Spinal aneurysmal Bone Cyst, surgical Treatment and Imaging (A Study of 8 Cases)

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Abstract: Patient with primary spinal aneurismal bone cyst was selected as regard to clinical presentation and radiographic findings. Surgical treatment with spinal radical bone curettage was performed for all patients without fixation. Neurological deficits marked improved after surgery in all paient. No radiological recurrence was detected along two years follow up period. Healing and ossification was found in 75% of cases.
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1. Introduction

Aneurysmal bone cyst (ABC) is rare, benign but locally destructive bone tumors. The term aneurysmal bone cyst was first described by Jaffe and Linchtenstien in 1992**1**. Since then, the name has been generally accepted although the lesion is neither aneurysm nor a cyst. The incidence of ABC amounts to 0.14 per 105 per year. Women are slightly more often affected than men, with a ratio of 1.04:1. The mean age of patients affected lies in the second decade of life. The long bones of the lower extremities are affected most often, with the tibia and femur being respectively affected in 24.7% and 7.3% of all cases, followed by the upper extremities 10%, and the pelvis 9%. About 14% of all ABCs are encountered in the spine, with those in the cervical spine making up only 2% (**2,3)**. The etiology of ABC is uncertain; it may occur in bone as a solitary lesion or can be found in association with other bone tumors such as giant cell tumor, chondroblastoma, chondromyxoid fibroma, and fibrous dysplasia or in association with a malignant process(**2,10) .**

Most of spinal ABCs affect the cervical region. Mutiple vertebral level is often affected and 50% occur in the pedicles, laminae and the spinous processes (**3,4)**. Most patients clinically present with symptoms of diffuse pain, neck back stiffness, and swelling. Symptoms usually persist for about 12 months before definitive diagnosis(**5).**

2. Clinical Material and Methods

Eight cases of primary ABCs of the cervical spine have been operated from 2008 to 2010 in the Prince Sultan Military Hospital, King Saudi Arabia. All patients were subjected to thorough general and neurological examination preceded by proper history taking including family history and previous trauma.

Radiological workup was done in the form of plain X-ray, CT scan, and MRI. Post operative following surgery patients were followed for a period of two years, both clinically and radiologically. During follow up period, cervical MRI and plain X-ray were done for all cases about 3 months after surgery and 6 months later to exclude recurrence.

Operative procedure

All patients were operated under general endotracheal anesthesia in the sitting position. This position helped us to decrease bleeding and wash blood from operative field in such bloody operation. Posterior cervical midline incision was performed and the area of the ABC was exposed after subperiosteal muscle separation. The thin eggshell wall of the cyst was first exposed with its usual intense bleeding. The shell is opened and the soft tissue content of the ABC was rapidly removed and curetted. The content is usually brownish, fleshy and highly vascular. There were thin trabeculae of gray tissue with variable amounts of soft tissue containing vascular channels and blood filled cysts of variable sizes.

Bleeding appeared to come from the soft tissue lining the cyst, which was profuse and difficult to control until complete removal of the soft tissue contents of the cyst. For this reason, trials of hemostasis go in vain before complete curettage of the soft tissue content of the ABC. Extreme caution was carried out during removal of the inner part of the cyst contents because usually the lamina protecting the cord was eroded. The eggshell wall was then dissected from the adjacent soft tissue and muscles, and if part was adherent to vital structures it was left behind. The wound was then closed in the usual fashion leaving a subfascial drain which was left for 48 hours.

3. Results

Patients in this study consisted of 3 males and 5 females (62.5%) with age ranging from 12 to 25 years with an average of 18 years, and 87.5% of them were under 20 years. Data regarding clinical presentation and examination are shown in table 1

Table (1): Clinical findings in 8 cases of ABC

Clinical manifestation	No	%
Neck pain and tenderness	8	100%
Myelopathy	3	37.5%
Radiculopathy	2	25%
Radiculopathy with myelopathy	2	25%
Palpable neck swelling	1	12.5%

One patient only has a history of trauma. Examination of cervical lymph nodes revealed no associated lymph node enlargement. Examination of the rest of the spine and other bones revealed no associated spinal or other bone involvement.

Data regarding the level of affection, the size, and the site of the vertebra affected are shown in table 2.

Table (2): Radiological findings in 8 cases of ABC

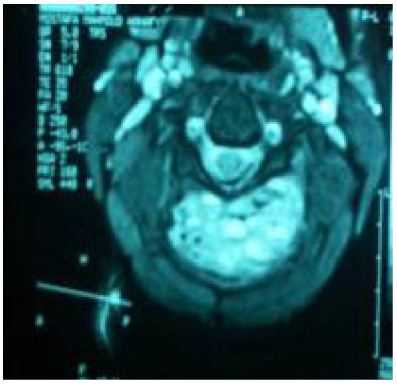
Parameters	No	%
- Level affected		
C2	4	50%
C5	1	12.5%
C6	2	25%
C7	1	12.5%
- Size in cm		
o 3cm or more	5	62.5%
o Less than 3cm	3	37.5%
- Site of vertebra affected		
o Both laminae and spinous process	3	37.5%
o One lamina and spinous process	4	50%
o One lamina, spinous and transverse process	1	12.5%

X-ray was performed to exclude other lesions. Lamina and spinous processes were affected in all patients, while the transverse process was only affected in one patient, and there was no pedicle or body affection.

The lesion was affecting C2 in 4 cases. During surgery 4 cases needed blood transformation and they were the cases with tumours 3cm or more. All cases improved following surgery as regard neck pain, myelopathy and radiculopathy with variable degree and rate of improvement. One patient showed transient Myelopathy, but she improved gradually within one month and improvement progressed with physiotherapy along the period of follow up.

Also, there was one case of wound infection that resolved completely with antibiotics according to culture and sensitivity. During the two years follow up period, radiological follow up showed no recurrence in any of our cases and cyst wall remnants showed later ossification which was found in 6 cases and it was considered a sign of healing.


A


B

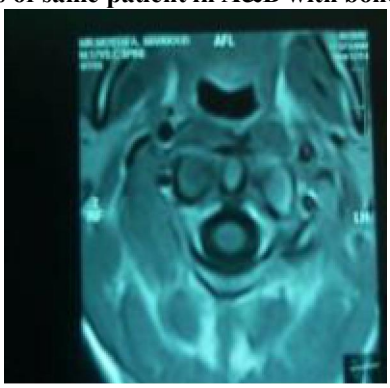
Pre operative Sagittal (A)T1 and axial (B) T2 MR images of Cervical spine aneurysmal bone cyst of its posterior element .

Spinal cord is free


C


D

Plain lateral films of same patient in A&B with bone erosion of its spinous process . No soft tissue mases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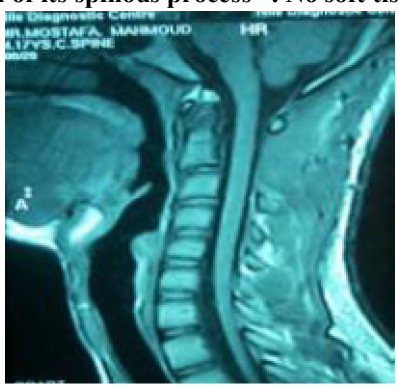
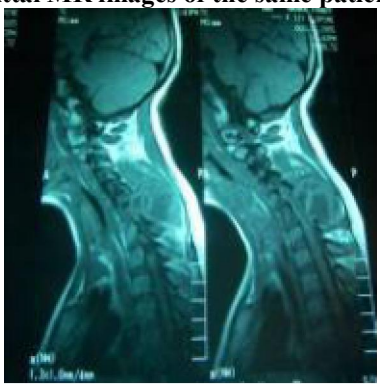
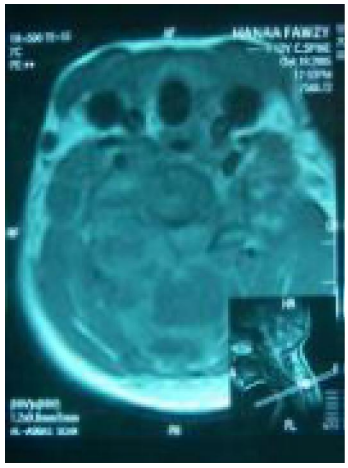
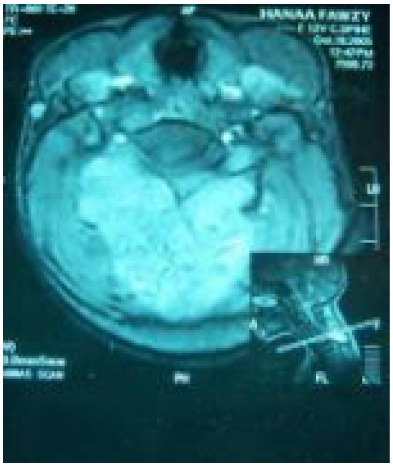

F

E, Axial and F sagittal MR images of the same patient in A and B showing complete excision of the cyst.


G


H

Pre operative Sagittal MR images of Cervical spine aneurysmal bone cyst of another case

I **J**

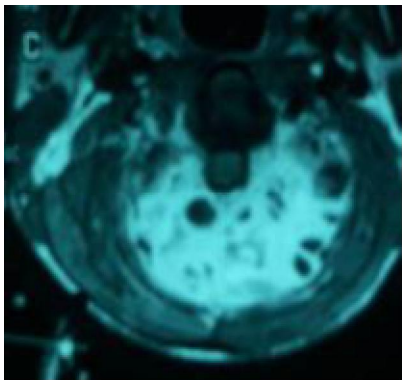
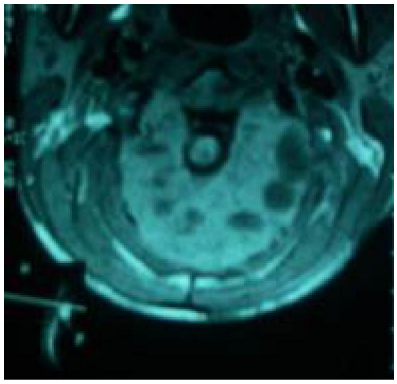
Axial T1 and T2 MR images of the same patient in G,H.

K

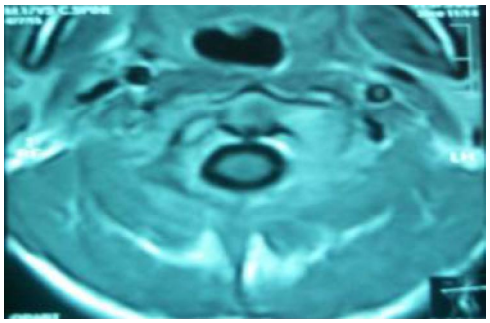
Plain X ray lateral view of the same patient in G and H. It shows complete bone erosion of the posterior elements of C6 and C7 with intact bodies.

L

Follow up sagittal T1 MR image of the same patient in G and H. It shows complete excision of the tumor

M  N 

Preoperative MRI images of a case with extensive aneurysmal bone cyst lesion. M, Axial T1 with contrast, N Axial T1 MR images

O 

Follow up axial T1 MR images of the same patient in M and N, showing complete excision of the tumor.

4. Discussion

Aneurysmal bone cysts are non neoplastic reactive condition, which is aggressive in its ability to destroy and expand bone.

Microscopically, the cyst may consist of a dense cellular composition containing plump stromal cells, multinuclear giant cells and thin-walled blood vessels, or a preponderance of fibrous tissue with enlarging vascular spaces (3, 6, 21). Both computed tomography and magnetic resonance imaging provide useful information in examining a possible ABC. CT typically reveals characteristic soap-bubble appearance, which represents a ballooning, multilocular lytic lesion. Thin walls of cortical bone result from erosion and expansion of the cortex (6, 9, 11).

Pathologic fracture or partial vertebral body collapse is a common finding.(7) Preoperative CT imaging is helpful for assessing pedicle and vertebral body integrity in anticipation of instrumented fusion (8). MRI is useful in examination of the full bone and soft tissue involvement including assessment of the potential compromise of the neural elements (7). Radioisotope bone scans can occasionally show increased uptake of traces in ABCs (8), and selective angiography can sometimes reveal the blood supply to these vascular lesions and may also occasionally reveal arteriovenous shunts(6)

While these tumours are benign, they rarely disappear spontaneously (7), and radiation therapy is ineffective and relatively contraindicated in young patients near the spine because of the risk of myelopathy (8), as well as the potential for radiation induced spinal deformity9. Accordingly, surgical excision with a complete marginal resection should be the goal. In this study the authors treated 8 cases of cervical ABC from 2000 to 2007 which is a reasonable number of cases compared to Boriani and Colleagues (20) who reported 41 cases in 44 years, and Ozakin (15) who reported 13 cases in 19 years, Papagelopous and Colleagues (16) reported 52 cases in 83 years. In this study no cases showed a relevant family history though some authors recorded a familial incidence in some of these lesions (14) they also stated that 75-80% of the cases occurred before the age of 20 years which corresponds with our results.

In this study, the cervical region was the only affected, where as in many published series, the lumbar vertebrae were the most affected. Malcolm and Colleagues (13) found equal number of cervical and lumbar vertebrae affected in their study and by taking into account the number of vertebrae in each region the dorsal spine should be the most affected. Camins *et al.* (21) found equal affection of thoracic and cervical spine.

Regarding the site of involvement in the vertebrae most authors agreed that posterior element was involved in all cases, and the lamina was the most commonly involved part(.21,22) This coincides with our results, but we have few numbers of cases, however most authors stated that the body was never affected alone but with the posterior element.(21,22) Schachar and his colleagues (17) described a case of complete destruction of the entire vertebral body which progressed to vertebral plane due to ABC.

In this study there was no affection of 2 adjacent vertebrae, Capanna *et al.* found (4) lesions involving adjacent vertebrae and even ribs in the thoracic region, they stated that the lesion had crossed to the adjacent bone either at the articulation between the posterior elements, at the facet joints or at the articulation of the transverse processes with the ribs. Malcolm *et al.* found affection of adjacent vertebrae in 5 cases out of 14. Complete surgical resection is the treatment of choice with aneurysmal bone cysts of the spine especially in patients who have a neurological deficit (22)

Preoperative selective spinal angiography was advocated by some authors (22) for defining the relationship of the arterial supply of the lesion to the arterial supply of the cord, also to define the involvement of the vertebral artery in cases of cervical lesion and the anatomic location of the artery of Adamkiewicz in lower thoracic or upper lumbar lesion. Moreover preoperative embolisation is a useful adjunct that may decrease the intra operative blood loss (22).

Other authors suggested the use of embolisation when the involvement of the lesion is so great as in sacrum, or when total excision or radical curettage would be too risky (7-9) Selective arterial embolisation should not be considered if there is pathological fracture or neurological involvement. Though the use of somatosensory evoked potential and the use of provocative sodium amytal test were described by some authors during selective arterial embolisation, other authors stated that embolisation should be contraindicated in the cervical location because of high risk of cord damage as well as the thoracic region because of the risk of Adamkiwicz artery occlusion(2,7-9).

Radiotherapy, which is another modality of treating ABC although effective in some cases, is contraindicated in children. The risk of malignant transformation (Radiation induced sarcoma) myelopathy and endplate growth disturbance compared with the benign behavior of the lesion does not justify this type of treatment. Capanna and colleagues (4) reported 6 cases treated with radiation alone using 2600 to 5000 rads with 50% complication rate, and one patient died from radiation induced septicemia and endocardities.

There seems to be little justification for needle biopsy. The results are likely to be negative, and the procedure is potentially dangerous owing to the risk of epidural hematoma(22) Subtotal surgical excision is followed by a high incidence of recurrence which is usually rapid (within 1year and often within 4 months). In this study there was no recurrence along the follow up period (1-2 years).

Conclusions

Complete surgical excision and curette of bone cyst have excellent prognosis. However, surgery has its complications and surgical difficulties which can be predicted and treated . MRI is the best imaging modality in spinal bone cyst

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