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A CASE OF BIZARRE PAROSTEAL OSTEOCHONDROMATOUS PROLIFERATION (NORA'S LESION) PRESENTING AS TRIGGER FINGER

Orthopaedics

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ABSTRACT

Introduction: Bizarre parosteal osteochondromatous proliferation (BPOP) is a rare benign, locally aggressive bony lesion mainly involving the bones of hands and feet. Due to its rarity, various questions are still unanswered about its aetiology, presentation, diagnosis and natural history of this condition.

Case presentation: Current report presents a case of BPOP involving the base of proximal phalanx and presenting clinically as a trigger finger in a 26 year old white female. It describes the initial misdiagnosis at the primary care level, radiological and histological findings and the final outcome of the patient. It counters the current belief that BPOP never involves the surrounding soft tissues.

Conclusion: BPOP still poses a challenge to the treating surgeon due to its rarity and varied presentations. This case highlights that our current knowledge about the condition is incomplete and it can involve the surrounding soft tissues which has never been reported before.

KEYWORDS

Bizarre parosteal Osteochondromatous proliferation; Nora's lesion; trigger finger; benign bone tumor.

Introduction

Bizarre parosteal osteochondromatous proliferation (BPOP), also known as Nora's lesion is a rare benign osseous lesion that presents as exophytic cortical growth consisting of bone, cartilage, and fibrous tissue primarily involving bones of hands and feet. However, lesions in the long bones, skull, maxilla and metatarsophalangeal sesamoid have been reported [1,2]. It was first described by Nora and colleagues in 1983 [3]. In the hand it usually affects the proximal and middle phalanges but can also involve metaetacarpals less frequently. It can occur at any age but mainly affect people in their 20s or 30s [4]. There is no sex predilection [3]. BPOP can be frequently misdiagnosed due to its varied clinical presentation and close resemblance to Osteochondroma histologically [3, 2]. It is locally aggressive lesion but no metastasis has been reported [4]. The local recurrence rate has been reported between 20%-55% in different studies [3,2,5]. In 2001, Horiguchi et al [6] suggested that BPOP represents a proliferative reaction secondary to periosteal trauma or ischemia as they identified the processes occurring in a BPOP lesion are similar to those of endochondral ossification in the normal growth plate, indicating a reactive/reparative process after periosteal injury. Dorfman and Czerniak [7] also suggested BPOP as a part of a spectrum of reactive lesions while, on the other hand; Zambrano et al. [8] suggested that BPOP could represent a neoplastic process.

Fewer than 170 cases have been reported in the literature to date but none has been reported to present as a trigger finger. The following case report describes a case of this rare lesion with its unique clinical presentation, radiologic and histologic findings and final outcome.

Case presentation

A 26 year old right hand dominant female was referred to the Orthopaedic clinic for evaluation of swelling over the base of left ring finger. She first noticed the lump about a year before being seen in the clinic. There was no history of any preceding trauma. The swelling gradually grew in size and was associated with occasional night pain. She had noticed that her finger was locking when she tried to flex it and she had to unlock it by pushing it straight using the other hand. By the time she was seen in the clinic, she was unable to straighten the finger fully.

In physical exam, she was noted to have a fixed flexion deformity of about 20 degrees at PIP joint but she could flex the finger fully. There was a hard lump over the volar aspect of MCP joint of the left Ring finger, just distal to distal palmer crease. The lump was non tender to touch, fixed to underlying bone and over lying skin was normal. Both the digital neurovascular bundles were intact and no regional lymphadenopathy was noted.

Radiographs of the hand revealed soft tissue swelling and some calcification within the swelling (figures 1 & 2). MRI scan showed that the lump was originating from the base of proximal phalanx and axial scans did not show any continuity with the medullary cavity (Figure 3). It also showed that it was closely related to the flexor sheath.

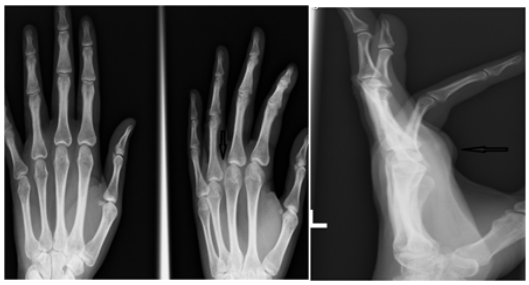


Figure 1& 2: AP, oblique & lateral Radiographs of hand showing the bony lesion (marked by arrow)

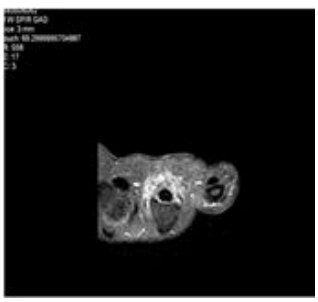
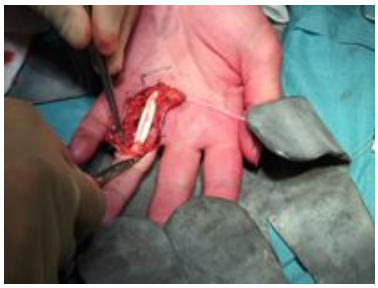


Figure 3 : MRI image (crossectional view) showing the mass encircling the flexor tendons of the finger.

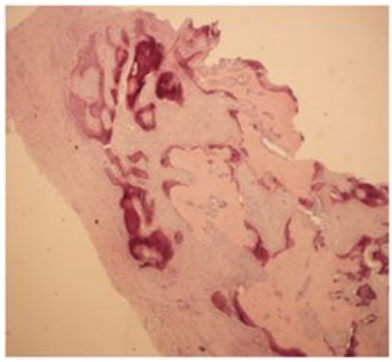
Surgical excision was discussed with the patient and was planned under general anaesthetic with Brunner incision for the skin. Intra-operatively it was noticed that the mass was completely encircling the flexor tendons and the A1 and part of A2 pulleys were involved, making a tight band around the tendons and preventing the gliding of the tendons within the pulleys, resulting in the flexion deformity (figure 4). The mass was completely excised along with the complete excision of A1 and partial excision of A2 pulley (figure 5). This resulted in correction of fixed flexion deformity and full range of movements of the MCP and IP joints of the finger.

Figures 4: Intra-operative picture showing the bony mass completely covering the flexor tendons (marked by arrow)

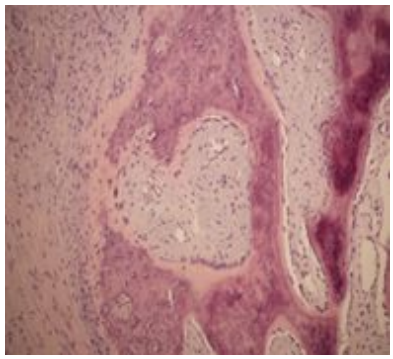


Figures 5: Intra-operative picture showing the freed up flexor tendons after removal of the mass with A1 and A2 pulleys

Histology of the tissue revealed an Osteocartilaginous lesion with "bizarre" chondrocytes consistent with BPOP (figures 6 & 7).



Figures 6: Low power histologic image showing cartilage with irregular calcification. The cartilage can be seen undergoing endochondral ossification



Figures 7: Medium power histologic image showing cellular cartilage with uneven calcification along with fibroblastic tissue. The new bone formed by enchondral ossification is lined by benign osteoblast

Patient made an uneventful recovery and attained full range of movements of the finger with no recurrence at one year.

Discussion

BPOP is a rare mineralizing mesenchymal lesion that typically affects the surfaces of bones in the hands and feet, usually the proximal, middle phalanges, and the metacarpal and metatarsal bones [9]. It is a benign but locally aggressive fibro-osseous mass that has striking clinical similarities with osteochondroma and periosteal chondroma. There are unresolved issues about this rare disease regarding its aetiology, diagnosis, and treatment. On one hand it has been described as a reactive lesion. Dorfman and Czerniak gave a "unitary hypothesis" and proposed that BPOP, florid periostits and Turret exostosis are all part of the same lesion spectrum [7]. On the other hand it has been described to represent a neoplastic lesion. Zambrano et al suggested that BPOP could represent a neoplastic process [8]. Further work by Nilsson et al & Endo et al supported this [9,10].

Radiographically these lesions seem to originate from periosteal aspect of an intact cortex of the affected bone. There is rarely any continuity between the lesion and the underlying medullary cavity in contrast to Osteochondroma where the presence of continuity between the lesion and the underlying medullary cavity is a key radiographic feature.

On MRI, BPOP has low signal intensity on T1 weighted and high signal intensity on T2 weighted sequences and short T1 inversion recovery (STIR) sequences. Torreggiani et al also suggested that the lesion never involves the surrounding soft tissue [11]. On the contrary this case report clearly demonstrates involvement of soft tissues (flexor pulleys) which is against the current belief.

Surgical excision is the treatment of choice for BPOP lesions [2, 12]. BPOP has high rate of recurrence with many studies showing recurrence rate of up to 50% within 2 years of excision [3]. Wide excision lowers the risk of recurrence as opposed to intra-lesional excision but it brings with it the risk of leaving some form of functional restriction [13].

This case report stresses upon the above fact that our current knowledge regarding BPOP is patchy and also, that these rare lesions can present in bizarre ways and can be misdiagnosed.

Conclusion

BPOP is a rare lesion and can present a challenge to the treating surgeon with regards to its diagnosis and treatment. At the present time, there is no standardized screening protocol or follow-up regimen given its rarity. Careful assessment of clinical features, imaging and histology may prevent misdiagnosis and help in optimal treatment and follow-up of these patients.

Take home message: BPOP should be considered as a differential diagnosis, if dealing with a bony mass involving the soft tissues in bones of hand and feet.

Consent

Informed consent was taken from the patient involved in this case report.

Competing interest

The author declares that they have no competing interests.

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