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Case Report

Painful and rare tumor of the pediatric hand – calcifying aponeurotic fibroma

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ABSTRACT

Pediatric hand tumors are rare and the commonest include ganglion cysts and giant cell tumors. Calcifying aponeurotic fibroma (CAF) is a rare, locally aggressive, benign fibroblastic tumor occurring usually in the palms of the hands and soles of the feet in young and adolescents under 20 years of age. Clinically, it presents as a painless palpable mass. Pathologically, it is characterized by fibroblast proliferation with an indistinct border and calcification. This soft tissue tumor typically infiltrates into the surrounding fascia or muscle and has a predilection for local recurrence after surgical excision of around 50%. Only 4 cases of pediatric CAF have been reported from India till date and all of them were painless swellings, at a site other than the hand. We present the first case of painful pediatric CAF of the hand in an 8 year old male patient. Clinical findings and imaging features (X-ray, USG, MRI) are presented. Patient underwent excision biopsy with complete removal of tumor and histopathological features were consistent with CAF. Patient was followed up for a period of 1 year post-resection and showed good function and no recurrence.

Keywords: Pediatric hand tumors, Calcifying aponeurotic fibroma, Pediatric CAF, Hand swelling, Fibroma, Benign fibromatoses

INTRODUCTION

Although many varieties of hand tumors exist, pediatric hand tumors in general are quite rare.[1] The vast majority are benign. Ganglion cysts are among the most commonly encountered pediatric hand masses. Giant cell tumors of the tendon sheath (GCT) are the second most frequent hand tumors.[2] Other more common, benign soft tissue tumors in the pediatric population include hemangiomas, and fibromatous lesions.[1]

CAF is a rare, locally aggressive fibroblastic lesion that most commonly affects the palms of the hand and soles of the feet in young children.[3] It commonly occurs in the first or second decade of life (8-14 years peak incidence) with higher prevalence in males over females[4]. It is one of the few fibrous lesions that can calcify.[4] It was originally described and referred to as juvenile aponeurotic fibroma by Keasbey in 1953.[5] The tumor presents as a very poorly circumscribed painless lump with a high tendency for recurrence.[6]

Pediatric CAF is a very rare tumor in India with only 4 cases reported in literature till date, none located in the hand and all of them painless.[7-10] We report the first case of a painful pediatric CAF of hand in an 8 year old male patient.

CASE REPORT

An 8-year-old boy presented with a solitary swelling on left palm since 2 years (Figure 1), which was painless to

begin with, but became so painful since 2 months that his school teacher alerted his parents about him not using the hand to pick up objects or play. Patient gave no history of trauma or previous surgery.

On clinical Examination, there was a solitary swelling in the left palm near the distal palmar crease between 4th and 5th metacarpals. It was 1.5×1 cm in size, firm, tender and mobile. Skin over swelling was normal. Range of motion in both fingers was unrestricted with no obvious neurovascular deficit.

Plain radiographs (Figure 2) of the left hand revealed no bony involvement and routine blood tests were insignificant. Ultrasonography of soft tissue showed evidence of focal proliferation of cutaneous layer in palmar aspect of left hand with surrounding soft tissue, underlying bony echoes and surrounding vascular structures appearing normal. On MRI (Figure 3 and 4) a well-defined T1 isotense and T2\PDFS hyperintense lesion measuring ~12×9 mm was noted in the subcutaneous plane of palmar aspect of left hand between head of 4th and 5th metacarpal bones. Posteriorly lesion was in close proximity to flexor tendon of left 5th digit and 4th lumbrical muscle. Bony structures of hand were in normal alignment with normal signal intensities.

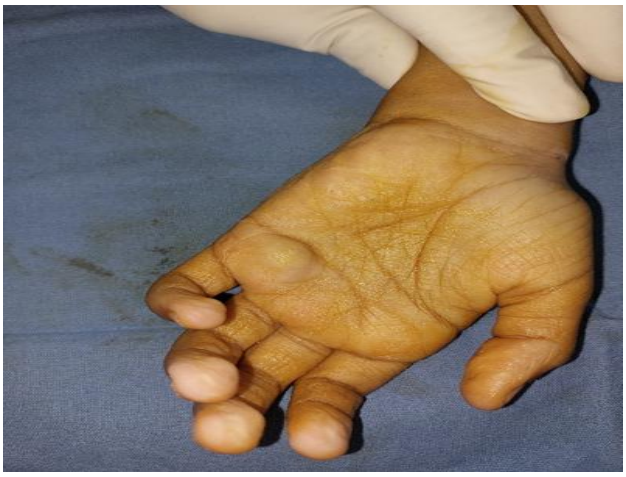

Figure 1: Pre-operative clinical photo.

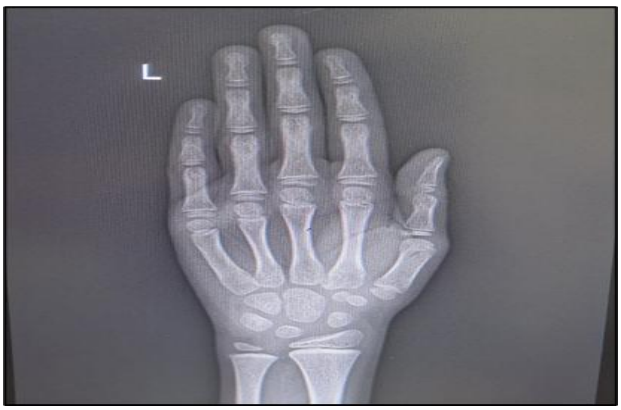

Figure 2: Pre-operative X-ray of hand.

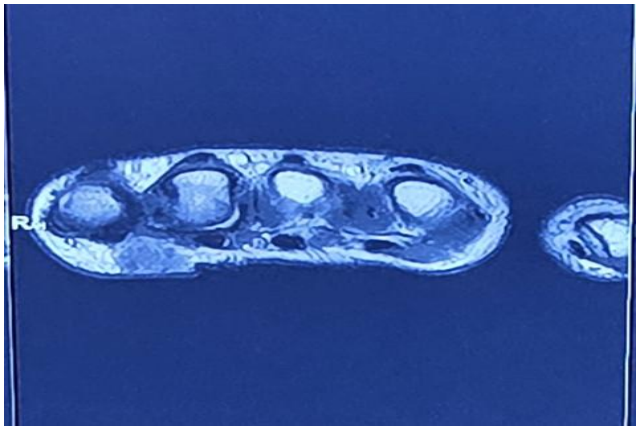

Figure 3: MRI (T1 weighted).

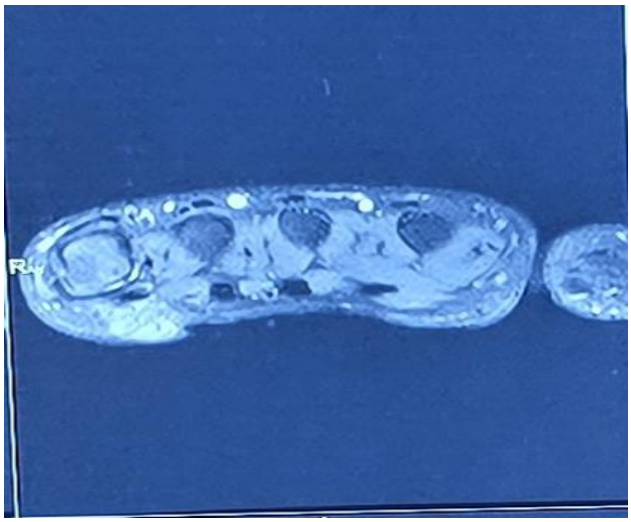

Figure 4: MRI (T2 weighted).

Patient was taken up for excision biopsy under general anesthesia (Figure 5-7). A chevron shaped incision was made on the left palm between the 4th and 5th metacarpal heads. The tumor was visualized, found to be loosely adherent to the neurovascular bundle, and was excised completely with a healthy margin, while taking care to safeguard the neurovascular bundle. On visual inspection, the mass appeared yellowish- white with speckled areas of calcification.

Histopathological examination (Figure 8) revealed the presence of fascicles of benign spindle shaped cells having bland oval shaped nucleus with moderate amount of cytoplasm, interspersed with areas of hyalinization and calcification. These areas were surrounded by plump spindle and epithelioid cells with multinucleated giant cell. No evidence of atypia/ mitosis/ necrosis. These findings were consistent with the pathological diagnosis of CAF.

Post-operatively patient had no neurovascular deficit and movements were normal. Patient was followed up for a period of 1 year after surgery and showed no evidence of recurrence (Figure 9 and 10).

Figure 5: Intra operative-exploration of the tumor.

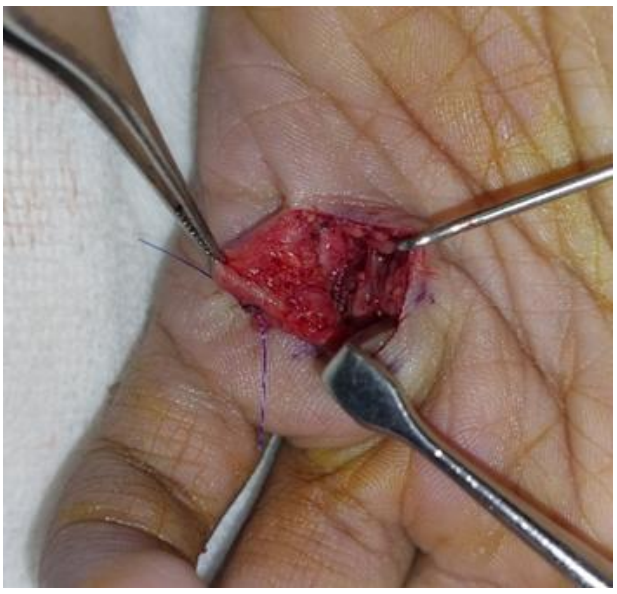

Figure 6: Post resection of tumor (neuro vascular bundle intact).

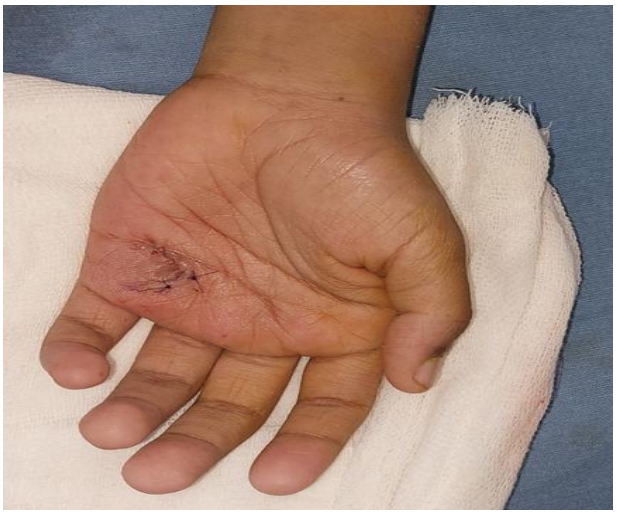

Figure 7: After wound closure.

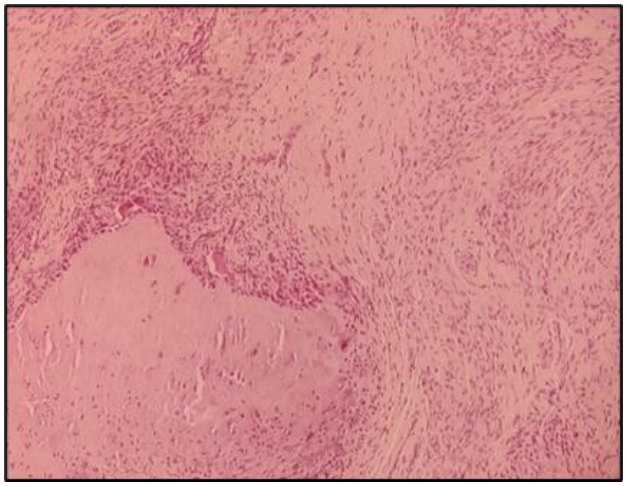

Figure 8: Histopathological examination.

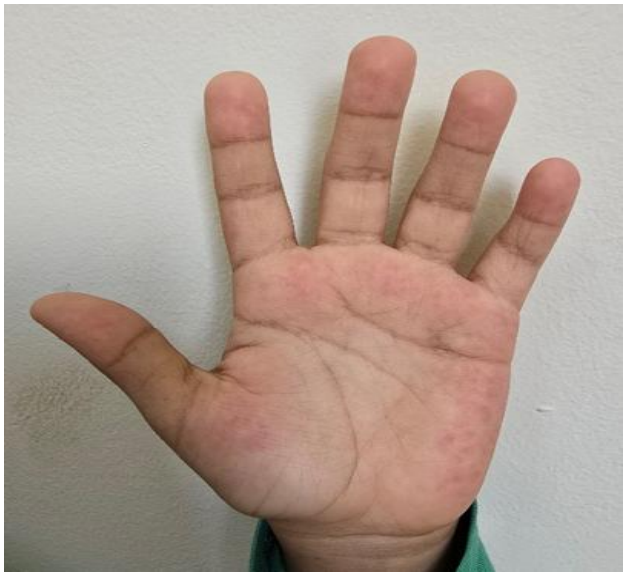

Figure 9: 1 year post operative.

Figure 10: 1 year post op normal hand function.

DISCUSSION

Musculoskeletal fibromatoses represent a wide spectrum of fibroblastic and myo-fibroblastic neoplasms with similar pathologic appearances and variable clinical behavior. These lesions can be categorized by location (superficial or deep) or by the age group predominantly affected. Superficial fibromatoses in adults (palmar and plantar) and children (CAF, lipo-fibromatosis, and inclusion body fibromatosis) are often small slow-growing lesions; their diagnosis is suggested by location.[3] The second subdivision includes aggressive fibromatoses, plantar-palmar fibromatosis, and nodular fasciitis.[11]

CAF is usually found superficially in subcutaneous tissue or in deeper musculofascial and para-skeletal tissue. The most constant clinical findings include a slow-growing, nontender, firm, mobile mass less than 3 cm in diameter in the distal portion of the extremities in children and adolescents.[12] The tumor is painless and rarely causes complications such as restriction of range of motion.[4]

Radiographic features are also non-pathognomonic. No calcification or only smudge-like radiopacities may appear initially. However, lesions that have been present for years may exhibit large calcified areas. Ultrasound examination excludes the more likely diagnosis of a ganglion, indicating a solid mass mainly fibrous with foci of calcification.[12] Because of the common nature of GCT of the hand, the anatomic location of the tumor in our patient, and its association with the flexor tendon sheath, this lesion could be wrongly presumed.[2]

MRI can provide useful findings for differentiating between GCT and CAF. CAF shows heterogeneous gadolinium enhancement, speckled calcifications, and ill-defined margins, whereas GCT shows lobulated well-defined margins, close association with tendon sheath and uniform gadolinium enhancement.[13]

As with any mass of unknown etiology, malignancy should always be considered. However, in the pediatric hand, malignancy is exceedingly rare accounting for approximately 1-2% of surgically addressed masses. Fibrosarcoma is one of the most common malignant masses found in the pediatric hand though synovial sarcoma, epithelioid sarcoma, melanoma, clear cell sarcoma malignant schwannoma, parosteal sarcoma, and angiosarcoma have been described, and should thus be considered entities that require a differential diagnosis.[1,13] In general, malignant soft tissue tumors reveal hetero-generous findings caused by various biological processes such as necrosis, hemorrhage, fibrosis, hyalinization, and infiltration. Internal irregularity, often evident on T2-weighted images and T1-weighted post gadolinium images, is the most specific criterion in the diagnosis of a malignant tumor.[13]

The definitive diagnosis is solely based on histopathological biopsy. Lesions typically contain two components: (a) fibromatosis-like spindle-shaped cell elements and (b) distinctive, round or oval, fibrocartilaginous, often calcified foci composed of epithelioid cells, as in our study.[3] Although the tumor is benign, it tends to invade surrounding fat and engulfs nerves, vessels and muscle fibers, without destroying them and has a propensity of local recurrence after excision.[4] Biopsy is also important to allow differentiation from synovial sarcoma, which can have similar imaging features and which may affect the hands and feet of younger patients.[3]

Despite the histologic appearance of CAF resembling an aggressive fibroblastic lesion, its biologic behavior is benign, with a tendency for local recurrence, with exceedingly rare cases of reported malignant transformation and subsequent metastasis.[6]

Recommended treatment is conservative local excision and function-preserving surgery, even if the tumor resection is incomplete/tumor recurs locally, because of the low biologic potential of this tumor.[3,6] Local recurrence is common (52% of all cases) and develops more frequently in patients younger than 5 years of age, typically within the first 3 post-operative years. Re-excision of local recurrence is usually performed, rather than more aggressive measures, to preserve function.[3]

An important message is that the neurovascular bundles should be preserved even if they are found to be invaded by the tumor. Different studies have proved that radical excision should be avoided as the natural course could be benign, considering that it matures and becomes more nodular with time, showing a less aggressive behavior.[7] In a case series by Corominas et al a patient underwent an incomplete resection as it was infiltrating the neurovascular bundles, albeit the patient remained symptom-free more than 3 years after resection, with no evidence of tumor regrowth.[7]

CONCLUSION

Thus, we present a case of an 8-year-old boy with a painful rare tumor of his left hand- calcifying aponeurotic fibroma. The awareness of the natural history of the lesion, combined with radiological correlation (including Ultrasound and MRI) can provide clues to the condition, exclude concerns related to malignant tumors and avoid the need for aggressive treatment.

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