Abstract: Primary Aneurysmal Bone cysts account for 1.4% of all primary bone tumours and 15% of all primary spine tumours. ABCs are expansile cystic lesions and can be locally aggressive. ABCs of the dorsal spine can cause neurological complications and parasthesia. In this case report we present a 24 year old male patient with aneurysmal bone cyst of the third dorsal vertebra, with neurological deficit who was treated with open biopsy and posterior stabilisation (Stage 1), selective arterial embolisation (stage 2), anterior decompression-intralesional excision of the tumour and fusion (stage 3) and radiotherapy (stage 4). Neurological status fully recovered after radiotherapy.

Keywords: Aneurysmal bone cyst-dorsal spine

CHALLENGES ENCOUNTERED IN THE MANAGEMENT OF ANEURYSMAL BONE CYST OF THE THIRD DORSAL VERTEBRA - A CASE REPORT

Presenting Complaints: A 24 year old male patient reported to the Ortho Spine surgery department with complaints of upper backache of 2 months duration. The backache was intermittent in character and progressively increasing in severity over the last two months. He complained of numbness in both lower limbs of 2 months duration. He also had urgency of micturition and symptoms of incomplete bowel evacuation for 1 month duration. On examination, he had tenderness over the upper dorsal spine. The motor power in the upper limbs was 5/5 and in the lower limbs the motor power was 4/5. Sensations were decreased below D6 level. The knee and ankle reflexes were brisk and plantar reflexes were extensor.

Radiology: X ray dorsal spine AP view
As part of the initial management, the D3 vertebral lesion was open biopsied and then posterior stabilization was done.

Post operative x ray after open biopsy and posterior stabilization
Post operative x ray lateral view after open biopsy and posterior stabilization

The Biopsy report came as aneurysmal bone cyst.

Stage II: Selective Arterial Embolisation

One month later the feeding artery was embolised. Repeat MRI showed decreased size of the lesion. The Neurological status was static. The patient was discharged with Taylor's brace and advised physiotherapy. The patient was again admitted after 10 days with sudden onset of weakness in both lower limbs with power of 2/5 and exaggerated reflexes.

Stage III: Anterior decompression

Anterior decompression was done by Left Thoracotomy approach. Intralesional excision or the

Expansile, lytic lesion was seen involving the pedicle, transverse process, lamina and extending into the vertebral body of D3 vertebra, with multiple internal septae. T2 weighted MRI imaging showed a hyperintense lesion of D3 vertebra with hypointense thin septa, with fluid-fluid levels.

DIFFERENTIAL DIAGNOSIS:
A differential diagnosis of Giant cell tumour, Aneurysmal bone cyst, Osteoblastoma, Ewings sarcoma, Chondroblastoma of the D3 vertebra was made. We decided to biopsy the lesion to confirm the diagnosis.

MANAGEMENT
Stage I: Posterior Stabilisation & Open Biopsy.
Intraoperative photograph of anterior decompression done by left thoracotomy approach.

Post Operative X Ray AP view after intralesional excision of the tumor and fusion using cage.

Post Operative X Ray lateral view after intralesional excision of the tumor and fusion using cage.

Post operatively patient had good improvement in power and attained 5/5 in right side and 4/5 in left side within a week. Patient was discharged.

ADMISSION 3-RADIOTHERAPY

Patient was again readmitted after one month with complaints of weakness of both lower limbs. The motor power was 2/5 in both lower limbs and reflexes were exaggerated. Patient was given radiotherapy of 30 gy in 15 cycles and the neurological status improved to 5/5 in both lower limbs.

DISCUSSION:

Aneurysmal Bone Cyst is a benign expansile osteolytic bone neoplasm, characterized by blood filled spaces separated by fibrous septa. They are locally aggressive. The peak incidence is in the 2nd decade. The etiology is unknown. Aneurysmal bone cysts constitute 1.4% of all bone tumors & 15% of all primary spine tumors. They commonly occur in flat bones and in the spine. Aneurysmal bone cysts are common in the thoracic and lumbar region. Pain is the main presenting symptom. In spine, Aneurysmal bone cysts arise commonly in the posterior elements and extend to involve the pedicle, body & spinal canal resulting in pathological fractures and neurological compromise. ABCs may occur secondary to Giant Cell Tumor, chondroblastoma, osteoblastoma and osteosarcoma.

TREATMENT OPTIONS

The treatment options for Aneurysmal bone cysts of the dorsal spine include simple curettage with or without bone grafting, complete excision, selective arterial embolisation, radiation therapy, or a combination of these. Staging system of the musculoskeletal tumor society (MTS) for Aneurysmal bone cyst summarized by Enneking.

Bush and Drane reported in 2000 treating an ABC of the dorsal spine with radionuclide ablation by injection of P-32 colloid into the lesion. Primary ABCs respond to radionuclide ablation, while secondary ABCs do not respond. Marushimo et al reported treating an aneurysmal bone cyst of the dorsal spine by Selective Arterial Embolisation using n-Butyl cyanoacrylate. Selective Arterial Embolisation is time consuming and there are chances of ischemic insult.

Selective arterial embolization of the feeding artery of ABC

Embolisation using NBCA improved clinical symptom by reducing cyst size and accelerating ossification. There was no recurrence in 3 years of follow-up. Long term follow up is needed as 2nd embolisation or surgical treatment may be needed in future. Marushimo et al from Childrens Hospital Boston reported their experience of surgically treating aneurysmal bone cysts of the spine. There was a high chance of recurrence after incomplete excision of aneurysmal bone cysts of the spine. Recurrence occurred even after 8 yrs and long term follow up was needed. Preop embolisation had no significant effect on blood loss. After complete excision and stabilisation, the long term prognosis was excellent. In 2008, Ohashi et al reported treating an aneurysmal bone cyst of the C2 vertebra with percutaneous intalesional injection of calcitonin and methyl prednisolone.
Percutaneous intralesional injection of calcitonin and methylprednisolone

This procedure is an important alternative to surgery, especially when a surgical procedure is technically difficult or unsafe in high-risk patients.

External beam RT:

The curative potential of RT in Aneurysmal bone cysts has been well documented in many clinical studies. The mechanism by which RT can affect ABC is not completely understood, but an obliteration of the small arteries has been suggested. This results in a reduction of the blood supply to the ABC lesion. RT is also alternatively used with other adjuvant therapies as a post-surgical therapy in anatomical sites where complete removal is impossible in order to sterilize the residual cyst tissue.

Furthermore, RT has to be considered after excision of recurrences in order to improve the local efficacy. The total RT doses reported in the literature range between 5-60 Gy. CT-based 3-dimensional RT treatment planning is mandatory to achieve an optimal coverage of the whole soft tissue mass and a protection of the surrounding normal tissue. The complications reported were myelopathy and radiation induced sarcomas.

Conclusion:

Bone pain is the presenting symptom in aneurysmal bone cysts of the Dorsal spine. The involvement of the spine is associated with various neurological symptoms possibly resulting from the intra spinal extension or the extradural compression of the spinal cord. A spinal parasthesis develops when the lesion progresses. Intralesional gross total resection reduces local recurrence. The development of recurrence is influenced by the completeness of resection. Radiation in the treatment of ABCs has been used in selected patients after biopsy or surgery. Embolization is performed primarily before surgery to decrease intraoperative blood loss through Selective Arterial Embolization (SAE).

References:

5. Campanacci M, Cervellati C (1976) Aneurysmal Bone Cyst (Study of 127 cases, 72 with long term follow up) Ita J Orthop Traumatol 2;341-353
9. Edling NP, (1965) Is Aneurysmal bone cyst a true entity, Cancer 18;1127-1130
12. Georgios Zenonos, Osama Jamil, Surgical treatment for Primary Spinal Aneurysmal Bone Cyst, Experience from Children’s Hospital, Boston, Spine 40 no 8 p 230-234,2011
17. Lichteinstein L, 1953 , Aneurysmal bone cyst, Cancer 6;1228-1237