Case Report Destructive osteoblastoma with secondary aneurysmal bone cyst of cervical vertebra in an 11-year-old boy: case report

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Abstract: Study Design: A case report and review of previous literature are presented. Objective and Background: The objective of this manuscript was to report a case of destructive osteoblastoma with secondary aneurysmal bone cyst of cervical vertebra in a child, and discuss the pathogenesis of this disease. The combination of osteoblastoma and aneurysmal bone cyst in the cervical spine is rare in primary bone neoplasm. To the authors' knowledge, only one case in a child has been reported. Method: Plain X-rays, technetium bone scanning, CT scan and MRI indicated an expansile, partially sclerotic lesion of the C4 involving the body of vertebra and appendix. The lesion was excised through anterior and posterior approach. Results: After operation the tumor was removed completely. There has been no sign of tumor recurrence or clinical or radiologic sign of instability in the follow-up investigations. Conclusions: We report a rare case of destructive osteoblastoma with Secondary aneurysmal bone cyst of cervical vertebra in a child, a full investigation indicated that complete resection of the tumor can prevent recurrence and malignant transformation. Long-term follow-up is needed to declare a lifelong cure of the disease.

Keywords: Osteoblastoma, aneurysmal bone cyst, cervical vertebra, child

Introduction

Osteoblastoma (OTB) and aneurysmal bone cyst (ABC) are infrequent primary neoplasm of bone and individually occur in less than 1% of bone tumors [1-4]. Furthermore, the combination of OTB and ABC in the cervical vertebrae is relatively rare in the subclass of primary tumors [4, 5]. We report a case of destructive osteoblastoma with secondary aneurysmal bone cyst in the C4 of an 11-year-old child, including the clinical symptom, radiographic findings, and surgical intervention with spinal fusion. One-year clinical and radiographic follow-up is presented.

Case report

An 11-year-old boy was referred for neck skew to left side and suffered mild pain for 3 years. The cervical destruction was found during a health examination through X-ray. The past history was negative for any major cervical trauma or bone tuberculosis. On physical examination, the cervical range of motion was not limited. There was tenderness on the acantha of C4 and C5. No motor, sensory or reflex changes were noted at physical examination. Routine laboratory blood tests were all within the normal range. Chest X-ray was normal.

Plain cervical X-ray (**Figure 1A** and **1B**) showed a expansile lytic lesion located in the vertebrae, spinous process and posterior elements of C4. The X-ray reveal that the cervical spine had a sign of kyphosis, and the preoperative Cobb angles was proximal 35°. Bony fusion of the posterior elements was noted at C2-3. Flexion and extension lateral radiographs showed instability of the vertebrae. Computed tomography (CT) showed a large expansile multiseptated mass, involving C4 vertebral body, the right and posterior elements. It contained some saccate separation in the nidus and a sclerotic surrounding rim as the outer layer (**Figure 2**). In magnetic resonance (MRI) imaging, the tumor



Figure 1. A: Preoperative plain radiographs showed the expansile lytic lesion present in the posterior elements of C4, and B: Showed a sign of cervical kyphosis.



Figure 2. Computed tomography imaging showing the lytic expansile lesion's extension into the right sides of C4.

appeared to be >4 cm³ in volume, containing fluid level in the nidus. It was noted that the compression of the spinal cord and paraverte-

bral involvement, both anteriorly and posteriorly (**Figure 3A** and **3B**). According to the Enneking score of benign musculoskeletal lesions the tumor was classified grade 3 [6].

Operation

The patient accepted one stage anterior-posterior approach to resect the lesion completely. In operation, it was found that the tumor involved almost the whole cervical vertebrae, including bilateral transverse foramina. The posteriorlateral part of the left transverse foramen was resected, while the right vertebral artery and vein were separated and the whole right transverse foramen was removed. The other involved bone was completely removed by rongeur and microdrill without neural or vascular complications. In gross, the excised tumor mass, which are pinkish gray and friable, consisted of multiple capsular space and some sclerotin. On histological examination, the tumor was composed of osteoid trabeculae rimmed by osteoblasts embedded in vascular stroma. The tumor also contained cystic cavities filled with blood. Thus, it demonstrated the tumor was osteoblastoma with a secondary aneurysmal bone cyst component (Figure 4).



Figure 3. Preoperative MRI T1-weight (A) and T2-weight (B) image revealed a destructive lesion on the anterior and posterior parts of C4 with signs of spinal cord compression.

To reconstruct the stabilization of cervical spine, the lateral mass screw and rod was used in both sides of C3/5/6 in posterior approach, and the defect of C4 vertebral body was planted with appropriate length titanium net filled with shattered bone from left cristailiaca and fixed with titanium plate.

The postoperative radiographs showed the lesion was completely resected, the cervical kyphosis was rectified and the Cobb angles was proximal 0°. The patient was pain free after one-year follow-up, and showed no evidence of recurrence or spinal instability (**Figures 5, 6**).



Figure 4. Microscopically, the tumor is mainly composed of haphazard proliferation of osteoblasts. The fibrovascular stroma was associated with osteoblasts, osteoid tissue (H&E×200).

Discussion

Osteoblastoma (OTB) is rare benign tumor that accounts for approximately 0.5% to 1% of all primary bone tumors [7, 8]. It is described that around 20% to 40% OTB affected in cervical vertebrae [9]. Lesion is most frequently found in the posterior elements with potential extension to the vertebral bodies. In clinically, progressive pain is the main symptom, followed by torticollis, scoliosis and neurologic manifestations [10, 11]. In gross, the OTB is observed as circumscribed, friable and hemorrhagic and often forms extraskeletal bone in the soft tissue [2]. Osteoblastoma is similar to osteoid osteoma in histology, but osteoid osteoma is defined smaller than osteoblastoma [12].

Aneurysmal bone cysts are rare, benign lesion of the bone. It can be located in all bone types and often found in femur, tibia, and small bones of the hands and feet [13]. Approximately 8% to 30% of ABCs arise in the spine, mostly in the thoracic and the lumbar regions [14]. It has been reported to be 25% in the cervical spine [15]. Aneurysmal bone cysts frequently affect the patients younger than 20 years and seem to be slightly more frequent in females than males. In clinical, patients present with insidious onset back pain, stiffness, and swelling [16]. Due to cord compression and spinal instability, some patients present neurologic dysfunction. In gross, the lesion consists of a soft fibrovascular core containing blood-filled cystlike cavities surrounded by a bone shell [17]. Aneurysmal bone cysts are divided into two



Figure 5. Plain radiographs showed no evidence of recurrence.

classes: primary lesions developing independently and secondary lesions. The secondary aneurysmal bone cysts can be associated with osteoblastoma, giant cell tumor, osteosarcoma, and fibrous dysplasia [15, 18].

The association between osteoblastoma and aneurysmal bone cyst is quite rare in the general population [19]. Several cases of this association, such as the posterior cranial fossa [20], skull [21], ethmoid sinus [22], mandibular ramus and condyle [23], sacrum [24], were reported. To our knowledge, only one case of the osteoblastoma with secondary aneurysmal bone cyst of cervical vertebra in child has been reported [25]. In our case, the lesion extends to the vertebral body and posterior elements of C4 and caused to cervical deformity. Operative therapy is necessary for the patient. It has reported that osteoblastomas and aneurysmal bone cyst with incomplete resection has a high rate of recurrence [26, 27]. To avoid the recurrence of tumor, we choose the complete excision. But for large and extensive lesions complete resection will likely incur iatrogenic instability and need instrumented fusion [28]. In our patient, both anterior and posterior approaches were used to rebuild the cervical stability. After one year follow-up, the patient has no evidence of local recurrence and rachiterata.



Figure 6. A and B: MRI showed sufficient decompression of the spinal cord in the cervical spine and no recurrence of lesions after one-year follow-up.

Conclusions

The combination of osteoblastoma and ABC in the cervical spine is a rare occurrence in child. We reported a rare case of destructive osteoblastoma with secondary aneurysmal bone cyst, including clinical symptom, imageology examination and operation. Complete excision of the tumor offer the best chance of cure and a low rate of recurrence. Furthermore, a long time follow-up is necessary for precaution of tumor recurrence and kyphosis.

Disclosure of conflict of interest

None.

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