Differential diagnosis of BPOP arising in relation to patella

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What to Learn from this Article?
In spite of some inherent weakness this publication will add to popularize one disease process (BPOP) which is believed to be rare. I consider this is not the situation. Possibly this is because of the ignorance by most of the clinician and pathologist too. Paucity of mention in text books makes the PG students unaware about the condition in D/D of osteochondroma.

Abstract

Introduction: Solitary exostosis is common at the metaphysis of long bones, and rarely may it develop in the lower pole of the patella. Usually it stops growing after skeletal maturity unless complicated. When the growth continues after skeletal maturity, other rare possibilities need to be considered such as bizarre parosteal osteochondromatous proliferation (BPOP). Though solitary exostosis is common at the metaphysis of long bones, very rarely it also develops in lower pole of the patella. Usually they stop growing after skeletal maturity unless complicated. When it starts after skeletal maturity and continues to grow, other rare possibilities like bizarre parosteal osteochondromatous proliferation (BPOP) are to be thought of.

Case Report: 21 years male student presented with anterior midline painless progressive swelling over right knee joint of one year duration which was hard, non-tender, fixed to patella but mobile with patella. X ray showed midline heterogeneously radio-opaque swelling attached to inferolateral aspect of the anterior surface of patella. Patellar outline is fully maintained except the narrow site of tumour attachment. After exposing through midline incision, the swelling was found to incorporate the patellar tendon completely and an anterior vertical midline cleavage was found. The mass was deliberately detached along the cleavage and from intact patellar tendon. Almost full range of knee movement is obtained in operation table. Immediate post operative 10° quadriceps lag was corrected with quadriceps setting exercises in two weeks time. Histopathological examination demonstrated thin layer of cartilage cover, irregular lamellar bone in deeper zone and spindle cells between them without cytoplasmic atypia. Plenty of cartilage cells in different stages of maturation are seen without column formation. Marrow elements are absent. Periosteum could not be demonstrated and there was no other evidence of malignancy. Features simulate ‘bizarre parosteal osteochondromatous proliferation’. There is no recurrence in five years of follow up.

Conclusion: When exostosis like lesions arise from unusual site and at an unusual age group, other rare conditions need to be investigated. Though the final diagnosis of BPOP is obtained after careful histo-pathological examination, the clinico-radiological findings are also relevant. As literature search indicates, this is possibly second incidence where BPOP arised from sesamoid bone and first from patella.

Keywords: Patella; Bizarre parosteal osteochondromatous proliferation; BPOP; Osteochondroma.
Introduction

Exostosis is a common benign tumour resulting from growth aberration particularly in metaphyseal region of long bones. Infrequently it arises from short and flat bones like scapula, clavicle, pelvis, rib, carpals and metacarpals, metatarsals etc [1]. Very rarely it arises from patella in its lower pole [1 to 7]. Usually it stops growing with skeletal maturity unless complicated with scomatous change. Hereditary multiple exostosis (HME) are a result of mutation of EXT gene which results in low pericellular production of heparan sulphate (HS) or it's over metabolism thus preventing normal maturation of growth plate [9,10]. Bizarre parosteal osteochondromatous proliferation (BPOP) is a rare condition that usually involves metacarpals and metatarsals, though one incidence of involvement of sesamoid bone of foot has been found in literature [8].

This case is being reported because of its rarity, evidence of continued growth even after skeletal maturity and its extent to envelopment of the adjacent patellar tendon. Differential diagnosis also discussed. Decision making whether to sacrifice the patellar tendon is also an issue.

Case Report

21 years male student presented at a rural tertiary care centre in March 2004 with anterior midline progressive swelling over right knee joint of one year duration. It started with a small hard nodule near inferior pole of right patella just lateral to the midline without any pain or constitutional symptom. During one year it progressed to the level of tibial tuberosity. It was attached to the patella but remained free from tibial tuberosity. Patellar tendon could not be palpated separately. Side to side movement of the swelling along with the patella was possible. 50% flexion was restricted where as rotations retained to its fuller extent. It was not associated with trauma.

X-ray showed midline heterogeneously radio-opaque swelling attached to inferolateral aspect of surface of patella. Patellar out line is fully maintained except the narrow site of tumour attachment. No evidence of cortical erosion, periosteal reaction or elevation. Cleavage between patella and swelling is well appreciated in the under surface [Fig 1]. Incision biopsy was planned. Through longitudinal midline incision, the mass was exposed and found a small site of attachment to the inferolateral aspect of the lower pole of patella. The patellar tendon could not be identified, as it was completely wrapped by the swelling with an anterior vertical midline cleavage [Fig 2]. Initially it was planned to excise the mass along with patellar tendon and its reconstruction. Before doing so one half of the mass was deliberately detached along the cleavage when the patellar tendon could be visible. So the entire mass was excised without damaging patellar tendon. The mass was not adherent to the tendon. It was little attenuated but integrity of the tendon was found satisfactory. Almost full range of movement could be obtained on operating table. Wound was then closed in layers leaving a closed suction drain which was removed after 48 hours.

Quadriceps exercise started on second post operative day. Normal activities were permitted after 2 weeks when stitches were removed. [Figs 3, 4] Patient was followed up at two weeks, 6 weeks and subsequently at 6 month intervals upto 5 years. Post operative, immediate 10° quadriceps lag was corrected in two weeks time. There has been no evidence of recurrence till date.

Macroscopically, the mass was covered with a thin layer of capsule and the lobulated surface was covered with thin layer of cartilage. Tumour tissue is friable and heterogeneous. Histopathological examination demonstrated thin layer of cartilage cover, irregular lamellae bone in deeper zone and spindle cells between them without cytoplasmic atypia. Plenty of cartilage cells are seen without column formation. Marrow elements are absent. Periosteum could not be demonstrated and there was no evidence of malignancy.

Discussion

Solitary exostosis in patella, as reported in literature, is very rare. Singh J et al (2009) [1] reported 26 neoplastic patellar lesions without any incidence of exostosis after a database study of four...
demonstrating space between tumour and cortex in certain stages of the disease and also heterogeneous character of the mass itself. Inherent weakness of the article is non application of these modalities due to nonavailability at that stage. But per-operative and macroscopic observations could demonstrate them. Microscopically, it is composed of hyper cellular cartilage with basophilic tinctorial character with evidence of calcification and ossification. Trabecular bone matures deeper zones. The presence of spindle cell the inter-trabecular space creates confusion with parosteal osteosarcoma and myositis ossificans traumatic. But in this case they are without hyperchromatosis and cytologicalalatypia. Thinning of cartilage layers are usual and more so in long standing cases[12 to 16]. These features are observed in the present case. 50% local recurrence is reported in literature [13]. But this patient did not have recurrence in 5 years.

BPOP, Turret exostosis and florid reactive periostitis are belied to represent different stages of a posttraumatic proliferative process [17]. Turret exostosis is a benign osteo-cartilaginous lesion. By the influence of mild trauma reactive periostium was believed to produce such tumour like mass[17]. Central area of mature bone from endochondral ossification with thin hypocellular peripheral rim of cartilage and absence of periostium are microscopic features. No history of trauma can be obtained in this case.

Osteochondroma or BPOP in unusual site may represent focal lesion of heterotrophic ossificansatraumatic and diffuse idiopathic skeletal hyperostosis (DISH) also known as Forestier's disease [19, 20]. Since there is no multifocal involvement they are excluded.

The disposition of mass resulted in incorporation of the tendon fully within the mass leaving an anterior longitudinal cleavage. Mass including tendon excision can be confidently avoided if preoperative biopsy and MRI studies are done. In this case same thing has been done on clinical basis only.

**Conclusion**

Exostosis like lesions when arising from an unusual site and at an unusual age group, other rare conditions need to be thought of. Though the final diagnosis of BPOP is obtained after careful histopathological examination, the clinic-radiological findings are also suggestive. As evident from literature search; this is possibly a second incidence where BPOP has arisen from sesamoid bone and first for patella.

**Clinical Message**

Managing such a lesion one should go for pre-operative biopsy and MRI study to avoid sacrifice of patellar tendon where reconstruction is difficult and morbidity is high.

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