Aneurysmal bone cyst of pubis

A rare presentation

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ABSTRACT

يعد كيس العظم الدموي عادة من الأورام الحميدة والمتمددة، مع انحلال العظم الوحيد، و دم يملأ تجويف الكيس الذي ينمو به بشكل غريب. و بالمناسبة، قد يصل نمو الورم الغريب إلى حجم ظاهر قبل أن يتم تشخيصه سريرياً، خصوصاً في حالة نمو الورم في مركز العظم. يعد الكيس العظمي الدموي للعانة على حسب علمنا حالة نادرة وصفت بشكل قليل في الأدب. في ما يلي تقرير حالة كيس العظم الدموي مصدره الجزء العلوي للعانة و ينتشر إلى عضلة الفخذ مع ألم خصوصاً مع تحمل الوزن. أظهر حجم و موقع الورم الوعائي صعوبات في العلاج. تمت السيطرة عليه عن طريق الكشط باستخدام مثقب عالي السرعة، و إعادة بناء الطعم العظمي الأسفنجي للعرق الحرقفي.

Aneurysmal bone cyst is a benign usually expansile, solitary lytic lesion of bone; with blood filled cystic cavities, which tend to grow eccentrically. Such eccentric growth may, on occasion, attain considerable size before it is clinically recognized, particularly if the tumor arises in a deeply situated bone. Aneurysmal bone cyst of pubis is a rare entity. In the following case report, an aneurysmal bone cyst, originating from the superior pubic ramus, expanded into the groin and first manifested itself as a gradually increasing fullness in groin and pain especially with weight bearing. The size and location of this vascular tumor posed difficulties in treatment. It was managed by curettage with high speed burr and reconstruction of the defect with an iliac crest cancellous bone graft.

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Aneurysmal bone cyst (ABC) is a benign, locally destructive lesion of bone that was first described as a distinct entity in 1942 by Jaffe and Lichtenstein.[1] Although the precise nature is not clear, most authors favor the opinion that these lesions are not neoplastic. Approximately 79% of cases, it arises as a primary tumor without any recognized precursor bone lesion or in about 30% of cases, as a secondary lesion when a pre-existing osseous lesion can be identified.[2] Aneurysmal bone cyst constitutes approximately 6% of the primary lesions of bone and is seen predominantly in children; 90% of these lesions occur in patients less than 20 years of age. There is a slight predilection for females and with great variability in the clinical course. In most cases there is pain and swelling and sometimes a pathological fracture.[3] It can involve any bone, but the most common locations include distal femur, proximal humerus, proximal tibia and so forth. The metaphysis of long bones is a frequent site of predilection, although ABCs may sometimes be seen in the diaphysis of a long bone, as well as in flat bones such as the scapula or pelvis and even in the vertebrae. The radiographic hallmark of an ABC is multiloculated eccentric expansion (blow-out appearance) of the bone, with a buttress or thin shell of periosteal response. Magnetic resonance imaging findings are rather characteristic and usually allow a specific diagnosis of ABC.[4] These include a well-defined lesion, often with lobulated contours, cystic cavities with fluid–fluid levels, multiple internal septations and an intact rim of low-intensity signal surrounding the lesion. Lack of understanding on ABC origin and growth makes treatment empirical. The most common treatment has been curettage with bone grafting which has a substantial rate of recurrence. Lower recurrence rates can be achieved by marginal or wide resection, but such treatment is accompanied by loss of bone and the need for reconstruction. Intralesional resection or curettage with effective adjuvant therapy to extend the surgical margin has been advocated.[3]

Case Report. A 17-year-old female presented with a dull pain and fullness in her left groin that had progressed over past 1 year. Pain was more troublesome while running or jumping. On examination, there was fullness in the left groin area. Palpation revealed a mildly tender, hard swelling seems to be arising from the left pubis without any signs of inflammation (**Figure 1**). The inferior extent of this mass could be palpated per vaginally. The radiological examination of the pelvis revealed a destructive lesion in the left superior pubic ramus and body of pubis with faint septations (**Figure 2**). The whole architecture of the superior pubic ramus and body of pubis is lost. The hemogram, total serum proteins, serum albumin-globulin ratio, serum calcium, serum phosphorus and serum alkaline acid phosphatase were all within normal limits. Magnetic resonance imaging of the pelvis showed well defined multiloculated thin walled cystic lesion with thin internal septae occupying almost whole of the left pubis with multiple fluid filled levels (**Figure 3**). The lesion showed multiple hypointense areas within it on T1W1 sequences suggestive of hemorrhage. The fine needle aspiration cytology report was inconclusive, which revealed the hemorrhagic smear. Based on these clinic-radiological features a provisional diagnosis of ABC of the left superior pubic ramus and its body was carried out. Surgery revealed a thin walled cyst with partially loculated areas with bony trabeculae that contained few milliliter of brown red un-clotted blood (**Figure 4**). The lesion was curetted out and high speed burring was carried out. The cavity was packed using iliac crest cancellous bone graft. Histopathological examination of the bone and fibrous tissue revealed blood filled multi-loculated cystic areas lined with fibrous tissue, osteoclasts such as giant cells and silvers of osteoid, consistent with the diagnosis of ABC. Post-operatively, patient was kept on absolute bed rest for 6 weeks, followed by partial weight bearing for next 4 weeks. Full weight bearing was allowed at 10 weeks after operation. Patient is now walking full weight bearing with the full range of movements after a follow up period of 12 months. At her last visit to the hospital, pelvic radiograph showed the lesion in the pubic bone has healed completely without any evidence of recurrence (**Figure 5**).

Discussion. Aneurysmal bone cyst is described as a locally destructive lesion of bone rather than a true bone neoplasm. The pelvis is not an unusual site for an ABC; approximately 12% of cases occur in this location.[5] Aneurysmal bone cyst of pubis is a rare entity. On occasion, ABC of pelvis may attain considerable size before it is clinically recognized, and may present as

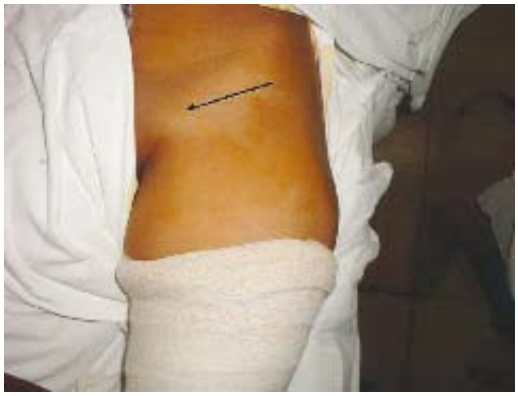

Figure 1 - Clinical photograph showing fullness in left groin area (pubic region [arrow]).

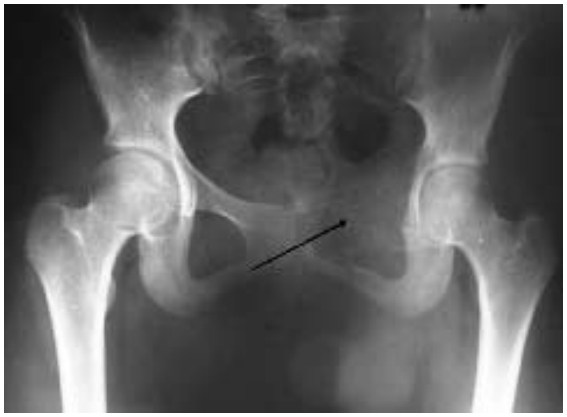

Figure 2 - Pre-operative radiograph of the pelvis showing a destructive lesion in the left superior pubic ramus and body of pubis with faint septations (arrow). The whole architecture of the superior pubic ramus and body of pubis is lost.

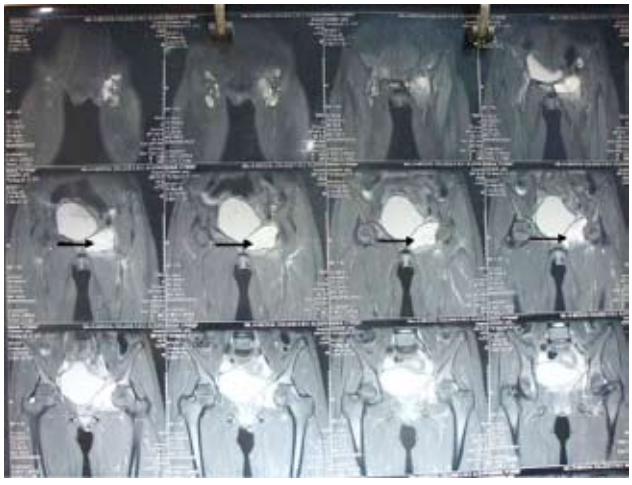

Figure 3 - Magnetic resonance imaging of the pelvis showing well defined multi-loculated, thin walled cystic lesion with thin septa occupying almost whole of the left pubis with multiple fluid filled levels (arrow).

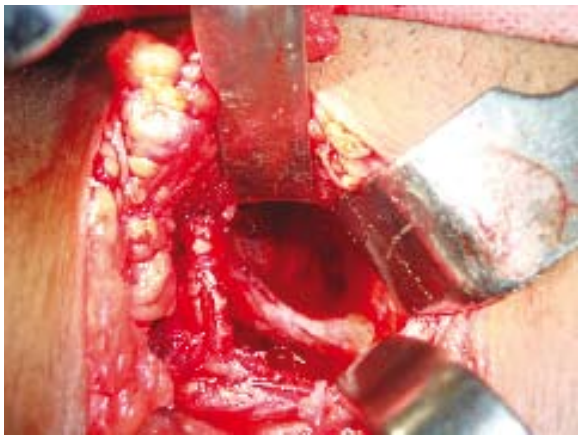

Figure 4 - Intraoperative photograph showing cavity after curettage with high speed burring.

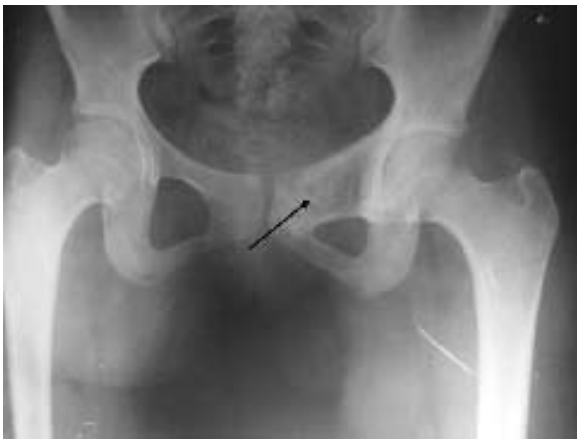

Figure 5 - One year postoperative radiograph of the pelvis showing the defect in the pubic bone that has healed completely without any evidence of recurrence. Note: reappearance of normal architecture of the pubic bone.

an abdominal mass.[6] Although radiologic features and therapeutic management of ABC have been extensively described, involvement of pelvis particularly of the pubis is relatively rare. The differential diagnosis of ABC includes The simple bone cyst, giant cell tumor, and intraosseous lipoma. In cases of ABC, MRI will typically show a well-circumscribed, macro-lobulated cystic lesion, often containing multiple fluid–fluid levels. The finding of fluid–fluid levels is non-specific; however, it can be seen with other osseous lesions including fibrous dysplasia, simple bone cysts and chondroblastomas.[7] Fluid levels are rare in giant cell tumor. Intraosseous lipoma may present as a sharply defined cystic radiolucent lesion. In our patient, MRI of the pelvis showed well defined multi-loculated, thin walled cystic lesion with thin internal septae occupying almost whole of the left pubis with multiple fluid filled level, which correlates with the histologic finding of large blood filled multi-loculated cystic areas lined with fibrous tissue, osteoclasts like giant cells and silvers of osteoid-consistent with the diagnosis of ABC. Some of the treatment modalities described for ABC - include curettage, cryotherapy, bone grafting, irradiation. Since ABCs are usually easily eradicated by curettage, irradiation should only be reserved for inaccessible lesions. Use of cryotherapy has been recommended as an adjuvant to the surgical treatment of ABCs.[3] On the assumption that ABC has an intrinsic potential to heal by ossification, a minimally invasive procedure was developed by Docquier. They introduced demineralized bone powder mixed with bone-marrow aspirate into the cyst to arrest the expansion phase and to allow the cyst to ossify. They found that this minimally invasive technique can promote the self-healing of ABC in 11 out of 13 cases. As no curettage is required, the proposed treatment avoids extensive surgery and blood loss and is convenient for the treatment of poorly accessible lesions in the pelvis.[8]

Papagelopoulos et al,[9] studied 40 consecutive patients with ABC of the pelvis and/or sacrum which were treated from 1921-1996. They found that ABC of the pelvis and sacrum are usually aggressive lesions associated with substantial bone destruction, pathological fractures and local recurrence. Current management recommendations include pre-operative selective arterial embolization, excision-curettage, and bone-grafting. Short follow-up and small sample is limitation of this study.

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Case Reports

Case reports will only be considered for unusual topics that add something new to the literature. All Case Reports should include at least one figure. Written informed consent for publication must accompany any photograph in which the subject can be identified. Figures should be submitted with a 300 dpi resolution when submitting electronically or printed on high-contrast glossy paper when submitting print copies. The abstract should be unstructured, and the introductory section should always include the objective and reason why the author is presenting this particular case. References should be up to date, preferably not exceeding 15.