

CLINICAL CHARACTERISTICS AND A BRIEF REVIEW OF OUTCOMES OF GIANT CELL TUMOR OF TENDON SHEATH: A CASE SERIES AND LITERATURE REVIEW.

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Abstract

Recurrence in a non-malignant condition like giant cell tumor of the tendon sheath (GCTTS) is still an unresolved issue. After fine needle aspiration cytology confirmation, we operated on twenty-one cases of GCTTS and did complete excision of the lesion including the satellite nodules. Recurrence was noted in two cases which were successfully treated by a second-wide excision. The predictive factors for recurrence were preoperative diagnosis and meticulous surgical excision including usage of a magnifying loupe for better visualization. 18 patients who were operated between year 2015 and 2020 (2 males, 16 females with a mean age of 29, ranging from 10–53 years) were included during this study. The lesions were more common over the thumb (n=10), ring finger (n=2), index finger (n=2), and over the hand (n=4). Al-Qattan classification was used to classify these lesions. The most common presentation was with a mass over the hand. Radiological changes were seen in only 4 cases in the form of bony indentation. In 3 out of the 18 cases FNAC were inconclusive. Preoperative planning aided by tissue diagnosis, wide surgical exposure and complete excision after meticulous dissection with the help of magnification are imperative for a successful outcome in GCTTS.

INTRODUCTION

Giant cell tumor of tendon sheath (GCTTS) is the second commonest tumor of the hand.^[1]Clinically it is a slow growing soft tissue tumour, taking months or years to develop. Common etiological factors include trauma, inflammation, metabolic disease and even neoplasm.^[2,3]Proximity to the distal interphalangeal joints, pressure erosions in the radiographs, presence of degenerative joint disease, increased mitotic activity, and type 2 lesions as described by Al-Qattan are many proposed factors leading to recurrence.^[1,3-5]Complete surgical excision with removal of all satellite nodules if present is the only consistent finding proposed by various authors in preventing recurrence.^[6-8]The use of an operating microscope,^[7] and or magnifying loupe 6 helps in radical excision. Few authors went on to the extent to suggest that the surgeons should take the help of a pathologist during surgery with frozen section,^[2,6] and or a fine needle aspiration cytology (FNAC).^[9]

MATERIALS AND METHODS

This is a multi-centric retrospective observational study. We consulted our institutional review board and because of the retrospective nature of the study no formal ethics approval was required. This research conforms to the Declaration of Helsinki as revised in 2008.

18 operatively treated cases of GCTTS between May 2015 and December 2020 were included in the study. Part of the cases were done in Palakkad government medical college, Palakkad and the rest from Dr Moopen's Medical College, Wayanad. After confirmation with FNAC and using magnifying loupe for meticulous dissection, a total of 21 cases were operated by the senior surgeon. Later all these patients were asked over telephone to come for follow-up. Follow up ranged from 3–9 years (mean 4.3 years). Three cases were excluded from the study as these were not available for follow up.

Patients' age ranged from 10 to 53 years (mean age 29.5 years). Of the 18 cases available for follow up

ten involved the thumb, two each in the index and ring fingers, and four were over the hand. Antecedent trauma history was present in only three cases (Fig. 1,2 and 3). Routine radiographs of the involved part and an ultra sound scan were done for all patients prior to FNAC (Fig.4). The FNAC procedure was performed by the treating surgeon. The slides were preoperatively analyzed by the histo pathologist for any giant cells so that extra caution could be taken during the dissection (Figs. 5 and 6). All cases were operated by the treating surgeon under Bier's block, under tourniquet control and using a magnifying loupe. Total excision of the tumor with it's retaining capsule and a margin of normal tissue was done in all cases. Again the operative field was searched for any satellite lesions or daughter cysts using a magnifying loupe. Histopathological examination of the entire specimen was then done, and the margins were observed for clearance.

Classification suggested by Al-Qattan was used to classify the tumors (Table 1). Single nodule surrounded by one thick pseudo capsule were seen in eight cases (Al Qattan I a), six were single nodule within a thin capsule (I b). Two lesions were found one in the ring finger and the other in the thumb were multi-lobulated surrounded by a common pseudo-capsule (I c). Another two cases were found in relation to the flexor carpi ulnaris and was diffuse with multiple granular like lesions without a pseudo-capsule (II b).

Two cases of recurrence were seen during our follow up period, which recurred after a period of 2 months. The masses were over the dorsum of the thumb near the interphalangeal joint, and clear indentation of the terminal phalanx over the radial aspect in both the cases, in the antero posterior radiograph. The patients underwent re-exploration and there was no recurrence after 5 years.

RESULTS

FNAC was positive for giant cells in 14 cases out of the 18 cases, and in the rest it was inconclusive or negative. Skeletal changes due to pressure effect of the mass in the form of bony indentation were seen in four cases. Two cases had recurrence with mass over the dorsal aspect of the thumb which were successfully managed with a second surgery. Inadequate resection of the margins could be the cause of recurrence. Superficial wound infection was seen in two cases which responded to a broad spectrum antibiotic cover. Post-operative stiffness were noted in three thumb cases and one in the little finger, and all of them regained full range of movements with mobilization from the physio therapy clinic.



Figure 1: Mass over the thumb in a patient with antecedent trauma

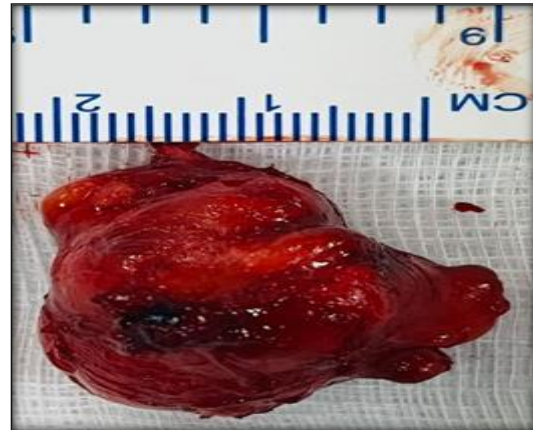


Figure 2 and 3: Type I c lesion: Multi-lobulated lesion surrounded by a common pseudocapsule, from volar aspect of the thumb, in a patient with antecedent trauma



Figure 4: Antero posterior radiograph of the patient in Fig. 1, showing scalloping of the proximal phalanx of the thumb

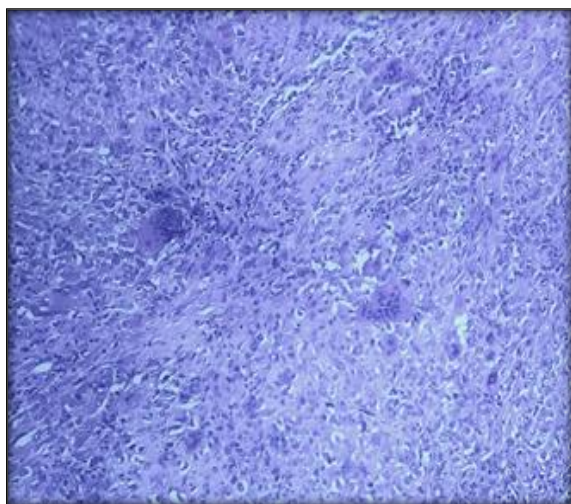


Figure 5: H&E 100x - Microscopic picture of Giant cell tumour of tendon sheath showing multinucleate giant cells in a background of mononuclear cells and collagenous stroma

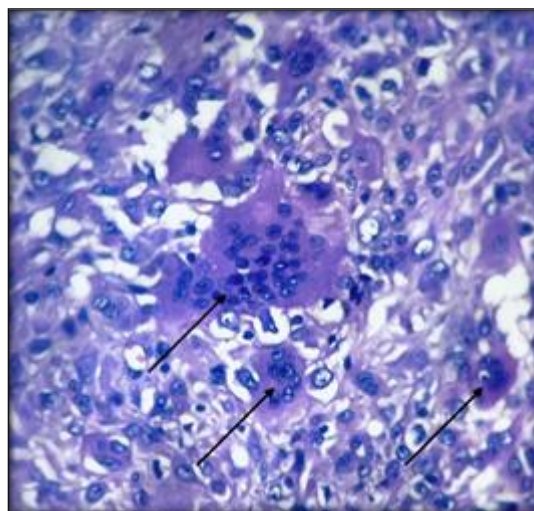


Figure 6: H&E 400x - Microscopic picture of Giant cell tumour of tendon sheath showing multinucleate giant cells (arrowed) in a background of mononuclear cells and collagenous stroma

Table 1: Demographic details of patients

No:	Age	Sex	Site of swelling	FNAC	Al Qattan type	X ray Changes	Recurrence
1	25	F	Volar aspect of Index finger	Positive	I a	Nil	0
2	30	M	Dorso medial aspect of thumb (Fig. 1)	Positive	I c	Bony indentation (Fig. 2)	0
3	15	F	Dorsal aspect of hand	Positive	I a	Bony indentation	0
4	31	F	Thumb (Dorsum)	Negative	I a	Nil	0
5	29	F	Thumb (Dorsum)	Positive	I b	Nil	0
6	39	M	Right thumb (Dorsum)	Positive	I a	Nil	0
7	28	F	Volar aspect of Ring finger	Positive	I b	Nil	0
8	35	F	Wrist in relation to FCU	Inconclusive	II b	Nil	0
9	35	F	Left thumb (Dorsum)	Positive	I a	Nil	0
10	10	F	Thumb (Dorsum)	Positive	I b	Nil	0
11	25	F	Volar aspect of Ring finger	Positive	I c	Nil	0
12	27	F	Dorsum of thumb (Near interphalangeal joint)	Positive	I b	Bony indentation	1
13	32	F	Dorsum of thumb	Positive	I b	Nil	0
14	53	F	Dorsum of hand	Inconclusive	I a	Nil	0
15	30	F	Thumb near interphalangeal joint	Inconclusive	I a	Bony indentation	1
16	32	F	Dorsal aspect of left wrist	Positive	II b	Nil	0
17	15	F	Right thumb (Dorsum)	Positive	I b	Nil	0
18	31	F	Volar aspect of Index finger	Positive	I a	Nil	0

DISCUSSION

As per the literature the rate of recurrence of GCTTS is 9–44%,^[3,5,6] with few authors reporting as high as 45%.^[2,4,8] Various predictive factors have been described for the recurrence. These include location at the inter phalangeal joint, pressure erosions on radiographs and presence of degenerative joint disease. Lowyck and De Smet^[2] in their study did not find any significant correlation between these factors and recurrence. Again the bony indentation due to pressure from the overlying

tumor should not be considered as intra osseous invasion and is not associated with a higher recurrence rate as per Al-Qattan study.^[1] Difficulty in excising the lesion completely due to limited space may be a reason for the higher incidence of recurrence in the distal joint.^[3]

Classification of these tumours has helped in analyzing the recurrence pattern. Byers classified this into localized nodular type (common in hand) and diffuse type (common in joints).^[1,7] Al Qattan on the other hand has analyzed the recurrence after surgical excision, where he has classified it as Type

I- as a single tumor which is round or multi lobulated, and Type II, where there are two or more distinct tumors which are not joined together.^[1]This new classification by Al-Qattan is particularly useful in predicting the recurrence as satellite lesions are often missed if not a magnifying loupe is used especially in Type IIa and Type II b lesions.^[1,10]

In a nodular tumor since there is clear margin it could be easily excised, where as in a diffuse tumor excision may be difficult due to its infiltrating nature. Ikeda et al. recommends microscopic excision even in nodular as there may be small part which is diffuse.^[7] Again he infers that there are more diffuse tumors in the hand than described.^[7] They associate diffuse tumors with location at the distal interphalangeal joint, adjacent degenerative joint disease and presence of pressure erosions of bone.

Recurrence can be prevented if a microscope is used for excision.^[7] Ikeda et al reported recurrence in only one out of 18 when they used the microscope and in that particular case an operating microscope was not used. Using a magnifying loupe to excise the satellite lesions after excision of the nodules was suggested by Ozalp.^[8]The usage of operating microscope or a magnifying loupe has been advocated by many authors for complete excision, as complete surgical excision is the only factor which has been proved to prevent recurrence.^[7,8]

Ultrasonography can detect whether the tumor is solid or cystic and if there are any satellite lesions.^[11] It can also give information regarding the extent of involvement with the underlying tendon and the percentage of circumferential involvement.^[11]From the histological point of view these are composed of multinucleated giant cells, polyhedral histiocytes, fibrosis and hemosiderin deposits.^[8,12,13]Cellularity and mitosis, though previously found significant, were not found to influence recurrence.^[1,10] But as per Rao and Vigorita high incidence of recurrence in tumors were found with increased mitotic activity^[5].All recurrent lesions showed increased mitotic activity even though there was no relation between the number of mitosis found initially and recurrence.^[5] However, Monaghan et al. has shown that mitotic figures do not indicate recurrence and suggested complete local excision as the treatment of choice to prevent recurrence.^[14]

Absence of a gene nm23 is associated with high rate of recurrence as per Grover et al.^[15]Again described nm23-H1 as an independent prognostic factor. However, Lorea et al. in 2004 found no correlation between nm23-H1 expression and recurrence.^[16]

Mainstay of treatment of these tumours is complete surgical excision assisted either with an operating microscope or a magnifying loupe. Radiotherapy has been suggested if complete excision was not possible and in patients with high mitotic activity to prevent recurrence.^[4] Recurrence rate of only 4% were reported with this method of management.

However Ozalp et al. said excision even for multiple recurrences, and none were managed with radiotherapy.^[8]

FNAC is now being used as a primary diagnostic tool and helps in proceeding with care of soft tissue tumours.^[17]According to these authors most of the management decisions can be made from a cytological analysis preoperatively thus preventing a recurrence.^[9,17] Patient should thus be counseled in the preoperatively with regard to the probability of recurrence. Most important factor deciding recurrence is incomplete excision and leaving behind satellite nodules. Thus, adequate surgical exposure, meticulous dissection and use of magnification are all necessary to reduce recurrence and should remain the mainstay of surgical management. Following the above surgical protocol, we were able to achieve almost complete clearance of the tumor with recurrence in only two cases [11%].

CONCLUSION

Recurrence in giant cell tumor of the tendon sheath (GCTTS) does occur and still an unresolved issue. Our technique of preoperative planning aided by tissue diagnosis, wide surgical exposure and complete excision after meticulous dissection with the help of magnification has proved useful and should be considered as a treatment option.

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