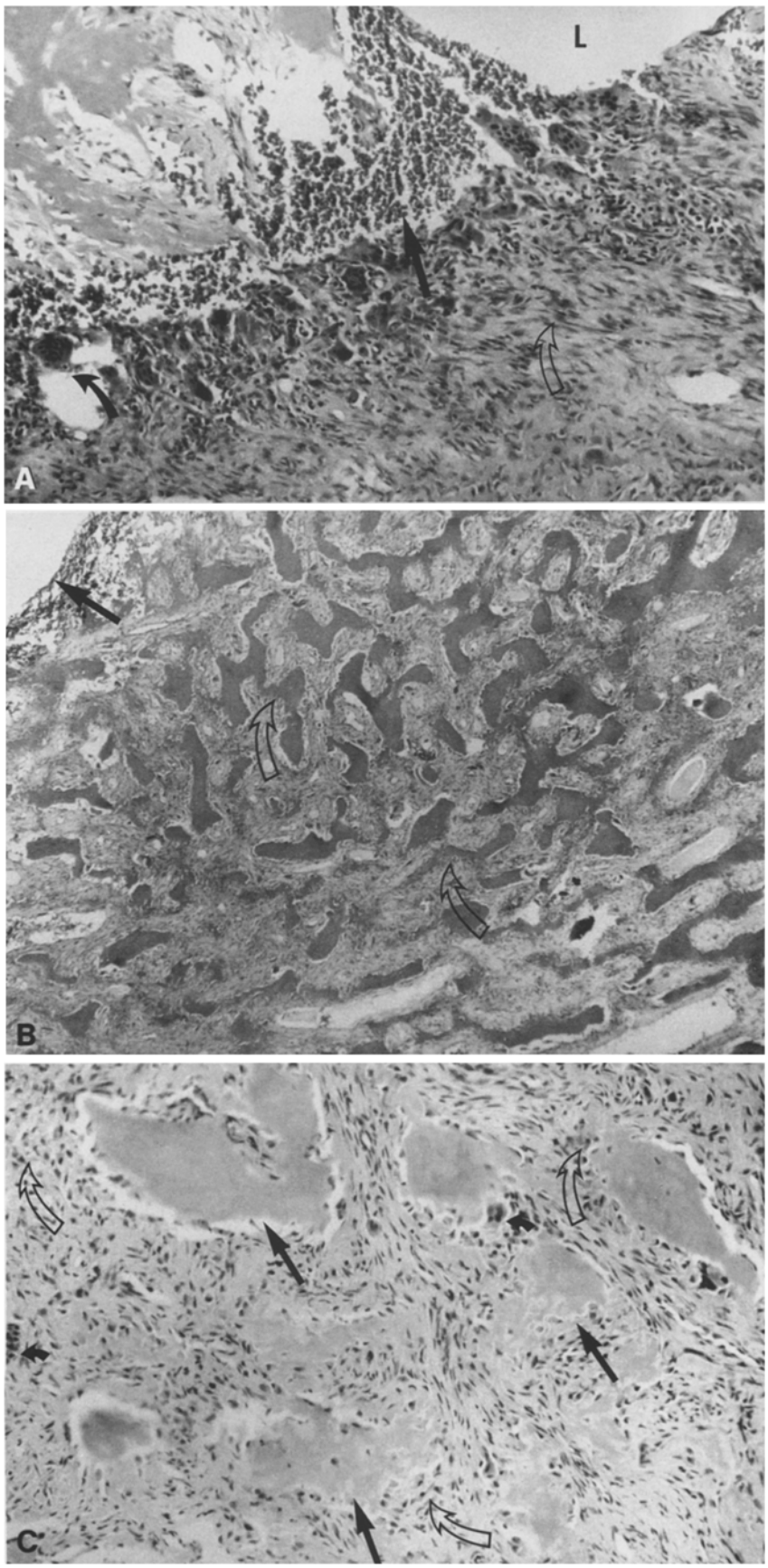

Fig. 4. **A** Photomicrograph (H&E ×100) of the wall of the aneurysmal bone cyst showing multinucleated giant cells (*curved black arrow*), fibroblasts (*curved open arrow*) and hemorrhage in the lumen (L).
B Photomicrograph (H&E ×25) of the osteoblastoma component showing heavy osteoid deposition (curved open arrows) and part of the cyst in the upper left corner (*black straight arrow*).
C Higher magnification of osteoblastoma component (H&E ×100) showing irregular osteoid (*straight black arrows*), giant cells (*curved black arrows*) and cellular stroma (*curved open arrows*)

having this feature [12]. Fluid-fluid levels in ABC represent sedimentation of red blood cells and serum in unclotted blood contained within the cysts [13]. Cortical expansion and thinning, cortical destruction, and soft tissue masses may be appreciated on CT and MRI. MRI is generally superior in depicting demarcation of the abnormality from adjacent soft tissue extension and CT is superior in demonstrating new bone formation and calcification [14]. Between the extraosseous mass and adjacent soft tissue planes, ABC has a thin cortical shell or a sharp interface representing calcified or noncalcified periosteal membrane. The thin cortical margin can be demonstrated by CT [12]. MRI may show the calcified periosteal membrane or cortical shell of the ABC as a rim of low signal intensity [15]. The cysts display a wide range of signal intensity due to intracystic hemorrhage of different ages [15]. Acute hemorrhage produces low signal intensity on T_1 images. After a few days, with oxidation of iron compounds and methemoglobin, the signal intensity increases greatly on T_1-weighted images. Even with T_2-weighted images there is still a significant T_1 effect causing a relatively high signal intensity. In our patient, the cystic lesions with fluid-fluid levels demonstrated supernatant fluid of high signal intensity on both T_1-weighted and T_2-weighted images. Normal marrow-filled vertebral bodies produce a homogeneous signal higher than that of the intervertebral disc on T_1-weighted images. In contrast, most tumors have long T_1, resulting in decreased signal intensity on partial saturation (TR 500 TE 40) or inversion recovery images (TR 200 TE 500). We did not obtain inversion recovery images in our patient. As the T_1 images were obtained only in the sagittal plane, the low signal intensity usually associated with tumor effect was not well seen. Most tumors also have a prolonged T_2 and typically show increased signal intensity with spin echoes that have long repeat times (TR 2000 TE 60).

In *summary*, benign osteoblastoma can resemble aneurysmal bone cyst both in location and in radiographic and histological appearance. The sacral lesion described in this case showed features of both BO and ABC on plain films, CT, MRI, and histological sections. CT demonstrated a combination of new bone formation and bone destruction in the posterior part of the lesion, suggesting a diagnosis of BO. Although these bone changes were not well appreciated by MRI, MRI showed multiple cysts with fluid-fluid levels in the same part of the sacrum which were characteristic of aneurysmal bone cysts and missed by CT. However, the long fluid-fluid level in the large anterior sacral ABC was well demonstrated by both CT and MRI. In our patient, CT and MRI proved to be complementary in the diagnosis of a secondary aneurysmal bone cyst engrafted onto the sacral benign osteoblastoma.

References

1. Bonakdanpour A, Levy WM, Aegerter E (1978) Primary and secondary aneurysmal bone cyst: A radiological study of 75 cases. Radiology 126:75–83
2. Dahlin DC (1978) Bone tumors: General aspects and data on 6221 cases, 3rd edn. Charles C Thomas Publisher, Springfield, Illinois, pp 370–376
3. Bloom MH, Bryan RC (1969) Benign osteoblastoma of the spine. Clin Orthop 65:157–162
4. De Souza Dias L, Frost HM (1973) Osteoblastoma of the spine. Clin Orthop 91:141–151
5. Janin Y, Epstein JA, Carras R, Khan A (1981) Osteoid osteomas and osteoblastomas of the spine. Neurosurgery 8:31–38
6. Azouz EM, Kozlowski K, Morton D, Sprague P, Zerhouri A, Asselah F (1986) Osteoidosteomas and osteoblastoma of the spine in children. Pediatr Radiol 16:25–31
7. Lichtenstein L, Sawyer WR (1964) Benign osteoblastoma: Further observations and report of twenty additional cases. J Bone Joint Surg (Am) 46:755–765
8. Epstein N, Benjamin V, Pinto R, Budzilovich G (1980) Benign osteoblastoma of a thoracic vertebra. J Neurosurg 53:710–713
9. Byers PD, McPath (1968) Solitary benign osteoblastic lesions of bone. Cancer 22:43–57
10. Dahlin DC, McLeod RA (1982) Aneurysmal bone cyst and other nonneoplastic condition. Skeletal Radiol 8:243–250
11. Zimmer WD, Berquist TH, Sim FH, Wold LE, Pritchard DJ, Shives TC, Mc Leod RA (1984) Magnetic resonance imaging of aneurysmal bone cyst. Mayo Clin Proc 59:633–636
12. Hudson TM (1984) Fluid levels in aneurysmal bone cysts: A CT feature. AJR 142:1001–1104
13. Hudson TM, Hanlin DJ, Fitzsimmons JR (1985) Magnetic resonance imaging of fluid levels in an aneurysmal bone cyst and in anticoagulated human blood. Skeletal Radiol 13:267–270
14. Zimmer WD, Berquist TH, McLeod RA, Sim FH, Pritchard DJ, Shives TC, Wold LE, May GR (1985) Bone tumors: magnetic resonance imaging versus computed tomography. Radiology 155:709–718
15. Beltran J, Simon DC, Levy M, Herman L, Weis L, Mueller CF (1986) Aneurysmal bone cysts: MR imaging at 1.5 T. Radiology 158:689–690