CASE REPORT

Solitary Bone Cyst of the Odontoid Process and Body of the Axis: a Case Report

Ze-yu Huang, MD¹, Jing Chen, MD², Fu-xing Pei, MD¹, Yue-ming Song, MD¹, Li-ming Liu, MD¹

¹Department of Orthopaedics, West China Hospital, and ²West China College of Stomatology, Sichuan University, Chengdu, China

Introduction

Solitary bone cysts, also known as simple bone cysts or unicameral cysts, are common, benign, fluid-containing lesions found commonly in the metaphyses of long bones and extremely rare in the spine. Up to now, only 11 histologically proven cases have been reported^{1–11}. Five of these involved vertebral bodies^{1,3,6,7,9}, three a spinous process^{2,10,11}, one a pedicle⁵, one both a vertebral body and pedicle⁸ and one all components of a vertebra⁴. Only two of them were in the upper cervical segments^{8,9}. We here report a case of a solitary bone cyst in the odontoid process and body of the axis of a 32-year-old woman. The clinical features, pathogenesis, radiology, differential diagnosis and treatment are discussed.

Case Report

History

A 32-year-old woman was hospitalized with a 1 month history of neck and shoulder pain and limited neck activity. A cystic, expanding mass was seen on cervical radiographs taken in another medical center and she was referred to our center for further evaluation and treatment.

Examination

Physical examination confirmed pain of the neck and shoulder with mildly limited neck rotation. However, no neurological abnormalities were found. Plain radiographs and a 3D computed tomography (CT) scan showed a balloon-like unilocular osteolytic lesion in the odontoid process and body of the axis, with thinning of the pedicle cortex (Fig. 1A,B). Because the differential diagnosis included solitary bone cyst, giant cell tumor, aneurysmal bone cyst, or some other cystic lesion, 1.5-Tesla magnetic resonance images (MRIs) were obtained. T1-weighted MRIs showed low signal intensity within the lesion and absence of an adjacent soft-tissue mass, whereas T2-weighted MRIs showed high signal intensity (Fig. 1C,D).

Based on the history and radiographic findings, a diagnosis of solitary bone cyst was made.

Interventions

After undergoing cervical traction for 6 days, surgery to remove the suspected solitary bone cyst was performed via an anterior approach to expose the odontoid process and body of the axis. The patient was positioned supine on a radiolucent table with her placed in a Mayfield pin headrest. A standard Smith–Robinson anterior approach was used. On opening the thin shell of the lesion, a cavity filled with serosanguinous fluid was found; there was little bleeding (Fig. 2A). A 20-G needle was used to aspirate the yellowish, clear fluid after which the lesion was curetted (Fig. 2B) and the specimen sent for pathological examination. The lesion was then packed with autologous bone from the left iliac crest, taking care to achieve 1–2 mm between the bone graft and the posterior border of the lesion. The final positioning was confirmed radiologically and the thin anterior shell replaced. Blood loss was 400 mL and the operation time was 1 h and 30 mins.

Histologically, the bone wall was lined by thin fibrous connective tissue (Fig. 2C,D). After wearing a head and neck thoracic brace for 3 months, the patient returned to work. She was followed up three monthly with regular physical and radiographic examinations (Fig. 3). One year after the surgery, plain X-ray films showed no cystic mass and good stability of the atlantoaxial joint (Fig. 4B,C), and a 3D-CT scan showed successful obliteration of the previous cystic lesion (Fig. 4A).

Discussion

Solitary bone cysts, which were first described as a distinct disease entity by Jaffe and Lichtenstein¹², are tumor-like lesions that characteristically distend and thin the overlying cortical bone. These common lesions usually present in the first two decades of life and occur almost exclusively in the metaphases of long bones, particularly the proximal humerus,

Address for correspondence Li-ming Liu, MD, Department of Orthopaedics, West China Hospital, Sichuan University, 37 Wainan Guoxue Road, Chengdu, China 610041 Tel: 0086-28-85422430; Fax: 0086-28-85423848; Email: LLM_WCH@163.com, solo1987@yahoo.cn

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Fig. 1 Preoperative radiological images of a 32-year-old woman with the neck and shoulder pain and limited neck activity. (A) Lateral radiograph of the cervical vertebrae, showing an osteolytic lesion in the odontoid process and body of the axis. (B) Sagittal CT reconstruction, providing a clear view of a balloon-like unilocular osteolytic lesion in the odontoid process and body of the axis. (C,D) Low signal intensity within the lesion and absence of an adjacent soft tissue mass are evident in T1-weighted MRIs, whereas T2-weighted MRIs showed high signal intensity.

proximal femur and proximal tibia, which account for 75%–90% of all solitary bone cysts. Only a small percentage of cases involve the spine and even fewer the upper cervical segments. Interestingly, when solitary bone cysts are found in the spine, they tend to be in older subjects^{3,11}. A search of published reports of cases with spinal involvement yielded eleven cases, only two previous of which were located in the upper cervical segments.

The exact pathogenesis of solitary bone cysts has not yet been established. Suggested mechanisms include developmental defects of the epiphyseal plate, venous obstruction of interstitial fluid drainage, trauma and posthemorrhagic

lesions^{8,13,14}. According to the first of these, classic solitary bone cysts of childhood are caused solely by developmental defects of the epiphyseal plate, whereas the other two possible mechanisms involving trauma are considered to account for atypical solitary bone cysts in unusual sites, especially the spine. Our patient was 32 years old, well above the usual age for lesions in the long bone. In addition, she had no history of injury to the cervical spine. Thus, none of the above three mechanisms adequately explain our case, more likely mechanisms are yet to be developed.

When an osteolytic lesion is detected in the spine, the differential diagnosis should include aneurysmal bone cyst and giant cell tumor. It should also include solitary bone cyst, even

Fig. 2 Intraoperative and histopathologic images. (A) The lesion was exposed by an anterior approach, complete curettage was performed and autogenous iliac grafted into the lesion. (B) A 20-G needle was used to aspirate the yellowish, clear fluid. (C,D) Histopathologic examination showing a fibrous membrane lining the cyst wall (hematoxylin–eosin stain; magnification, 100 in C and 400 in D).

though these lesions rarely occur in the spine. In our case, radiologic examination showed that the lesion was unilocular. In addition, no fluid–fluid levels, hematomas or soft tissue masses were identified radiologically; thus, aneurysmal bone cyst was excluded. Giant cell tumors are destructive or aggressive and multiple compartments and heterogeneous signal intensity are commonly shown by MRIs². Thus, our preoperative diagnosis was solitary bone cyst, which was confirmed by the histological examination.

So far, there is no gold standard for treatment of solitary bone cysts. Because of the high recurrence rate, surgery is not the treatment of choice for young patients except when they have large lesions or lesions in weight-bearing locations. In our case, the location of the lesion was distinctive: it involved the odontoid process and body of the axis, leaving a very thin cortex of odontoid and thus could have led to fracture of the odontoid process during normal activity of the atlantoaxial joint. Such a fracture might have resulted in instability of the atlantoaxial joint or even migration of the fractured bone tissue into the spinal cavity, resulting in severe neurological

impairment. Our patient was a young to middle-aged woman who put strong demands on the atlantoaxial joint in fulfilling her daily work and life duties. We therefore chose to treat her lesion by curettage and autogenous iliac bone graft to preserve both the stability and activity of the atlantoaxial joint.

Recurrence is the main concern after surgery for a solitary bone cyst: the rate of recurrence is high. It has been postulated that the rate of recurrence is associated with age and size, younger patients having a much higher recurrence rate than older ones¹⁵. However, the rate of recurrence in rare sites is relatively low. None of the 11 reported cases which involved the spine developed recurrence. Our patient had no evidence of recurrence after 1 year of follow-up, no restriction in daily activities and was satisfied with the outcome.

To our knowledge, this is the third reported case of solitary bone cyst involving the upper cervical segments. We treated it with curettage and autogenous iliac bone graft to preserve the stability and activities of the atlantoaxial joint. At one year follow-up the outcome was satisfactory and there was no evidence of recurrence.

Fig. 3 Postoperative images on the day of discharge showing adequate filling of the osteolytic lesion by autogenous iliac crest graft in the sagittal CT reconstruction image.

Fig. 4 Postoperative images at the one year follow-up. (A) Sagittal CT reconstruction showing a good result from the autogenous iliac bone graft. (B,C) Flexion and extension radiographs of the cervical spine, demonstrating that stability and good activity of the atlantoaxial joint has been preserved.

References

- 1.** Brodsky AE, Khalil M, VanDeventer L. Unicameral bone cyst of a lumbar vertebra. A case report. *J Bone Joint Surg Am*, 1986, 68: 1283–1285.
- 2.** Coskun B, Akpek S, Dogulu F, Uluoglu O, Eken G. Simple bone cyst in spinous process of the c4 vertebra. *AJNR Am J Neuroradiol*, 2004, 25: 1291–1293.
- 3.** Dawson EG, Mirra JM, Yuhl ET, Lasser K. Solitary bone cyst of the cervical spine. *Clin Orthop Relat Res*, 1976, 119: 141–143.
- 4.** Dhaliwal J, Abbas N, Hamid NA. CSF bone cyst in a cervical vertebra. *Br J Neurosurg*, 2010, 24: 600–601.
- 5.** Ha KY, Kim YH. Simple bone cyst with pathologic lumbar pedicle fracture: a case report. *Spine (Phila Pa 1976)*, 2003, 28: E129–E131.
- 6.** Matsumoto K, Fujii S, Mochizuki T, Hukuda S. Solitary bone cyst of a lumbar vertebra. A case report and review of literature. *Spine (Phila Pa 1976)*, 1990, 15: 605–607.
- 7.** Nakagawa T, Kawano H, Kubota T. Solitary bone cyst of the cervical spine—case report. *Neurol Med Chir (Tokyo)*, 1994, 34: 558–560.
- 8.** Park CK, Cho KK, Lee SW, Jeon JS, Kang JK, Choi CR. Simple bone cyst of the axis. *Childs Nerv Syst*, 1997, 13: 171–174.
- 9.** Shen Q, Jia L, Li Y. Solitary bone cyst in the odontoid process and body of the axis. A case report and review of literature. *J Bone Joint Surg Br*, 1998, 80: 30–32.
- 10.** Wu KK, Guise ER. Unicameral bone cyst of the spine. A case report. *J Bone Joint Surg Am*, 1981, 63: 324–326.
- 11.** Zenmyo M, Komiya S, Hamada T, Inoue A. A solitary bone cyst in the spinous process of the cervical spine: a case report. *Spine (Phila Pa 1976)*, 2000, 25: 641–642.
- 12.** Jaffe HL, Lichtenstein L. Solitary unicameral bone cyst with emphasis on the roentgen picture, the pathologic appearance and the pathogenesis. *Arch Surg*, 1942, 44: 1004–1025.
- 13.** Cohen J. Etiology of simple bone cyst. *J Bone Joint Surg Am*, 1970, 52: 1493–1497.
- 14.** Aegerter EE, Kirkpatrick JA Jr. Miscellaneous diseases of the skeleton. In: Aegerter EE, Kirkpatrick JA Jr, eds. *Orthopedic Diseases*. Philadelphia, PA: Saunders, Corp, 1968; 491–500.
- 15.** Mirra JM. Cyst and cystlike lesions of bone. In: Mirra JM, ed. *Bone Tumors: Clinical, Radiologic, and Pathologic Correlations*. Philadelphia, PA: Lea & Febiger, Corp., 1989; 1233–1261.