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Aneurysmal Bone Cyst of the Dorsal Spine: Case Report

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***Abstract*— Aneurysmal bone cysts are uncommon lesions, especially in the spine. They account for 1.4% of all primary bone tumors and 15% of all primary spine tumors. ABCs are expansible cystic lesions and can be locally aggressive.**

There is a definite predilection for the lumbar region and the neural arch is the part of the vertebra most commonly affected. It is recommended that treatment should consist of total excision or when this is not possible, curettage. Radiotherapy should be reserved for those few cases where operation is inadvisable.

***Index Terms*— Aneurysmal bone cyst, ABC, Dorsal spine**

I. INTRODUCTION

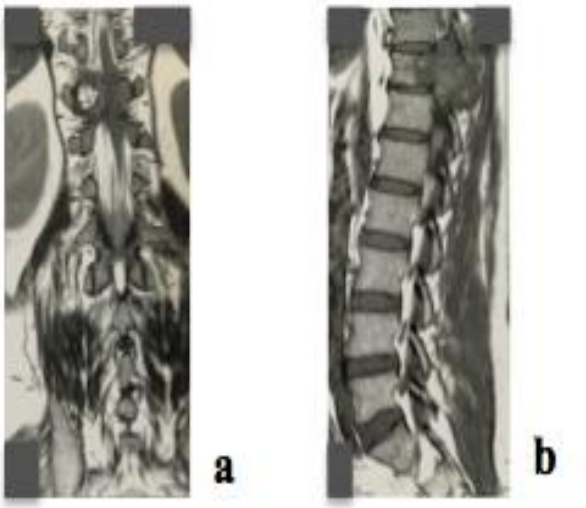
In 1942 Jaffe and Lichtenstein described two cases of a peculiar blood cyst which they termed an aneurysmal bone cyst. They recognized that the nature debated of the lesion was not clear, but they were of the opinion that the blood-filled cavity should be regarded as a large venous pool" . The term aneurysmal was used to denote the " blow out" radiographic appearance which resembles the saccular protrusion of the walls of clinical features of an aneurysm and also because cystic blood-filled spaces are encountered at operation. The name has been generally accepted, though indeed it is neither aneurysm nor a bone cyst [1] - [2].

Current theory holds that it is a non-neoplastic reactive condition, which is aggressive in its ability to destroy and expand bone. The histogenesis of ABC remains unclear. It may occur in bone as a primary lesion or can be found in association with other tumors, such as GCT and chondroblastoma [3] - [4].

Aneurysmal bone cyst (ABC) is an expansible cystic lesion, which has the distended appearance of an aneurysm and controversial pathogenesis [5] - [6]. The age distribution of ABC is distinct, with the majority of lesions occurring in skeletally immature patients. Although benign, ABCs can be locally aggressive. Primary ABCs, that is, arising independently with no underlying lesion, account for 1.4% of all primary bone tumors and 15% of all primary spine tumors [5] - [7]. Treatment modalities for aneurysmal bone cysts of the dorsal spine have included en-bloc excision, curettage, radiation therapy, and curettage with radiation therapy. Surgical excision is often complicated by profuse hemorrhage. Angiography and selective arterial embolization has been used as a preoperative means of reducing tumor vascularity [8] - [9].

II. CASE REPORT

A 46 years old male patient reported with complaints of upper backache of 6 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. 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.
Spinal MRI showed an expansive process implanted on the right posterior arch of T11 of 5cm of transverse diameter and 5cm of Antero-posterior diameter. This lesion has polylobed contours made of multiple boxes with hydro-hematic content and an inclined hematic component. It overflows forward on the point Antero-postero-lateral right of the vertebral body and causes spinal cord compression at this level.



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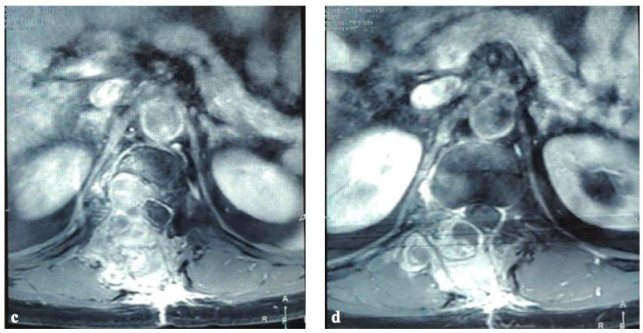

Figure: Medullar MRI showing an ABC of the dorsal spine a/ Coronal reconstruction b/Sagittal reconstruction c-d/ axial plane

A complete surgical resection was performed and showed discovertebral tissue dissociated by aneurysm fissures filled with hemorrhagic suffusions. These slits are lined with numerous inflammatory cells with histiocyts and giant multinucleated cells within a fibrous and siderophagic background. The diagnosis of ABC was established. After 3 months, the patient had a relapse of the process and had a radiation therapy on the T11 with a total dose of 30Gy, 15 fractions of 2 Gy in 23 days.
After 1year, the patient had no complaints and on the last MRI of spine, there were a 28% increase in aneurysmal cyst size of T11. He was referred to his neurosurgeon for a surgery.

III. DISCUSSION

Aneurysmal bone cysts (ABCs) are expansible osteolytic lesions that contain blood-filled spaces separated by fibrous septa containing giant cells [5] - [10] - [11].

Aneurysmal bone cysts constitute 1.4% of all bone tumor & 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 [10].

Recent studies have suggested that the pathophysiology of ABC is similar to that of giant cell tumors of bone (GCTB). In GCTB, the malignant cell secretes receptor activator of nuclear factor k-B ligand (RANKL) [12] - [13] - [14].

The pathogenesis of ABC has been postulated as vascular [15] - [16], neoplastic [17] - [18] and endocrine [19] - [20].
The Peak incidence is in the 2nd decade [10]. ABC is typically found in children and adolescents under the age of 20 years, affecting females more often than males [21] - [10].

Aneurysmal bone cysts have been described at every level of the spinal column except the coccyx. There is a definite predilection for the lumbar spine [23].

These lesions are often not confined to one vertebra and it has been noted that the neural arch is the most common site. In an analysis of seventy-four cases, 40 percent occurred in the bodies and 60 per cent in the pedicles, transverse processes, laminae and spinous processes [2].

Patients typically present with pain and an enlarged mass in the region of lesions that progress for months. On radiographs, ABC lesions appear as an expansible, radiolucent cyst most often in metaphysis [22]. The natural history can be divided into 3 or 4 phases. The initial phase is a small lytic lesion with evidence of lifting off of the periosteum. The second phase (or mid-phase) is the growth phase; the lesion is rapidly enlarging and demonstrates a characteristic blown out appearance. These lesions are noticed clinically at this phase. On initial radiograph, they appear very aggressive and are very commonly mistaken for a malignant tumor. The third phase is the stabilization phase, when growth is slowed or stopped by some type of intervention, allowing the periosteum to lay a new collar of bone down as the lesion moves into the healing phase, which is characterized by progressive ossification and calcification of the cavity [24].

Staging system of the musculoskeletal tumor society (MTS) for Aneurysmal bone cyst summarized by Enneking [10] - [25].
Treatment of ABCs is usually by primary surgical resection, which achieves very good control rates. For expendable bones, an en bloc resection can be performed with local control approaching 100% [25]. However, when en-bloc resection cannot be performed, the usual surgical procedure is curettage with or without the use of a physical adjuvant (cryosurgery, phenolization) and bone graft or bone graft substitute (methylmethacrylate), which results in local control rates ranging from 65–90% [26] - [27]. At sites where an adequate surgical procedure would result in significant morbidity and/or poor cosmetic results (such as vertebral lesions), radiotherapy (RT) has been used as an alternative.
The curative potential of RT in Aneurismal bone cysts has been well documented in many clinical studies [25]. The mechanisms by which RT can affect ABC are not completely understood, but an obliteration of the small arteries has been suggested.
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 [10].
Furthermore, RT has to be considered after excision of recurrent lesions in order to improve the local efficacy [28]. 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 [10] - [25].

Historically, RT for ABCs has resulted in local control rates of 75–100% in many small series (comparable with surgical results). Several investigators reported an incidence of radiation-induced malignancies as high as 12–25%, causing RT to fall out of favor. In the literature, all of the patients who developed radiation-induced malignancies were treated

suboptimally, using obsolete equipment and techniques. Low-energy photons are acceptable for treating superficial tumors (less than 2 cm deep). Unfortunately, for deeper tumors the skin or superficial dose can be significantly higher than the dose prescribed to the tumor. In addition, when treating near the bone, the biologic effective dose can be increased by the increased absorbed dose in bone and by increased scatter. When megavoltage therapy is used, both of these issues are eliminated, thus reducing the radiation dose to the overlying skin and the surrounding tissues, making the distribution of dose more homogeneous [20] - [25].

RT is administered using megavoltage therapy and CT treatment planning to a prescribed tumor dose of 26–30 Gy using standard fraction sizes [20].
Denosumab is a monoclonal antibody to RANKL, which had beneficial effects in the case of ABC. He reduces osteoclasts and increases serum sclerostin, which leads to depressed bone formation, and inhibits DKK1, which inhibits the sclerostin effect, thus increasing osteoblastogenesis [29] - [30] - [31].
Denosumab can result in symptomatic and radiological improvement in ABC and may be useful in cases not amenable to surgical interventions, although the long-term outcome is unknown. [12] - [32].
About 90% of the relapses of ABC lesions occur within the first 2 years after therapy. Thus, careful follow-up exams should be conducted for at least 2 years after treatment, but a follow-up of 5 years and more seems to be more adequate.
All patients who receive irradiation should undergo lifelong monitoring for secondary neoplasms.

IV. CONCLUSION

The optimal prescription of radiation dose is essential for avoiding the development of adverse reactions to RT. At our institution, we treat ABCs by delivering between 26 and 30 Gy at 1.8 to 2.0 Gy per once-daily fraction or 1.2 Gy per twice daily fraction to yield the most optimal outcome.

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