Case Report

ISSN (0):2395-2822; ISSN (P):2395-2814

Solitary Osteochondroma: Rare Occurrence A Report of Two Cases.

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Received: December 2016 Accepted: December 2016

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ABSTRACT

Solitary Osteochondroma is the most common benign bone tumor, and occurs most frequently in the proximal humerus, tibia, and distal femur. It rarely affects proximal femur and talus. Therefore, we report two cases of solitary osteochondroma, which were found at these rare sites (one at lesser trochanter and another at talus) with a brief review of literature and discussion of clinical features and management.

Keywords: Osteochondroma, exostosis, proximal femur, lesser trochanter, talus.

INTRODUCTION

Osteochondroma is also known as an osteochondromatous exostosis^[1], osteocartilaginous exostosis^[2,3] or simply exostosis, is defined by World Health Organization (WHO), as bone projections enveloped by a cartilage cover that arise on the external surface of the bone^[1]. Despite their predominant composition of bone, their growth takes place in the cartilaginous portion^[2]. Debate continues as to whether osteochondroma is a developmental disorder (pseudotumoral lesion) or a neoplasm^[1].

They present two distinct clinical forms^[3] : single lesions (solitary osteochondromas) and several lesions (multiple osteochondromas).Solitary form constitutes 10% of all bone tumors and, among these, 35% (20–50%) of the benign tumors^[1-4]. Single lesions are found in 85% of the individuals diagnosed with osteochondroma^[3]. The exostosis is commonly identified during childhood or adolescence^[1,2].

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Osteochondromas more frequently affect the appendicular skeleton (upper and lower limbs).It most frequently occurs in the distal femur, proximal tibia and proximal humerus^[3]. It rarely affects the proximal femur or the talus.

Here we report one case of such rare occurrence at each site.

CASE REPORT

Case report 1:

A 20 year old male, presented with complaints of pain and swelling in left groin and limp on left side since one year. Patient initially noticed swelling 4-5 years ago in his left groin, which was not growing in size, or causing any discomfort to the patient. 1 year ago, he started noticing pain in his left groin, which was insidious in onset, gradually progressive, non radiating, dull aching type, aggravated with sports activities, prolonged walking or standing and relieved by analgesics. He also started noticing a limp while walking on the left side.

On examination , a diffuse swelling was below gluteal fold about 10x12 cm in size, ill defined, globular, lobulated surface, bony hard, moving with the movement of femur with overlying skin being normal. External rotation was painful and restricted. No regional lymphadenopathy was appreciated and there was no distal neurovascular deficit. Skeletal survey revealed no other swelling.

Radiograph showed a large sessile mass arising from lesser trochanter of left femur [Figure 1,2].

CT scan of pelvis was done with 3d reconstruction for better delineation and extent of the tumour [Figure 3].

CT scan showed: bony outgrowth involving medial aspect of proximal shaft of left femur, which was continuous with the medullary cavity, and causing compression of gluteus medius muscle postero-

Jha et al; Solitary Osteochondroma

medially, with bony matrix showing stippled calcification suggestive of exostosis-proximal shaft of femur.



Figure-1:



Figure 2:



Figure 3:??????????????

In view of patient's complaints and taking into consideration the preponderance for malignancy, the tumour was excised. Usingmidline vertical incision, sciatic nerve was identified and protected by retraction. Tumour mass was identified, sessile in nature, arising from lesser trochanter and was excised in to along with its capsule [Figure 4,5]. The tumour along with its capsule was sent for histopathological examination and bone wax was applied over the raw bone. Wound was closed over a drain and the patient was allowed to bear weight from 2^{nd} post op day. Histopathological examination was consistent with osteochondroma.



Figure 4:



Figure 5:

Post op radiographs were taken [Figure 6,7]. Movements of hip were regained and the patient became pain free.

Follow up uptill 3 years showed no recurrence of the tumour.



Figure 6:





Section: Orthopaedics

Case report 2:

An 18 year old female, presented to our outpatient department with complaints of pain in the left ankle joint since 3 months, aggravated by walking , running and sports activities. There was no history of trauma or fever. Examination revealed a bony swelling on the anterolateral aspect of the right foot [Figure 8], around 2x2 cm in size, tender on palpation. Dorsiflexion of the ankle was painful and restricted. No other swelling was noticed on skeletal survey.

Radiograph showed a bony mass arising from the talar neck [Figure 9].

Surgical excision of the mass was done. Longitudinal incision was made and after adequate exposure a hard whitish grey mass [Figure 10] was visualized and it was excised from its base. Post op x ray showed no residual mass [Figure 11] Histopathological study of the specimen was Consistent with the diagnosis of osteochondroma. No recurrence was found till 3 yrs follow up.



Figure 8:



Figure 9:



Figure 10:



Figure 11:

DISCUSSION

Osteochondromas are the most common benign bone tumours^[5]. The cause of osteochondromas remains unknown. Based on the similarity of the cartilaginous cover of the exostosis to the growth cartilage (growth plate) of the bone, several hypotheses have been put forward, all of them relating to alterations to the growth plate 1 the lesion seems to result from separation of a fragment of growth cartilage (from the immature skeleton), which suffers herniation^[6].

Continuous growth of this loose piece of cartilage and its subsequent endochondral ossification forms a salience that projects from the bone surface, coated with a covering of cartilage^[6]. However, it is still unclear how this separation actually occurs.

Proximal femur and talus have been reported as rare sites of occurrences of these tumors.

Proximal femur was noted to be a site of occurrence in approximately 4.8% of the 313 cases described by saglik et al^[7] with lesions around hip joint forming a meagre 5.1% of all cases. At the same time talar region was noted to be involved in only 3.5% of cases in the same series^[7].

In one report of a series of 783 osteochondromas, only 15 osteochonromas were encountered in the tarsal region, and 10 of these were in the calcaneus^[8]. An osteochondromain the talus is very rare and only a handful of cases have been reported.

An osteochondroma of the talus was first reported in 1984 by Fuselier et $al^{[9]}$.

They reported solitary osteochondroma of the dorsum of thetalus in a 22-year-old female presenting with ankle discomfort. They found 2.0 cm long pedunculated osteochondroma, protruding from the dorso-lateral head of the talus with multiple toe deformities.

In 1987, Chioros et al reported an atypical osteochondroma that originated from the posterior aspect of the talus in a 34-year-old male^[10].

In 2003, Erler et al^[11] reported a case of an osteochondroma located on the dorsum of the talus, which is similar to this cases, in 6-year-old boy

Jha et al; Solitary Osteochondroma

without other foot deformities. There are very few other reports of osteochondroma in talus^[12-14].

Among solitary osteochondromas, the vast majority are asymptomatic. Symptomatic cases are often related to the size and location of the exostosis. In the immature skeleton, the osteochondroma grows slowly and progressively along with the bone involved, and it stops when skeletal maturity is Reached^[15]. In a few cases, pain of greater intensity may be present, associated with complications of a mechanical origin^[1].

The fixed anatomic effects of solitary exostoses of the proximal femur have been well described in cases involving trochanteric bursitis, external snapping of the hip, and sciatic nerve compression, hip dislocation etc leading to either local treatment or surgical excision^[16,17].

Unlike osteochondromas associated with hereditary multiple exostosis, a solitary osteochondroma in proximal femur typically does not cause coxa valga or overgrowth of femoral neck or increased femoral ante-version^[18,19].

An osteochondroma in the talus may present with variable symptoms, including Pain^[10], ankle swelling^[11], painless mass^[12,14], and a limited range of ankle motion.

Even though osteochondroma is a benign tumour, malignant transformation is one of its severe complications.

The incidence of sarcomatous change in patients with a solitary osteochondroma has been reported to be between 1% and 2%, whereas the incidence in patients with multiple hereditary osteochondromas varies between 1% to 25%.^[20-22]

Centrally located osteochondromas about the pelvis, hips and shoulders are reported to be particularly more prone to undergo malignant transformation^[23-25]. The cause may be the delayed diagnosis of the lesions in these areas^[23-25].

Of the 313 cases reviewed by saglik 2.2% (7 cases) sarcomatous degeneration were noted. Of the seven cases 2 were located in proximal femur^[7].

Since the risk of malignant transformation of these lesions are high,^[21,26] it is important to consider surgery in patients with radiographic or imaging evidence of malignant change, however, minimal. Also, delay in surgery may adversely affect the outcome. Various approaches have been described to ascess the tumour which include posterior approach, used if the tumour is projecting posteriorly, medial approach and an extensive surgical approach with dislocation of femoral Tschokanow^[27]. Tschokanow described two separate incisions in a one stage or two staged procedure to avoid rare complication of vascular damage while resecting an osteochondroma. Siebenrock and Ganz^[28] recommended a versatile surgical approach with dislocation of the femoral head for osteochondroma of the femoral neck. While this approach offers excellent exposure, there may be an increased risk of necrosis of the femoral head.

Almost all studies conclude that surgery is the mainstay of therapy for symptomatic osteochondroma. A complete excision is mandatory in the treatment of osteochondroma.

Local recurrence after excision has been reported with variable frequency approximating around 2%. Of the 114 patients presented by Humber et al^[29], there were two patients with a recurrent solitary osteochondroma are requiring a second resection who were less than 4 years of age. It has been suggested that incomplete removal of the cartilage cap is responsible for local recurrence, but young age at the time of surgery maybe an additional prognostic factor. Bottner et al reported recurrence rate of 1.8% after surgical excision^[30].

The treatment of an asymptomatic osteochondroma of the talus might be just observation. However, surgical excision is a good treatment method for a symptomatic osteochondroma of the talus, as in our case. Unlike a simple bony protrusion, an extraperiosteal complete excision is the key to complete eradication and for preventing a recurrence.

CONCLUSION

In conclusion, solitary osteochondromas rarely affect the talus and proximal femur. These are usually detected incidentally however; it can cause variety of symptoms. Patients with solitary osteochondroma at high-risk sites should be warned of malignant transformation and be thoroughly followed. Centrally located tumours have higher chances of malignancy and should be thoroughly investigated. A carefully planned surgical excision is safe and effective treatment. Major complications and local recurrence are rare if done meticulously. This case report is presented due to its rarity of occurrence.

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Jha et al; Solitary Osteochondroma

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How to cite this article: Jha V, Munde SL, Gulia A, Middha S, Lamba D, Kamra H. Solitary Osteochondroma: Rare Occurrence A Report of Two Cases. Ann. Int. Med. Den. Res. 2017; 3(1):OR01-OR05.

Source of Support: Nil, Conflict of Interest: None declared