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Case Report

CALCIFYING APONEUROTIC FIBROMA: A CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Calcifying aponeurotic fibroma, also called juvenile aponeurotic fibroma or Keasbey tumour, is a rare slow growing myofibroblastic tumour. It is most commonly seen in children and young adults in the extremities like the hand or foot. This tumour has a 50% chance for recurrence. This is a case of a 22 year old girl who presented with a swelling in the dorsal aspect of right index finger since 1 year. The lesion was excised and sent for histopathological examination which showed a lesion composed of cartilage, areas of calcification with osteoclast like giant cells and palisading fibroblasts. A diagnosis of Calcifying aponeurotic fibroma was made.

INTRODUCTION

Calcifying aponeurotic fibroma is a rare tumour and only 150 cases have been reported in the literature. This tumour has a 50% chance of local recurrence as reported by Okhwa kim *et al.,* 2004 .The cell of origin is believed to be fibroblastic or myofibroblastic (Jun Nishio *et al.,* 2014). It was first reported by Keasbey in 1953 and initially called juvenile aponeurotic fibroma. In 1961 Keasebey and Fanselau found that it was not only seen in children but also in adults, hence he coined the term aponeurotic fibroma. In 1964 Litchenstein and Goldman defined it as the cartilage analogue of fibromatosis. In 1973 Iwasaki and Enjoji defined it by the term "Calcifying aponeurotic fibroma". This term aponeurosis was used because of the transition from fibrous to fibro cartilage at the region where the tendon/ligament/aponeurosis attaches to the bone (Melike Oruc, 2007). The most common locations are the hand, forearm, foot, leg. Other locations include arm, neck, abdominal wall, lumbar, parvertebral area (Ralph *et al.,* 2001) and mandible (Okhwa kim *et al.,* 2014). This tumour has a predilection for males (70%)(Weiss *et al.,* 2008) and usually seen affecting individuals with a median age of 12 years (Ralph *et al.,* 2001). This tumour rarely exceeds 3 cm in the greatest dimension.

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Most of these lesions are painless, however a few cases with pain have also been reported. A few cases with malignant transformation have also been reported in the literature. Hence it is very essential to completely excise the lesion and follow up the patient to rule out any possible chances of recurrence.

Clinical details

A 22year old female came to the general surgery OPD with complaints of a swelling in the dorsal aspect of the right index finger near the nail bed since 1 year. The swelling was measuring approximately 2/2cm, (Figure 1). An X ray of the right hand revealed a dense soft tissue nodule in the periarticular region with pressure erosions in the distal interphalangeal joint of the right index finger, (Figure 2). A transverse elliptical incision was made over the swelling, it was excised in total under digital block and sent for histopathological examination. Grossly we received multiple skin covered, hard tissue bits which were gritty to cut. It was altogether measuring 1.8/1.7/0.8cm.It was decalcified. Haematoxylin and eosin sections studied showed skin with a poorly circumscribed lesion in the dermis and subcutaneous tissue composed of nodules of cartilaginous and calcified tissue surrounded by plump fibroblasts and occasional osteoc last like giant cells, (Figure 3,4). No evidence of atypia, mitosis or necrosis was seen in the sections studied. Hence a histopathologic diagnosis of calcifying aponeurotic fibroma was arrived at.

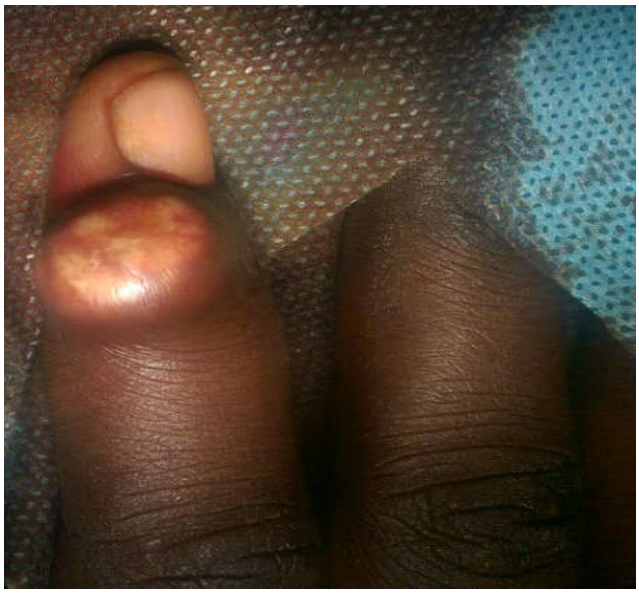

Fig. 1. An irregular swelling in the right index finger at the distal interphalangeal joint measuring 2/2cm

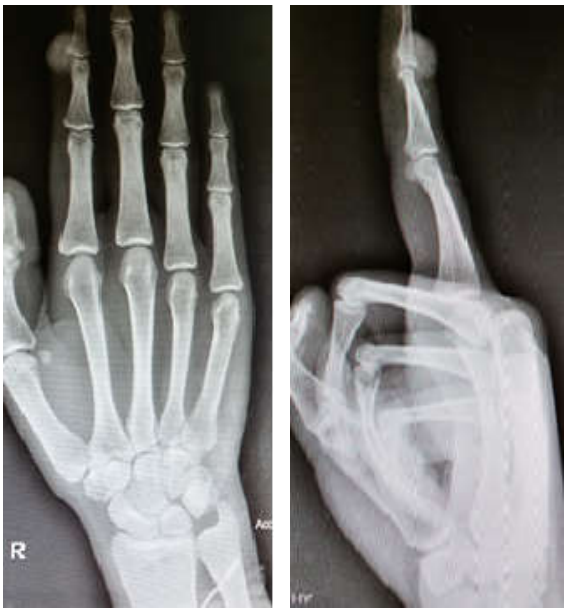

Fig. 2. X ray AP and lateral view showing a dense lesion in the periarticular region of the distal interphalangeal joint of right index finger

DISCUSSION

Calcifying aponeurotic fibroma as reported in the literature is a very rare and recurrent tumour. It is sometimes under diagnosed because of its rarity. Seventy percent of the cases are seen in males (Weiss, 2008) but in our case it was seen in a female. Though this lesion is more commonly seen in the younger ages, the median age at the time of diagnosis is 12yrs (Ralph *et al.,* 2001).The oldest reported case is from a 69 year old female(Jun Nishio *et al.,* 2014). It presents mostly as a painless lesion but 2 cases have been reported in the literature with mild pain (Ralph *et al.,* 2001) and 3 cases with major pain(Ralph *et al.,* 2001) .77% cases has been reported in the hand like that seen in our case compared to 13% in the foot(Ralph *et al.,* 2001).

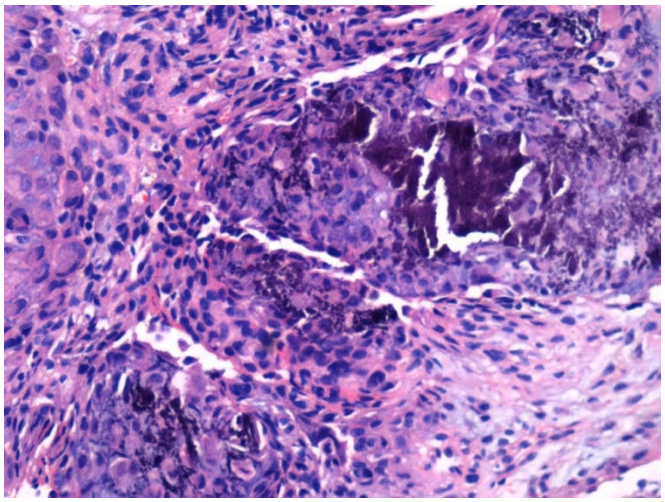

Fig. 3. 200x, H and E sections shows areas of calcification with palisading fibroblasts in the periphery

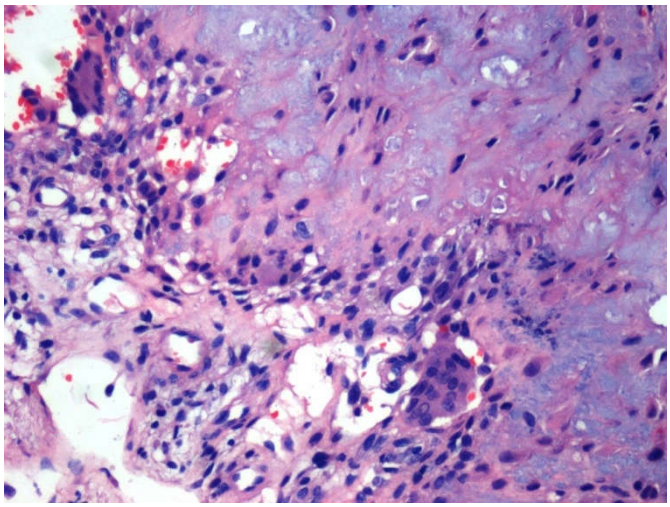

Fig. 4. 200x, H and E section shows osteoclastic giant cells with chondroid areas

The characteristic histopathological feature of this lesion includes fibroblast proliferation with hyalinised stroma , nodular deposits of calcification and cartilaginous metaplasia. This is a biphasic lesion. The initial phase, which is mostly seen in infants /children is characterized by diffuse cellular proliferation.

The late phase of this lesion has the following features: it is well demarcated, less cellular, has prominent calcification and increased collagen between fibroblasts .It denotes a maturation process seen mostly in adults. Our case represented a late phase lesion. As reported in the literature, immuno histochemical staining in the cellular component shows positivity for vimentin and smooth muscle act in (Jun Nishio *et al.,* 2014). X ray in the early phase shows a poorly defined lesion with fine stippled calcification and in the late phase shows extensive calcification, but MRI is more precise in delineating the extent of lesion and useful in planning the extent of excision. MRI findings include, low intensity lesion in T1 weighted images and high intensity lesion in T2 weighted images (Jun Nishio *et al.,* 2014). Ultrastructural study have demonstrated fibroblasts, myofibroblasts and

cartilaginous components of the tumour (Hiroshi Iwasaki *et al.,* 1983). The recurrence as noted by Allen and Enzinger (2008) is 10 out of 19 cases ,in whom recurrence occurred between 1 month to 11 years after initial excision .Other cases of recurrence was reported by Mehmet *et al.,* 2013 and Giuffre *et al.,* 2011. One case of malignant transformation was reported (Amaravati *et al.,* 2002) and another case of juvenile aponeurotic fibroma with disseminated fibrosarcoma was also reported (Lafferty *et al.,* 1986). Treatment involves excision and re excision in cases of recurrence.

Conclusion

To conclude, calcifying aponeurotic fibroma was presented here for its rarity and as it was observed in a female patient as opposed to males seen commonly in the literature .It must be considered in the differential diagnosis of calcified soft tissue lesions of the extremities and elsewhere too.

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