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Aneurysmal bone cyst of the spine

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Abstract
The authors present the case of an 11-year-old boy with a painful, rapidly expanding lesion in the posterior triangle of the neck. There was no history of cervical trauma. Computerized tomography of the neck revealed a unicameral (single-chambered) aneurysmal bone cyst involving the C3 vertebra. Treatment was by open resection and curettage; no recurrence was seen at six months. We discuss the natural history, differential diagnosis, radiographic appearance and treatment modalities for this unusual, benign, expanding, osteolytic lesion containing blood-filled cystic cavities.

Key words: Aneurysmal Bone Cyst; Cervical Spine; Surgical Procedures

Case report

An 11-year-old boy presented with a 10-month history of pain and swelling in the right posterior triangle of the neck. There was no history of trauma and he was otherwise well.

The swelling had been seen by the boy's general practitioner eight months previously, at which time it was considered to be of musculoskeletal origin and was treated with three months of physiotherapy. During this time the patient's symptoms were reported to have resolved. Two months after cessation of physiotherapy his symptoms of pain and mass had returned.

Examination revealed a 10 by 7 cm hard, immobile swelling in the right posterior triangle that was warm to the touch; there were no other masses or lymphadenopathy present. The remainder of the examination was unremarkable.

Fine-needle aspiration cytology (FNAC) of the neck mass was difficult to perform because of the hard consistency of the mass and three needles were blunted in an effort to gain a satisfactory sample. The FNAC revealed muscle fragments, osteoid, osteoblasts, osteoclasts and calcific material.

A computed tomography (CT) scan of the neck was performed revealing a large, cystic, bony lesion with a thinned but intact bony cortex and fluid / fluid levels of varying density. The lesion did not involve the spinal canal and from its appearance was thought to be an aneurysmal bone cyst arising from the third cervical vertebra (Figure 1).

An intra-arterial digital subtraction angiogram was performed in order to define vascularity with a view to embolization. Selective injections into the right common carotid and right vertebral and right costocervical trunk revealed no evidence of abnormal circulation to the tumour and embolization was thought not to be practicable as no definite feeding vessel was outlined.

Excision and curettage were performed by a neurosurgeon via an anterolateral approach. There was no pathological fracture and internal fixation was unnecessary. The patient made an unremarkable recovery without neurological deficit. Six months later he had no evidence of recurrence and was symptom-free.

Discussion

Aneurysmal bone cysts (ABC) are rapidly expanding, destructive, benign tumours of bone containing thin-walled, blood-filled, cystic cavities. They do not metastasize. The term 'aneurysmal bone cyst' was first coined in 1942 by Jaffe and Lichtenstein[1] based on radiographic appearances. Aneurysmal bone cysts make up 1.4 per cent of all bone tumours and 15 per cent of all primary spinal tumours.

Aneurysmal bone cysts occur predominately in children and adolescents, with the peak incidence occurring in those aged 16 years, and there is a slight female preponderance.[2] They occur most commonly in the metaphyseal region of the knee.[3] Other common sites are the forearm bones and the tibia; however ABC have been reported in all skeletal bones.

Presentation depends on the anatomical location of the cyst, but in the spine they present as a rapidly enlarging, painful mass which may appear 6–8 months following trauma. The skin temperature over the palpable swelling may be increased. There may be some crepitation as the egg-shell-like outer cortex is stressed and breaks. Cysts arising in the cervical spine may present with neurological symptoms including radiculopathy and even paralysis due to acute compression of the cord when the bone cyst arises in or extends into the spinal canal.[4] This might occur acutely following pathological fracture through a lesion. Cysts have also been described arising within the cranial cavity in which case the patient may present with the symptoms and signs of a space-occupying lesion.[5] One report has described progressive airway obstruction from subglottic stenosis due to an ABC situated in the larynx.[6]

These lesions may coexist with other bone conditions and should always be considered in the differential diagnosis of osteoblastoma, brown tumours occurring in

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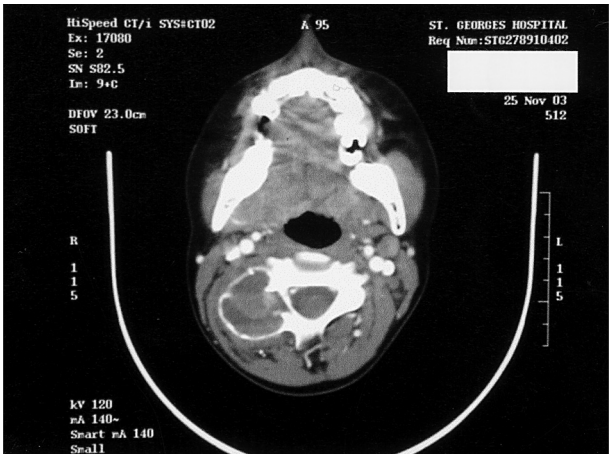

FIG. 1
Axial CT at the level of C3 demonstrating the radiographic features of an aneurysmal bone cyst.

FIG. 2
Egg-like appearance of the bone cyst.

hyperparathyroidism, enchondroma, haemangioma, giant cell granuloma, chondroblastoma, non-ossifying fibroma, fibrous dysplasia, telangiectatic osteosarcoma and chondromyxoid fibroma.[7] In the presence of other pathology they have been termed 'secondary ABC'.

The pathogenesis of ABC is unclear but an association has been suggested between ABC, bony development and repair with fracture as an initiator, or pre-existing bone lesions which feature arteriovenous malformations. It has also been suggested that there may be a familial element of inheritance in the formation of ABC but this is based on only one report of a primary ABC occurring in two family members.[8]

Successful FNAC diagnosis has been reported only once before in the literature but the cytological features may be indistinguishable from those of giant cell tumours of bone and of brown tumours of hyperparathyroidism.[9] As these usually arise in children or adolescents sampling may not be well tolerated due to the resistance of a surrounding shell of bone and the force required. There is also a risk that the sample may not be representative of the cytological field.

Open biopsy generally reveals an eggshell-thin layer of subperiosteal bone and a fibrous capsule with septa surrounding blood-filled channels. Microscopically, the fibrous septa contain fibroblasts, myofibroblasts and connective tissue containing numerous multinucleated giant cells. There are also haemosiderin-laden macrophages and new bone formation is found within the stromal matrix.[10]

On X-ray cysts have a characteristic well defined, expanded cortex and classic 'soap bubble' appearance due to the trabeculation and multiple septa.

Ultrasound imaging differentiates solid and fluid-filled collections, and fluid / fluid levels in the cyst particularly suggest the diagnosis.

Characteristic appearances on magnetic resonance imaging (MRI) include a ring of intact bone completely surrounding the lesion, multiple internal septa, contrast-enhancing cyst walls, fluid / fluid levels of varying intensity and a surrounding area of inflammation. The CT may be less sensitive and specific in the detection of ABC as it may not show the fluid / fluid levels or the cyst walls as clearly as MRI.[11] However CT is obviously invaluable when bone settings are used to aid the identification of fractures and to plan reconstruction. However MRI is more likely to reveal compression of the cord or rami when there is extension of the lesion into the spinal canal or vertebral formina.

Selective angiography has a role in defining abnormal vascularity of the ABC, and pre-operative embolization is possible.

Management over the last few decades has been surgical removal by opening the cyst and curetting the fibrous wall. More recently, this has been preceded by selective angiography with a view to pre-operative

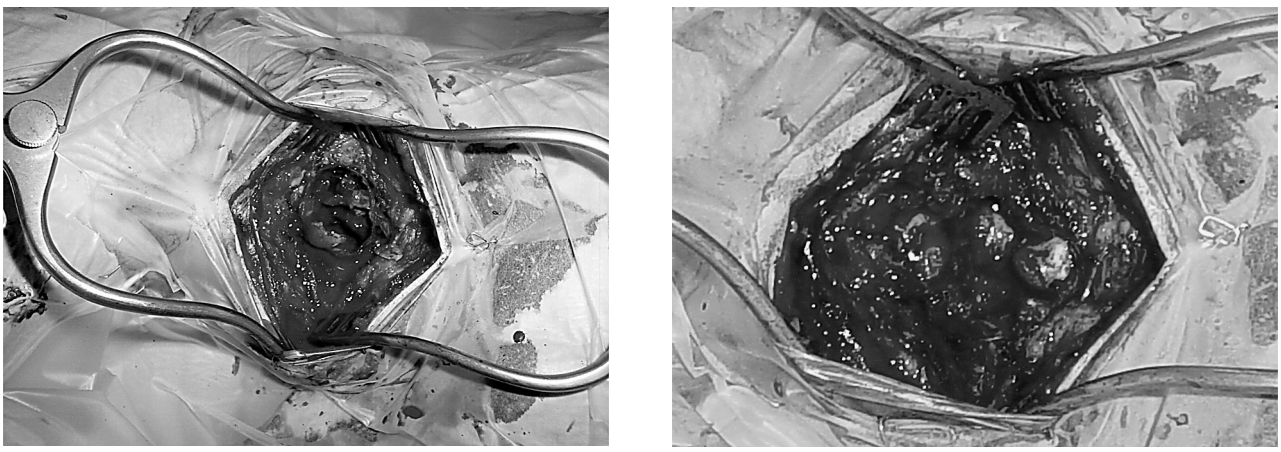

FIG. 3
(a) The opened bone cyst, and (b) underlying dura exposed at the C3 vertebra.

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embolization of any large feeding vessels. Operation within 48 hours of embolization is recommended to minimize blood loss from this highly vascular lesion. Prompt curettage of the cyst wall after opening the outer bony shell has also been recommended to reduce blood loss. If there is impairment of the structural integrity of the originating bone then autologous bone grafting may be required.

- **Aneurysmal bone cysts are uncommon, benign tumours occurring predominantly in children and adolescents**
- **This case reports a large aneurysmal bone cyst arising from the third cervical vertebrae and presenting as a neck swelling**
- **Treatment was by open resection and curettage**

Historically, radiotherapy was used but is now seldom employed as the majority of patients are less than 20 years old and there may be a risk of late-onset sarcoma in this group in whom there is no initial tissue diagnosis.[10] Some complications of radiotherapy may be avoided by radioablation of the cyst by injection into the cyst of radioactive colloids. Successful management has been reported for other sclerosing agents such as alcoholic zein and calcitonin.

Intriguingly, trauma also has been implicated in the spontaneous regression of ABC especially after pathological fracture.[12] In this case open biopsy had been performed previously when the radiological diagnosis was uncertain, and in one case a spontaneous regression occurred although this appears to be the exception rather than the rule.

Conclusion

Aneurysmal bone cysts are fortunately rare but can present with a rapidly expanding, palpable and painful mass occasionally complicated by pathological fracture and neurological compression.

It is important to exclude bony malignancy. Of the available treatment options surgical exploration with excision curettage and sometimes reconstruction offers the advantage of a clear tissue diagnosis.

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