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Simultaneous existence of unicameral bone cysts involving the femur and ischium

Femur ve iskiyumda eşzamanlı basit kemik kistleri

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We report a 30-year-old male patient with two unicameral bone cysts (UBC) simultaneously located in the proximal third of the right femur and ipsilateral ischium ramus, respectively. Fine needle biopsies were attempted for both lesions. Biopsy of the femoral lesion under local anesthesia was unsuccessful, so an open biopsy was performed which confirmed the diagnosis of UBC. Biopsy of the ischial lesion was not sufficient for diagnosis. Cytological examination of both specimens showed no other benign or malignant pathology. The femoral lesion was treated with intralesional (due to its large size) excision-curettage, bone grafting, and the introduction of a long gamma locking intramedullary nail to prevent the occurrence of a pathological fracture. The ischial lesion was left untreated and followed conservatively. The patient was free of any symptoms and complications three years postoperatively. This is the first report of an adult patient with UBCs simultaneously located both in a long tubular bone (femur) and a flat bone (ischium ramus).

Key words: Bone cysts/diagnosis/pathology; femoral neoplasms/surgery; ischium/pathology.

Bu yazıda, sağ femurun proksimal üçte birlik bölümünde ve aynı tarafta iskiyum ramusta eşzamanlı olarak basit kemik kisti (BKK) saptanan 30 yaşında erkek hasta sunuldu. Her iki lezyon için de ince iğne aspirasyon biyopsisi yapıldı. Femurdaki lezyondan lokal anestezi altında biyopsinin başarılı olamaması nedeniyle açık biyopsi uygulandı ve biyopsi sonucu tanı BKK olarak kondu. İskiyumdan alınan biyopsi ise tanı için yeterli olamadı. Her iki lezyondan alınan biyopsilerde başka benign ya da malign patolojiye rastlanmadı. Femoral lezyon, büyük olması nedeniyle intralezyonal eksizyon-küretaj, kemik greftleme ve patolojik kırık riskine karşı uzun gama kilitli intramedüller çivi ile tedavi edildi. İskiyumdaki lezyona ise bir girişimde bulunulmadı ve konservatif olarak izlendi. Hastada ameliyat sonrası üç yıl boyunca semptom ve komplikasyon görülmedi. Sunulan olgu, hem uzun kemikte (femur) hem de düz kemikte (iskiyum ramus) eşzamanlı BKK bildirilen ilk erişkin hastadır.

Anahtar sözcükler: Kemik kisti/tanı/patoloji; femur neoplazileri; iskiyum/patoloji.

Unicameral bone cysts (UBC) are benign lesions that typically occur during growth and are usually accidentally discovered. They are frequently located in the proximal part of the humerus or the femur.[1,2] They are seldom found in adults; nevertheless, UBCs may develop at unusual sites (e.g. calcaneum, pelvis) in older patients.[1]

We report on an adult patient with simultaneous UBCs located in the proximal third of the right femur and the ipsilateral ischial ramus.

Case report

A 30-year-old, otherwise fit and well, Caucasian male patient, visited our hospital's accident and emergency

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department reporting pain (mainly during weight bearing) in the right hip area following a relatively minor injury due to a road-traffic accident. Physical examination showed a large, slightly tender on palpation, solid mass located in the proximal third of the ipsilateral femur.

Standard radiographs revealed the existence of two cystic lesions, both with a well-defined, central osteolytic area surrounded by a thin sclerotic margin (Fig. 1a). The larger one was located in the proximal part of the femur, starting just below the intertrochanteric line and extending to the metaphysis; the other was situated in the ischial ramus, starting at the mid-horizontal level of the acetabulum and extending distally. The patient was well aware of only the femoral lesion, which he had accidentally discovered at a much younger age and had not accepted any treatment since then. All his hematological, biochemical and hormonal tests were normal.

Findings of computed tomography (CT) (Fig. 1b, c), magnetic resonance imaging (MRI) (Fig. 1d, e), and technetium bone scanning (Fig. 2) supported our original (based on the plain radiographs) diagnosis of UBC. Nevertheless, since the double existence of UBC (especially simultaneously) is highly uncommon, we performed

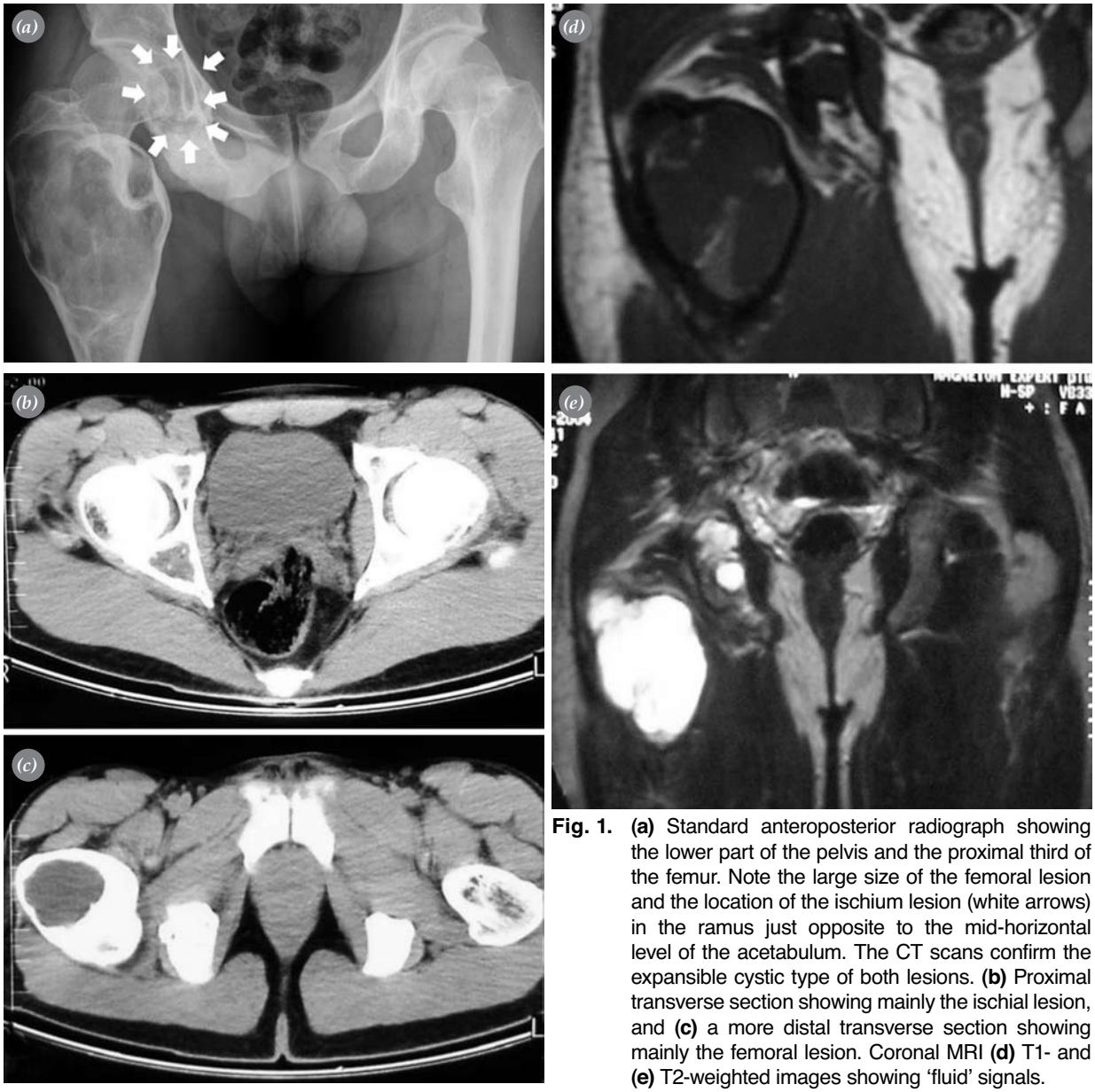

Fig. 1. **(a)** Standard anteroposterior radiograph showing the lower part of the pelvis and the proximal third of the femur. Note the large size of the femoral lesion and the location of the ischium lesion (white arrows) in the ramus just opposite to the mid-horizontal level of the acetabulum. The CT scans confirm the expansible cystic type of both lesions. **(b)** Proximal transverse section showing mainly the ischial lesion, and **(c)** a more distal transverse section showing mainly the femoral lesion. Coronal MRI **(d)** T1- and **(e)** T2-weighted images showing 'fluid' signals.

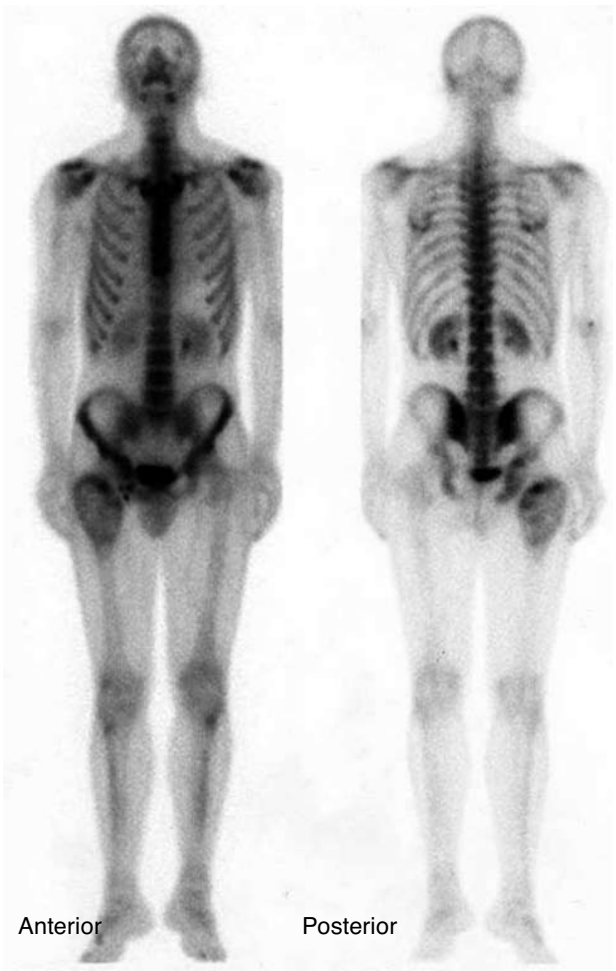

Fig. 2. The technetium bone scan showing light peripheral uptake and a 'cold' center for both lesions.

fine-needle biopsies for both lesions. The biopsy of the ischial ramus lesion was performed under local anesthesia and CT control (Fig. 3). The first attempt, however,

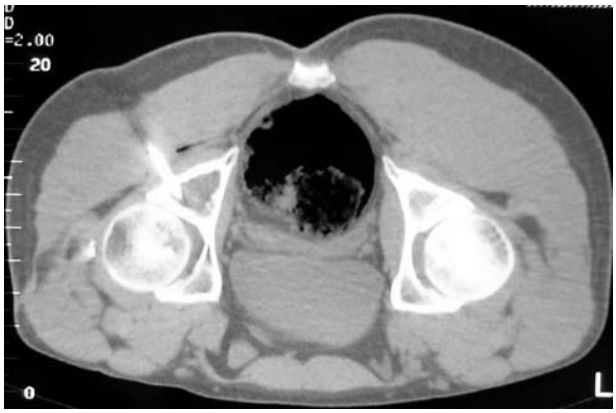

Fig. 3. Fine-needle biopsy from the ischial ramus lesion under CT control.

to enter the femoral lesion under local anesthesia was unsuccessful. As a result, an open biopsy (under general anesthesia) followed. The aspirated fluid from both cystic lesions resembled that of a synovial origin (clear serous yellow fluid) and its cytological examination failed (as expected in cases of UBC) to reveal any sinister pathology. The pathologic examination of the femoral lesion confirmed the diagnosis of UBC. Unfortunately, the specimen from the ischial ramus was not sufficient to confirm the diagnosis of UBC, even though no other pathology was apparent. We did not proceed to an open biopsy of the ischial lesion for the following reasons: all previous examinations convinced us that this was also a UBC, the pathological examination clearly showed that this was not a malignant lesion, and the patient refused to undergo an open biopsy.

The patient was informed that the development of a pathological fracture in the near future was almost inevitable due to his young age, the large size of the

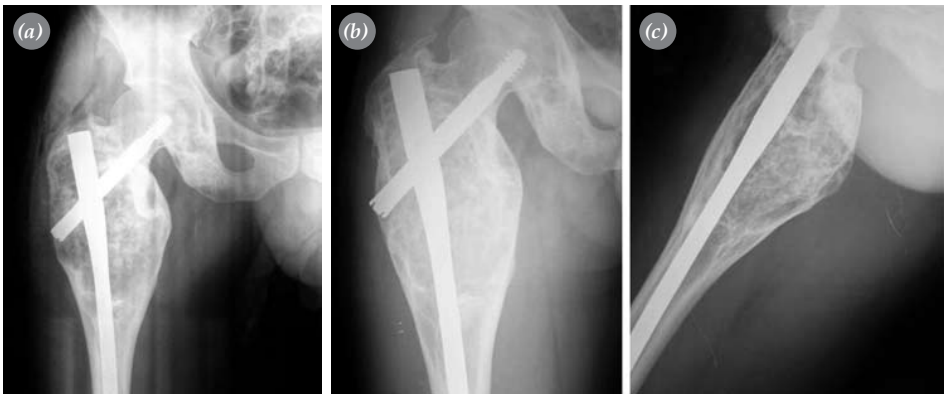

Fig. 4. **(a)** Plain radiograph of the patient at 12 weeks postoperatively. Note the long gamma locking intramedullary nail used for stabilization of the femur. **(b)** Anteroposterior and **(c)** lateral radiographs at four years postoperatively showing unchanged features of the ischial lesion and advanced consolidation of the femoral lesion.

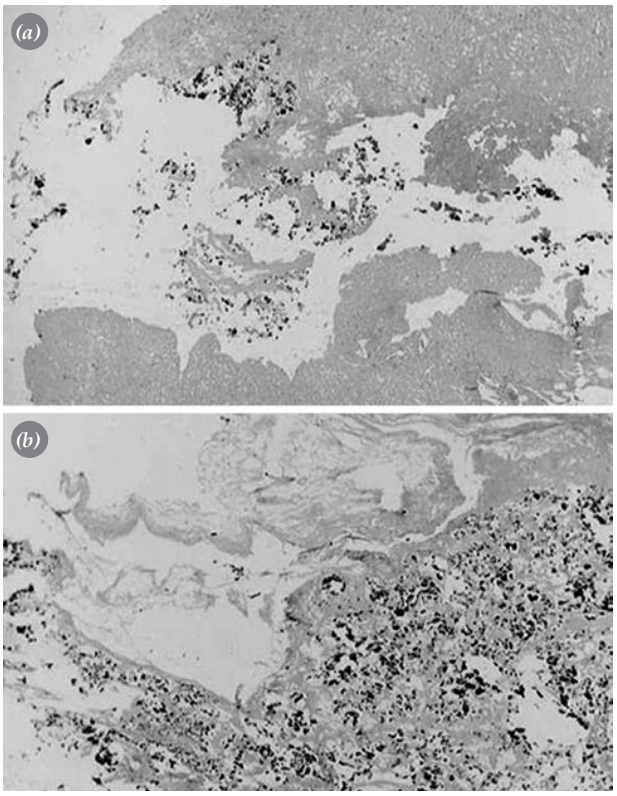

Fig. 5. Histologic sections of the femoral lesion: The cyst wall consists of fibrous tissue with fibrin deposits and calcifications. [H-E: **(a)** x 100, **(b)** x 400].

femoral lesion, and its location just below the intertrochanteric line. In order to prevent this, he agreed to undergo a wide intralesional excision-curettage of the femoral lesion, followed by the instillation of autologous bone graft (contralateral ilium) and allograft (demineralized bone graft), and the introduction of a long gamma locking intramedullary nail (Fig. 4a). The pathological examination of the excised specimen reconfirmed the diagnosis of UBC (Fig. 5). The ischial lesion remained untreated as the patient refused any further therapy. During four years of medical observation postoperatively, the patient was free of symptoms and complications (Fig. 4b, c).

Discussion

Unicameral bone cysts (also known as simple bone cysts) are benign, metaphyseal, fluid-filled, lytic lesions subsumed in the group of tumor-like lesions. They usually originate in the metaphyseal part of the long bones of skeletally immature persons, immediately adjacent to the growth plate, and may -with continuing growth- involve the diaphysis. They are 2-3 times more common in males.[2] In approximately 50% to 70% of all cases, they become clinically apparent due to a spontaneous pathological fracture, usually following a minor trauma.[2] The occurrence of a UBC in a nontubular bone is unusual accounting for about 4% to 10% of all UBCs and it is usually manifested at a higher age.[1] It seems that these cysts have a more favorable prognosis, with a less aggressive behavior and a lower recurrence rate than that of classic metaphyseal UBCs.[1,3]

There are some reports of multiple localizations of UBCs, mainly in adult patients.[1,4,5] These present as symmetrical bilateral localizations involving the calcaneus,[1,6] hamatum, and possibly the tibia.[1] To the best of our knowledge, this is the first report of an adult patient with UBC simultaneously located both in a long tubular bone (femur) and a flat bone (ischial ramus). Even though the pathological examination of the specimen taken from the ischium failed to histologically confirm our diagnosis, it is our belief that this was also a UBC. This certainty is based on the following: the location of the lesion in a flat bone in an adult patient; its clinical and other (radiographs, MRI, CT, bone scintigraphy) features; unchanged radiographic characteristics 36 months postoperatively, and absence of any other benign (e.g. aneurysmal cyst, fibrous dysplasia) or malignant pathology on pathological and cytological examinations.

Our decision not to 'press' the patient to accept an open biopsy of the ischial lesion and/or further aggressive treatment was mainly based on its benign radiographic features and (to a lesser extent) on the findings of cytological examination. Therefore, the radiologist and the orthopaedic surgeon must always keep in mind that UBC may simultaneously exist in a long tubular bone and a flat bone in adults as well, especially when dealing with diagnostic dilemmas and particularly when planning (aggressive) treatment modalities and strategies.

The actual etiology of this asymptomatic lesion remains more or less unknown, representing a difficult puzzle for pathologists, radiologists, and orthopaedic surgeons. It has been demonstrated in several reports that the cyst fluid plays an important role in the pathogenesis of UBC. Cyst fluid causes an elevation in intracystic pressure and contains proteolytic enzymes and oxygen radicals which are responsible for the degradation of bone matrix.[7] Chromosome anomalies have also been documented in UBCs.[8-10] Vayego-Lourenco et al.[8]

raised a question attributing the probable etiology of the UBC to physical trauma that may render traumatized bone regions at risk for generating chromosome alterations leading to the development of the lesion. Another quite popular theory is that a developmental anomaly in the metaphyseal veins of the affected area may be the actual cause that triggers the development of a UBC.[7]

Chromosomal alterations following a physical trauma (certainly at a much younger age) may be the actual cause for the development of the lesions in our patient, as they c located in "neighboring" parts of the skeleton. Metachronous development of the ischium ramus lesion is another interesting possibility that inevitably alters our theory about the causes of their appearance. Regardless of what the actual cause of the femoral lesion was (trauma and/or chromosomal alteration, developmental venous alteration and/or increased intracystic pressure, genetic factors, other), if we consider that their simultaneous appearance is somehow related, a connection factor must be determined. Might this be the local venous system (full of shunts) that made possible the transportation of genetically altered cells from the femoral lesion to the ischium ramus area? Or could it be the fact that both UBCs (for a considerable time period during the patient's skeletal development) were intracapsular, thus permitting the transportation of (any) pathogenetic factors from the femoral to the ischium ramus lesion? Unfortunately, these questions remain to be answered.

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