Cytological Diagnosis of Aneurysmal Bone Cyst of Pubis with Radiological and Histopathological Correlation A rare finding

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Abstract

Aneurysmal bone cyst is an expansile, destructive lesion of bone first recognized as a pathologic entity in 1942. Historically,they have been considered nonneoplastic cystic masses that have been divided into primary and secondary (histologically indistinguishable) types. The former occurs de novo, and the latter arises adjacent to and in conjunction with primary neoplasms of bone, ex. giant cell tumors and chondroblastomas. It represents approximately 1- 2% of primary bone tumors. Of all aneurysmal bone cysts, about 8-12% of them occur in the pelvis.Aneurysmal bone cyst of pubis is a rare entity.

Case report : A 16 year male patient presented with swelling in right groin measuring 5x4 cm for past 1 year, firm in consistency. Smears studied in H&E/Pap revealed moderate cellularity showing benign appearing oval to spindle shaped cell clusters interspersed with multinucleated osteoclast giant cells. Background showed cyst macrophages, histiocytes and proteinaceous material in a hemorrhagic background. Radiological and further histopathological findings correlated with our findings.

Conclusion : Although excisional biopsy is diagnostic for aneurysmal bone cyst, fine needle aspiration cytology is very helpful in pre-operative diagnosis of aneurysmal bone cyst because of its peculiar findings on smear.

Key words : Aneurysmal bone cyst, cytology, pubis.

Introduction

Aneurysmal bone cyst (ABC) is an expansile, destructive lesion of bonefirst recognized as a pathologic entity in 1942.The origin of the term "aneurysmal bone cyst" stems from two cases reported by Jaffe and Lichtenstein in their article on unicameral bone cysts in 1942. In this report, they noted two "peculiar blood-containing cysts of large size, "which were described as aneurysmal cysts. In a subsequent paper, Jaffe chose the name "aneurysmal bonecyst" as the descriptive term for this lesion, with the word "aneurysmal" to emphasize the "blown-out," distended contour of the affected bone, and the words "bone cyst" to underscore that when the lesion is entered through a thin shell of bone, it appears largely as a blood-filled cavity [1]. Historically, ABCs have been considered nonneoplastic cystic masses that have been divided into primary and secondary (histologically indistinguishable) types. The former occurs de-novo, and the latter arises adjacent to and in conjunction with primary neoplasms of bone, eg, giant cell tumors and chondroblastomas. Histologically, ABC is characterized by channels and multiloculated cyst-like spaces filled with blood and lined by fibrous septa that may or may not contain osteoclast-like giant cells, osteoid, woven bone, and chondroidmatrix material [2,3]. ABC represents approximately 1% to 2% of primary bone tumors, occurs predominantly within the first 2 decades of life with peak incidence in 2nd decade and its incidence is 0.14 / 100,000 and female predominace with a ratio of 1:1.3. The routine skeletal radiologic features of ABC vary depending on its developmental phase. In the long bones, most cases arise in the metaphysis and produce a lytic lesion that is circumscribed in the early phase and progresses to an expansile mass with cortical destruction. The most common site is metaphysis of the knee. Pelvis involvement ranges from 8-12%[4].They may easily be mistaken for a malignant tumor both radiographically and pathologically because it has essentially no metastatic potential, distinction between idiopathic causes and tumorous conditions is of paramount importance. The use of fine-needle aspiration cytology (FNAC) for the initial evaluation of primary bone

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tumors is controversial, although it is gaining popularity at several large medical centers [4-11]. The evolution of less invasive procedures for the initial evaluation of clinically primary bone tumors has evolved out of necessity to minimize disruption of the tumor bed to prevent potential dissemination of disease. In addition, it is required that data from the clinical examination and radiographic findings be correlated with the FNAC features (triple test)[5-12]. Aneurysmal bone cyst of the pubis is a rare entity. Cytological studies are done rarely.

Case report

A 16 year male patient presented with swelling in right groin measuring 5x4 cm for past 1 year and it was firm in consistency (Figure 1).

Figure 1. Swelling at the right groin region

Radiological examination revealed characteristic expansile, eccentric, well-circumscribed zone of rarefaction which appeared as a blow - out lesion of superior ramus of the pubis (Figure 2).

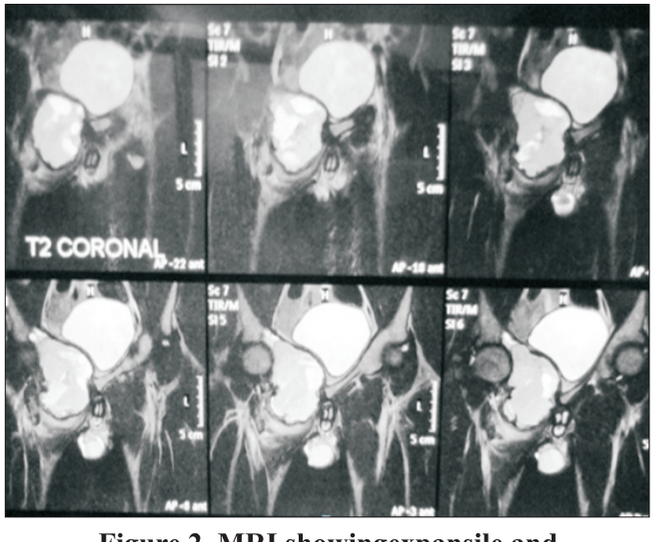

Figure 2. MRI showingexpansile and eccentric growth from the pubic ramus.

On performing fine needle aspiration, an aspirate of 10 ml of blood tinged serous fluid was aspirated. Smears studied in H&E/Pap revealed moderate cellularity showing benign appearing oval to spindle shaped cell clusters interspersed with multinucleated osteoclast giant cells. Background showed cyst macrophages, histiocytes and proteinaceous material in a hemorrhagic background (Figures 3-5).

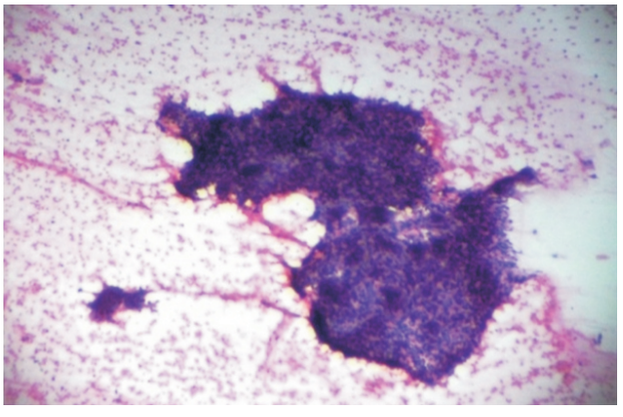

Figure 3. Spindle shaped cell clusters interspersed with multinucleated osteoclast giant cells, cyst macrophages and histiocytes. (H&E,100X)

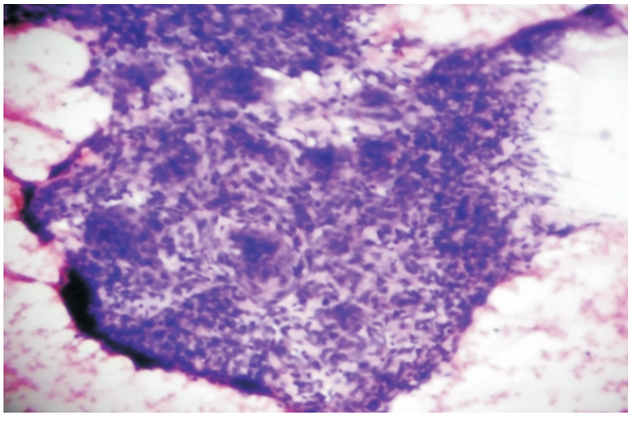

Figure 4. Spindle shaped cell clusters interspersed with multinucleated osteoclast giant cells (H&E,400X)

Hematological investigations were within normal limits. Serum alkaline phosphatase level was increased. Histopathology confirmed the diagnosis which on section showed large cystic spaces of various sizes with few of them blood filled, separated by fibrous tissue septae, containing many multinucleated giant cells (Figure 6).

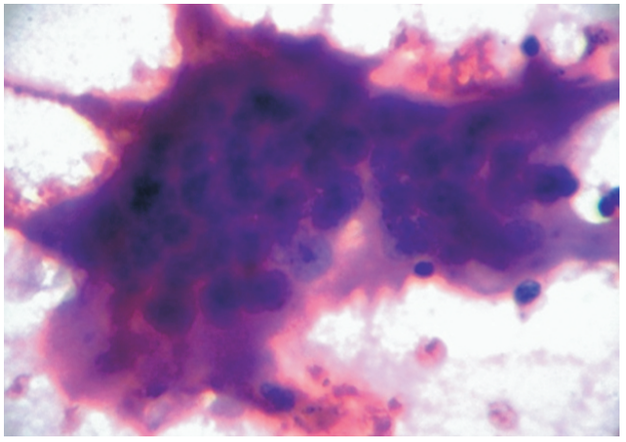

Figure 5. Multinucleated osteoclast giant cell with histiocytes (H&E,400X)

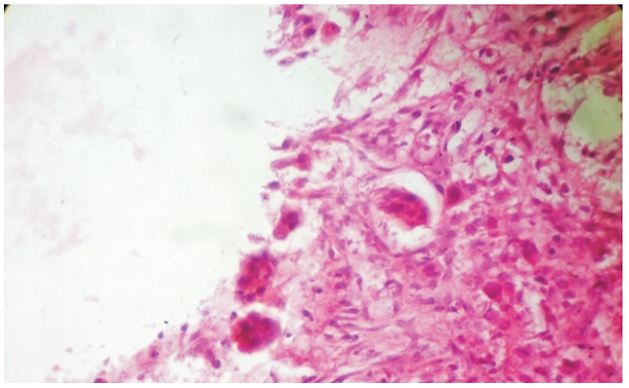

Figure 6. Histopathological confirmation showing cyst wall and multinucleated giant cells (H&E,100X)

Discussion

Aneurysmal bone cyst is a non-neoplastic expansile bone lesion consisting of blood-filled spaces separated by connective tissue septa containing bone or osteoid and osteoclast giant cells [4]. Ninety percent of patients with ABC present before the age of 30 years [13]. A study of pelvic bone cysts by Hammound et al. revealed that the majority of bone cysts occur in the ilium, followed by pubic ramus and ischium respectively [14]. Radiologically, ABCs from pelvis usually demonstrate a fusiform expansile "blowout" lesion. Sometimes, an expansile, lobulated, lytic, multiseptated cystic lesion will contain fluid-fluid level. In our patient radiological findings were consistent with expansile, "blowout", lytic changes. Although the findings mentioned above may suggest ABC, it also may suggest many other disease entities such as osteosarcoma and giant cell tumour [4, 15]. Historically, ABCs have been considered nonneoplastic entities that account for 1% to 2% of clinically significant, noninflammatory mass lesions. They are divided into primary and secondary types, the former unassociated with another primary bone tumor. Evaluation of primary bone tumors by FNAC is controversial, although it is gaining popularity. The close differential diagnosis is Giant cell tumor of the bone and granulomatous lesions of bones. In the Giant cell tumor, main cytological findings are abundant cellularity, a double cell population of mononuclear spindle cell population and giant cells of osteoclastic type and these giant cells are attached to the periphery of the clustered spindle cells.In our case the presence of histiocytes and cyst macrophages in addition to above features on cytology and histopathological confirmation helped to differentiate the two entities. FNAC is minimally invasive compared with open biopsy and has a low complication rate and high diagnostic value. Complications such as poor wound healing, inappropriate placement of the incision, and contribution of open biopsy to unnecessary limb amputation have been cited as disadvantages of open biopsy procedures and can be avoided by using closed biopsy procedures such as FNAC. The complications of FNAC are rarely reported. Radiologic correlation is the most critical factor, and, in general, it has been demonstrated that the ability to review these findings with the appropriate clinician increases the accuracy of FNAC[7-12]. In our case, preliminary cytological findings along with the radiological investigations helped us to clinch the diagnosis of aneurysmal bone cyst of pubic ramus which itself is a rare location for ABCs. Further histopathological findings correlated with our initial findings thus showing the significance of fine needle aspiration cytology as an initial mode of investigation.

Conclusion

Fine needle aspiration cytology plays a pivotal role in the surgical decision making for aneurysmal bone cysts, as a rapid, easy, cost effective and non-traumatic maneuver which can be carried out as an outpatient department procedure. It along with radiological investigations provides high diagnostic accuracy with further confirmation by histopathology of the excisional biopsy. Recurrence occurs most commonly during the first two post-operative years. The factors influencing recurrence are still unknown. Age, lesion location, lesion size and number of mitotic figures have been suggested. Aneurysmal bone cyst of pubis is a rare entity and a preliminary cytology investigation along with the radiological findings helped clinch the diagnosis. Hence, we are presenting this case.

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