discouraging: only 50% of patients achieve seizure freedom, and 65% present a refractory course.[10] However, our case suggests that, in selected patients with drug resistant epilepsy related to focal cortical polymicrogyria, surgery can be offered with good results.

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Vertebral fracture due to a solitary bone cyst

Dear Editor,

Simple bone cysts (SBC), also known as solitary bone cysts [1] or unicameral bone cysts,[2] are common, benign, fluid filled, cystic lesions that occur mostly in the metaphysis of the long bones. Atypical bone involvements occur, however, in less than 5% of the cases [3, 4] including the vertebrae throughout the spinal axis.[2, 4-6] Most SBCs are self-limiting tumors and they nearly always occur in children and adolescents with a reported male-to-female predominance of 2:1.[4] Since Dawson *et al*.[3] reported the first case of SBC in the spine, only 17 cases of spinal SBC have been reported. Most of these cases are discovered incidentally and involve the cervical and lumbar spine (Table I). Tsirikos *et al*. reported the only case of thoracic SBC in the literature.[4] These tumors can involve both the vertebral body and the posterior elements.[2, 5] Elsewhere in the body these tumors are naturally painless and mostly remain silent.[1] However, when they occur in the spine they may come to clinical attention for spinal pain solely or with vertebral fractures accompanied by potential radicular symptoms. The clinical picture after vertebral collapse may range from painful neck or back to slight neurological deficits.[2, 7-9] In this report, a patient with T1 vertebral fracture due to a simple bone cyst is presented. Histological confirmation of the diagnosis is provided. Also, the differential diagnosis and planning of treatment is discussed in light of relevant literature.

Our patient was a 27-year-old woman who presented at our outpatient clinic for neck pain that had been getting worse for the last three months. She had no history of trauma and her neurological examination revealed no neurodeficits. Initially, a cervical XR was obtained which revealed normal results on lateral view, however a suspicious irregularity was noted on the upper end-plate of T1 on A-P view (Figure 1A, arrow). Further investigation with CT and MRI was performed (Figure 1B-F). The CT images revealed loss of height of T1 vertebra due to a fragmented compression fracture. An osteolytic lesion limited to the vertebral body was noted with

TABLE I.—*Summary of reported cases of spinal SBCs in the literature.*

Authors (year)	Age and sex	Vertebral level	Symptoms	Vertebral fracture	Treatment
Dawson *et al.*[3] (1976)	37y/M	C4	Neck pain	–	Curettage and bone graft/cement
Sawai *et al.*[18] (1980)	16y/F	L2	Back pain	–	Simple resection
Wu and Guise [19] (1981)	30y/M	L3	Back pain	–	Simple resection
Brodsky *et al.*[21] (1986)	31y/M	L1	Back pain	–	Curettage and bone graft/cement
Matsumoto *et al.*[22] (1990)	40y/M	L2	Back pain	–	Curettage and bone graft/cement
Nakagawa *et al.*[23] (1994)	63y/F	C5	Coincidental with radicular symptoms due to cervical disk hernia	–	Curettage and bone graft/cement
Park *et al.*[25] (1997)	12y/F	C2	Neck pain	–	Curettage/partial resection
Lee *et al.*[20] (2000)	14y/M	C7	Neck pain	–	Curettage
Zenmyo *et al.*[7] (2000)	13y/F	C7	Neck pain	–	Curettage and bone graft/cement
Shen *et al.*[24] (1998)	4y/F	C2	Neck pain and torticollis	–	Curettage and bone graft/ cement
Snell *et al.*[9] (2001)	10y/F	C7	Neck pain	Present	Resection with instrumentation
Chang *et al.*[16] (2001)	25y/M	L5	Coincidental with radicular symptoms due to lumbar hernia	–	Simple resection
Tsikoros and Bowen [4] (2002)	17y/F	T9	Back pain	–	Simple resection
Fujimoto *et al.*[26] (2002)	27y/F	L2	Back pain	–	Curettage
Ha and Kim [8] (2003)	53y/F	L1	Back pain, radicular symptoms	Present	Resection with instrumentation
Coskun *et al.*[5] (2004)	26y/F	C4	Neck pain, radicular symptoms	–	Simple resection
Ogata *et al.*[6] (2004)	50y/F	L3	Back pain	–	Curettage and bone graft/cement
Matsubayashi *et al.*[2] (2013)	20y/M	L3	Back pain	–	Curettage and bone graft/cement
Present study	27y/F	T1	Neck pain	Present	Resection with instrumentation

an anterior expansion of the anterior vertebral wall (Figure 1B). MRI study showed a cystic lesion, the content of which seemed hypointense on T1 weighted images (T1WI) and hyperintense on T2WI. The middle part of the vertebral body had a less hyperintense impression on T1WI and hypointense impression on T2WI, which was attributed to bleeding due to the fracture (Figure 1C-E). No edema was noted on adjacent vertebrae. No enhancement was noted in or around the lesion after IV gadolinium injection (Figure 1F). A differential diagnosis between an aneurysmal bone cyst and simple bone cyst was considered. Studies of CBC and blood biochemistry revealed normal results. MRI and blood tests did not provide any evidence to consider a neoplastic vertebral fracture. Since the radiological studies revealed that the lesion spared the posterior elements, a surgical treatment via an anterior approach was planned. A right-sided vertical incision was performed following the medial border of the sternocleidomastoid ending one fingerbreadth above the manubrial notch. The spinal column was reached via blunt dissection. The fractured vertebra was immediately noted due to its surface irregularity. T1 corpectomy as well as the discectomies for the adjacent levels were completed using microsurgical techniques. Intraoperatively, cortical thinning of the T1 was notable. The lesion was composed of a lobulated unicompartmental cyst surrounded by normal bone cortex (Figure 2). No inner lining membrane was found. The content of the cyst was made of a serous clear fluid, although streaks of blood oozing from fractured vertebral wall were noted. Both upper and lower endplates looked healthy. Upon completion of the T1 corpectomy, a titanium expandable mesh cage was put in place and supported by a plate fixation between C7-T1 (Fig3A-C). Histopathological study of the cyst wall with eosin-hematoxylin and Masson's trichrome staining showed that the cyst was surrounded by acellular collagenous tissue rimmed by cortical bone (Figure 2A, B). Neither epithelial lining nor inflammation was seen and normal degenerative fibrous tissue was noted on the inner lining of the cyst. No neoplastic process was reported. Therefore, a final diagnosis of simple bone cyst was made. The recovery of the patient was uneventful. She was discharged from hospital and advised to use a cervical collar. She reported significant pain relief after surgery.

Originally**,** **s**imple bone cysts were described by Virchow in 1876 [2, 10] under a generic categorization. However, it was Bloodgood who made their distinction from other cystic bone diseases in 1910.[11] Three decades later, Jaffe and Lichtenstein recognized them as the only true cyst of primary intraosseous origin and suggested that they were caused by a localized defect in the ossification process, occurring in the metaphyseal region adjacent to the growth plate during rapid bone growth.[12] The exact pathogenesis of SBC, however, still remains to be determined.[2] Several other theories exist, including venous obstruction due to increased intracystic pressure,[13] encapsulation of a metaphyseal hemorrhage, microtrauma,[14] and synovial rest.[1, 2, 4] Among them, the venous obstruction theory is one of the most accepted models.[2] Also, for SBCs occurring in the spine, microtrauma may play a role, as the overwhelming majority of the reported lesions involved the cervical and lumbar spine where there is more mobility in an older population of patients (Table I). This may explain why simple bone cysts occur in an older age group than the cysts of long bones.[5] Komiya *et al*. demonstrated that the cyst fluid plays an important role in the pathogenesis of SBC.[1] Cyst fluid causes an elevation of intracystic pressure [15] and contains proteolytic enzymes and oxygen radicals,[1] which are responsible for the degradation of bone matrix.

Because, SBCs occur in the spine very infrequently, their presentation always generates a diagnostic problem. The differential

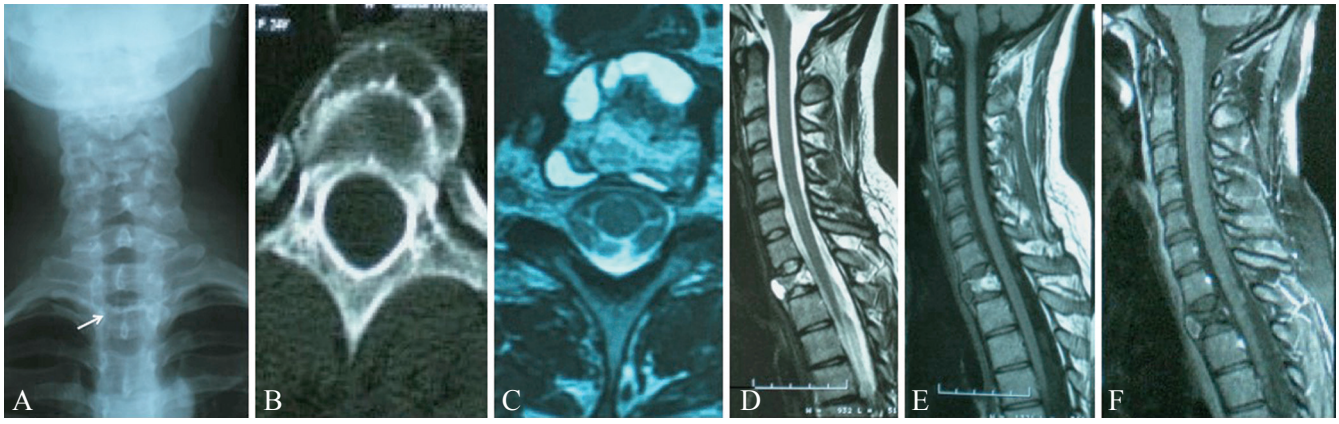

Figure 1.—Preoperative radiological studies: A) preoperative XR imaging; a subtle irregularity is noted on the upper lamina of T1; B) axial CT image of the lesion. C-F) Preoperative MRI study of the patient: C) axial T2W MRI; in comparison with B, the lytic anterior aspect of the vertebra on CT seems to have a hyperintense fluid content on T2W MR images; D) sagittal T2W MRI; E) sagittal T1W MRI; F) sagittal T1W MRI shows no contrast enhancement of the lesion after gadolinium injection.

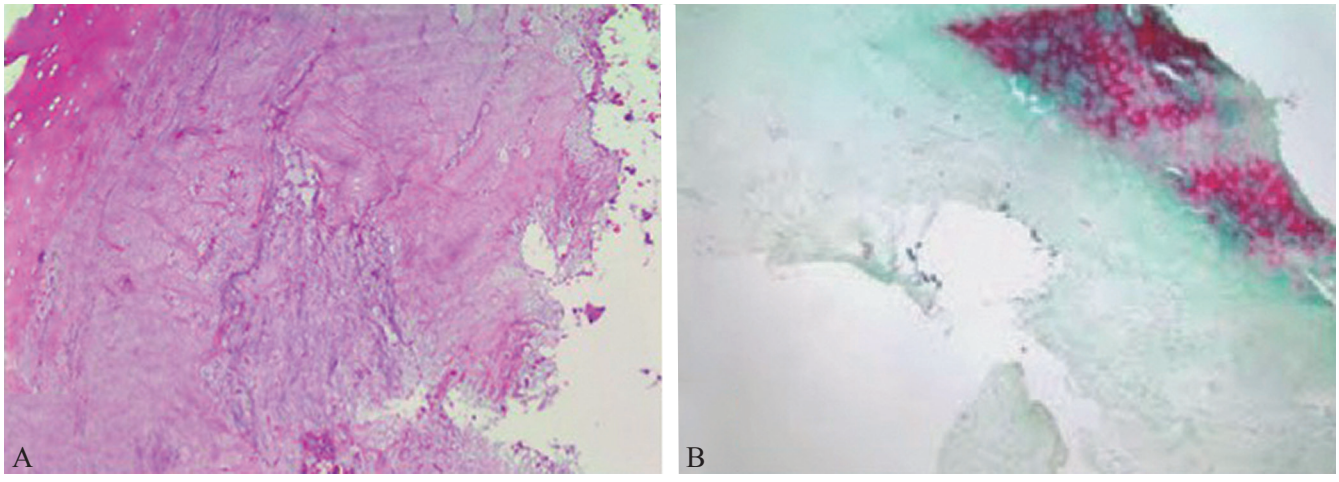

Figure 2.—Histopathological study of the cyst wall: A) hematoxylin and eosin stain shows that the cystic space is surrounded by a collagenous fibrotic tissue rimmed by cortical bone. (H&E ×100); B) note the green colored acellular collagenous lining of the cystic space (Masson Trichrome ×100).

diagnosis should include giant cell tumors of the bone as well as aneurysmal bone cysts.[4] Giant cell tumors are expansile, lytic, locally aggressive, primary benign bone tumors with thinning of the cortex. Most patients are between 20 and 40 years old. Giant cell tumors typically involve long bones. In the spine, the most typical site of localization is the sacrum.[5] These tumors may create osteolytic lesions on roentgenograms and CTs. However, they can be easily differentiated from SBCs on MRI, by demonstrating a solid mass with contrast enhancement.

SBCs generally appear as lytic lesions on roentgenograms and CT scans. However, the classic radiographic characteristics of solitary bone cysts of the tubular bones may not be easily identified in the spine, especially if the lesion involves the posterior bony elements.[4] In our patient plain radiographs showed a subtle endplate irregularity on AP view only with a careful inspection (Figure 1A, arrow). Further studies with a CT scan and MRI are useful in that they provide useful information concerning the osteolytic and cystic features of the pathologic process and define the surgical margins, help in the planning of surgical treatment. MRI is especially useful in providing information of the surrounding soft tissues and help in the differential diagnosis in collapsed lesions. In our case, the lack of enhancement of the lesion after gadolinium injection helped us rule out metastasis and other neoplastic fractures. Therefore, given the location of the lesion, the first consideration for differential diagnosis was aneurysmal bone cyst, even though the slight possibility of simple bone cyst was considered.

Aneurysmal bone cysts are multiloculated, expansile, highly

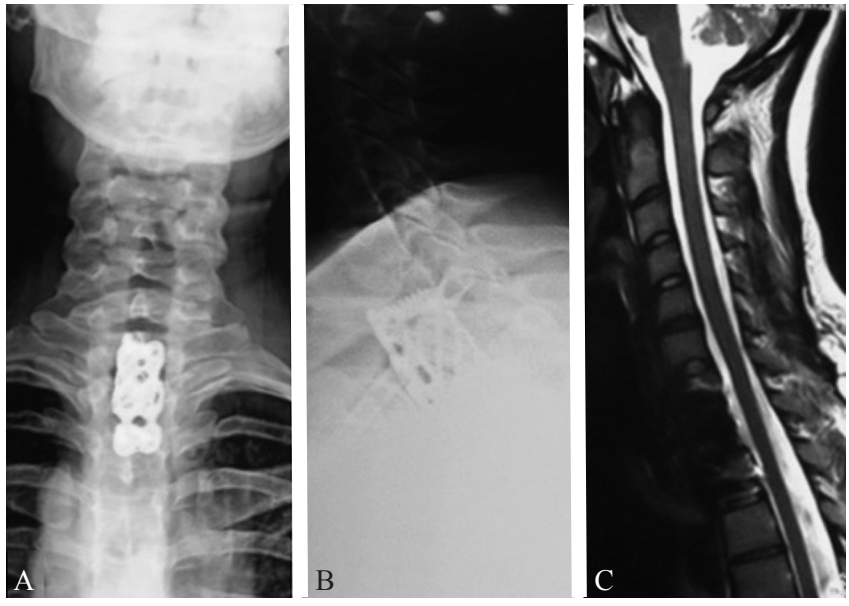

Figure 3.—Postoperative radiological studies: A, B) postoperative X-ray; C) postoperative sagittal T2W MRI shows the titanium mesh implant and anterior plate fixation.

vascular, osteolytic lesions that are filled with free-flowing blood products with fluid levels. They are common in patients younger than 30 years, with a slight female predominance. They particularly involve the long bones, however they may occur anywhere in the skeleton. The spine is rarely involved as well.[5] Unlike the simple bone cyst, which mostly involves the vertebral body and spinous processes, aneurysmal bone cysts mainly involve the laminae.[16] Similar to SBCs, radiographs and CT images indicate an osteolytic lesion that results in an expansion and thinning of the surrounding cortical bone. In contrast, however, a soft tissue mass is often present. Aneurysmal bone cysts are typically characterized on MRI images by their multiseptated appearance with fluid-fluid levels and blood degradation products.[17] Nonetheless, in a collapsed vertebra, it becomes more difficult to tell apart these two types of lesions and MRI, too, may become inadequate (Figure 1).

Intraoperatively, aneurysmal bone cysts also show thinning of the vertebral cortex but can be differentiated by their blood content,[5] in contrast to the serous content of an intact SBC. However, as in our case, collapsed SBCs may also contain blood. Therefore, the definitive diagnosis of a simple bone cyst will generally depend on histopathological examination of the tumor.[4] A serous cyst lined with fibrous collagenous lining and surrounded by thinned irregular cortical bone are the typical findings (Figure 2).

Review of the literature reveals only 18 cases of spinal SBCs reported up until today. Among these SBC cases, 8 were in the cervical spine, 1 in the thoracic spine, and 9 in the lumbar spine (Table I). Our report will add to the literature as the second case of SBC in the thoracic spine and third case of vertebral fracture. Various treatment modalities have been used in the past. The treatment is mainly based on where the cyst occurred and whether it resulted in a vertebral fracture. For non-weight bearing locations like the spinous processes, a simple resection is the treatment of choice.[5, 18, 19] If the lamina or the pedicles are involved, simple curettage [20] or curettage with bone graft or osteoinductive bone graft substitutes [3, 7, 21-24] have been used with success. These cysts may also be amenable for minimally invasive treatment modalities. Recently, Matsubayashi *et al*. reported a case of SBC involving the pedicle and lamina of L3, which they successfully treated with endoscopic curettage and bone graft filling.[2] However, for SBC with vertebral fractures, the issue of spinal instability has to be addressed. A radical excision and instrumentation is required. In our case, an anterior approach alone allowed us to resect the lesion totally, because the lesion was limited to the vertebral body. However, when all three vertebral columns are involved, combined anterior and posterior approaches may be necessary.[9] Surgical treatment seems to provide a cure for these patients, as there have been no reports of recurrence for spinal SBCs.

In conclusion, simple bone cysts of the spine are extremely rare and they may present with vertebral fractures. They should be included in the differential diagnosis of vertebral fractures due to bony pathologies.

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