



# Giant Cell Tumor of the Tendon Sheath of the Hand: Analysis of Factors Impacting Recurrence

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## Abstract

**Background** Giant cell tumors of the tendon sheath (GCTTS) of the hand are considered the second most common benign tumors of the hand after ganglion cysts. Excision biopsy is considered the standard treatment at present. They are notorious for having a very high rate of recurrence as given in many studies. Many factors are said to be associated with recurrence of the tumors. The goal of this study is to evaluate the long-term results of a series of 48 patients operated on at a single institute and to find out if there is any correlation between the proposed risk factors with recurrence.

**Methods** A retrospective analysis was done in cases of GCTTS operated on between 2015 and 2021. The patients were invited for follow-up for a minimum of 2 years, and the patient files were reviewed. Further data were collected at follow-up including recurrence, range of movement, sensation, skin necrosis, scarring, and digital neuropathy. A data analysis was done. The correlation between the proposed risk factors and recurrence was calculated with the Pearson correlation coefficient. A *p*-value of less than 0.05 was considered statistically significant.

**Results** During the 6 years, 48 patients were operated on. Recurrence was observed in eight patients (16%) at an average of 38.7 months from the time of surgery. Of the risk factors, tumors with satellite nodules and tumor adjacency to joint correlated significantly with recurrence. No complications were observed during follow-up.

**Conclusion** GCTTS of the hand has a high propensity to recur. The presence of satellite nodules and proximity to interphalangeal joints are two important risk factors for recurrence. Magnification during surgery ensures complete excision of the tumor and reduces the chance of recurrence.

## Keywords

- ▶ giant cell tumor
- ▶ hand tumors
- ▶ tendon sheath tumor
- ▶ recurrence

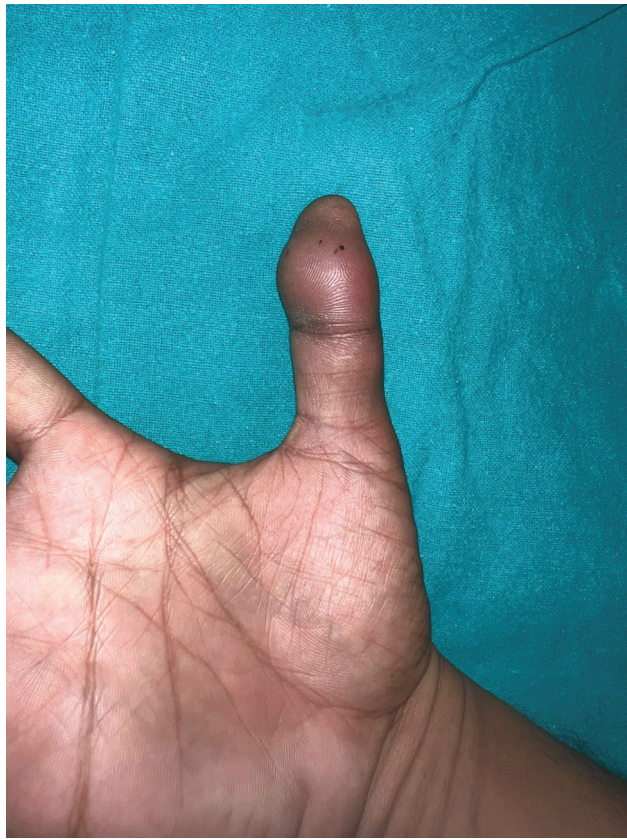
## Introduction

A giant cell tumor of the tendon sheath (GCTTS) of the hand is a benign tumor of unknown origin. It is considered the second most common benign tumor of the hand after

ganglion cysts.<sup>1</sup> Peak incidence is seen in the third and fourth decades, with a female predilection.<sup>2,3</sup> These tumors present as firm painless swellings over flexor aspects of the hand or feet (► **Fig. 1**). On radiographs, they appear as soft-tissue shadow or bony erosions. Magnetic resonance imaging (MRI)

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**Fig. 1** Presentation of a giant cell tumor of the tendon sheath (GCTTS) as painless swelling over the right thumb.



**Fig. 2** Intraoperative image of the same patient showing a well-encapsulated type I tumor encased by a thick capsule arising from the flexor pollicis longus (FPL) tendon sheath, close to the thumb interphalangeal joint.

and ultrasound can support the diagnosis.<sup>4</sup> The pathogenesis of GCTTS is still debated. The most accepted theory is that reactive or regenerative hyperplasia is accompanied by an inflammatory process. Recent studies detected chromosomal translocation in *chromosome 1p13* in most of the tumors.<sup>5</sup> They are classified as nodular (►Fig. 2) and diffuse types (►Fig. 3) according to Al-Qattan's classification.<sup>2</sup> Excision biopsy is considered the standard treatment at present. They are notorious for having a high rate of recurrence as given in many studies. It has been reported to be as high as 15 to 48%.<sup>3,6–8</sup> Many independent risk factors have been listed in the literature (►Table 1). Factors favoring recurrence are the presence of Al-Qattan type II tumors, indentation of tumor on the bone cortex, proximity of the tumor to the joint, presence of degenerative joint disease, tumors with increased mitotic activity, encasement of neurovascular structures, and incomplete excision.<sup>2,6–11</sup> There is plenty of variation in the literature stating which of the above-mentioned conditions are more relevant in causing recurrence. The goal of this study is to evaluate the long-term results of a series of patients operated on at a single institute and to find out the correlation of the proposed risk factors with recurrence.

## Material and Methods

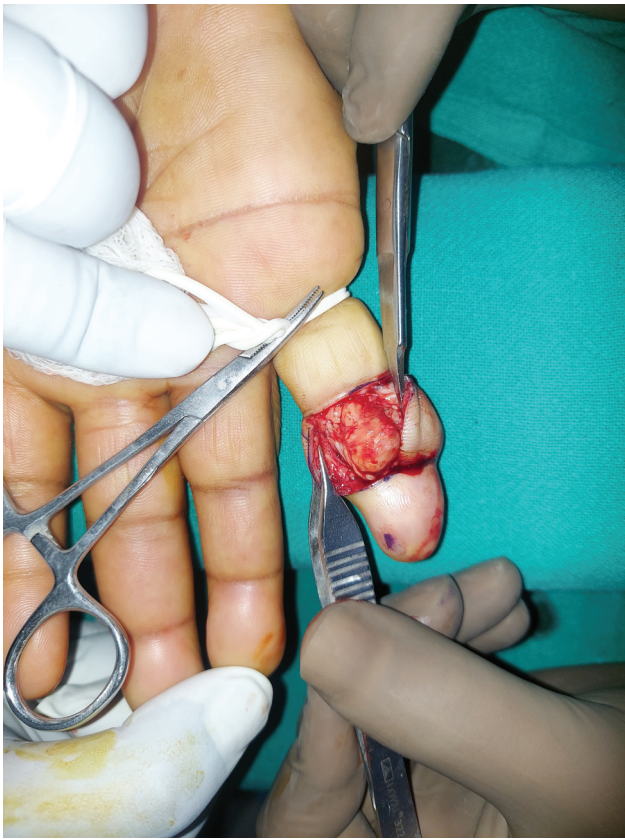
We retrieved the clinical data of 48 patients operated on between 2015 and 2021 with a histologically confirmed

diagnosis of GCT and analyzed them retrospectively. All the documentation was reviewed, and the patients were invited for follow-up for a minimum of 2 years. Patient demographic data, clinical features, imaging findings, operative notes, and histological features were reviewed.

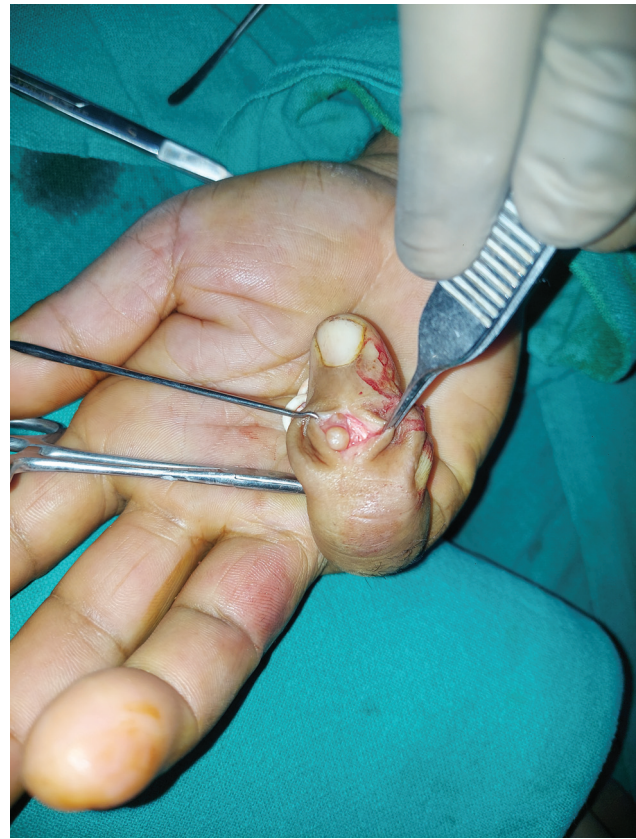
We performed X-ray and ultrasound examinations in all our cases and MRI in selected cases. All the patients underwent surgery under regional anesthesia, with tourniquet control and 5x loupe magnification. The incision was placed according to the location of the tumor on dorsal, lateral, or volar aspect of the hand. The tumors that were extending from volar to dorsal and vice versa. The tumors with multiple satellite nodules required more than one incision (►Figs. 3 and 4). In toto tumor excision was done whenever possible, preserving neurovascular structures. Encasement around the nerve or vessel is not usually circumferential, and we often do find a plane to dissect around them. If that is not possible, we open the tumor to dissect out the structures. We ensure removal of all satellite nodules and parts of tumor before wound closure. Postoperatively, we start active range of motions and physical therapy after 1 week and continue until full range of motion is reached.

Further data were collected at follow-up including recurrence, range of motion, sensation, skin necrosis, scarring, and digital neuropathy.





**Fig. 3** Tumor over the volar aspect of the middle phalanx of the left little finger.



**Fig. 4** Presence of satellite nodules on the dorsal aspect in the same patient (Al-Qattan type II).

**Table 1** Proposed risk factors for GCTTS recurrence

Location at the DIP joint of the finger or thumb IP joint
Incomplete excision of the tumor
Pressure indentation over bone
Increased cellularity or mitotic activity on histologic examination
Degenerative joint disease near the tumor
Tumors that are nm23 negative
Al-Qattan type II tumors

Abbreviations: DIPJ, distal interphalangeal joint; GCTTS, giant cell tumor of the tendon sheath; IP, interphalangeal.

## Statistical Analysis

Data entry and statistical analysis were done with Microsoft Excel 2019. Continuous variables were presented as mean  $\pm$  standard deviation. The chi-squared test is used to compare categorical variables. The correlation between proposed risk factors and recurrence was calculated with the Pearson correlation coefficient. Statistical significance was considered at  $p < 0.05$ .

## Results

During the 6 years, 48 patients with histological evidence of GCTTS were operated on. All the patients could be traced and

followed up. The average age at operation was 48.35 years (range: 36–55 years). Contrary to other series, most of our patients were males (75%). In 38 cases, the tumors were found in the palmar aspect of the finger or hand. The anatomical distribution of tumors on the hand is given in ►Table 2. Most of the patients presented with a painless swelling that developed pain as the size of the swelling increased. The range of motion of the hand and fingers was affected in 16 patients. The tumor was noted by 40 patients at an average of 16.4 months before surgery (range 6–39 months). The remaining eight patients could not say when exactly they first noticed the tumor. Only 16 patients could recollect trauma to the hand. However, this could be incidental.

Bone indentation or periosteal reaction is seen in nine patients, of whom two had recurrence. In 13 (27.08%) patients, satellite nodules were found. In six patients, there was an extension into the joint. X-ray and ultrasound findings were sufficient in most patients. MRI was done in cases of suspected neurovascular involvement and recurrence (►Figs. 5 and 6). All cases were operated on under 5x loupe magnification. Pseudocapsule (Al-Qattan type I or nodular type; ►Fig. 2) was found in 35 tumors (72.9%) and 13 tumors were type II (without pseudocapsule and presence of satellite nodules; ►Figs. 3 and 4).

Lesions averaged  $28 \pm 12$  mm in size. The largest tumor measuring 6.5 cm (►Fig. 2). Tumors were adherent to the tendon substance in 28 cases. In 32 cases, the tumor was

**Table 2** Anatomical distribution of tumors

Digit	Metacarpal	MCPJ	Proximal phalanx	PIPJ	Middle phalanx	DIPJ/thumb IPJ	Distal phalanx	Total
Thumb	–	3	3	–	–	2	0	8
Index finger	–	2	3	1	4	3	2	15
Middle finger	–	1	2	0	3	2	1	9
Ring finger	–	2	1	1	2	3	1	10
Little finger	–	0	2	1	1	0	0	4
Palm of the hand	2	–	–	–	–	–	–	2
Total	2	8	11	3	10	10	4	48

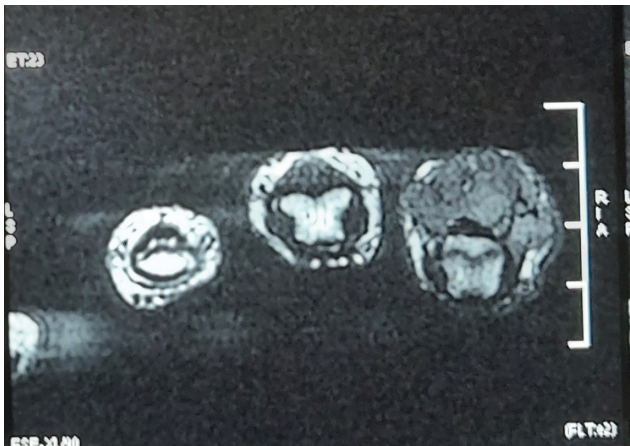
Abbreviations: DIPJ, distal interphalangeal joint; IPJ, interphalangeal joint; MCPJ, metacarpophalangeal joint; PIPJ, proximal interphalangeal joint.



**Fig. 5** Appearance of giant cell tumor of the tendon sheath (GCTTS) of the same patient in Fig. 1 on T1-weighted magnetic resonance imaging (MRI).

encasing a digital nerve or artery. Joint stiffness was noted in 10 cases during early follow-up, which improved with physical therapy.

The average follow-up time was 44 ± 18 months (range: 24–72 months). Recurrence was observed in eight cases (16%). The average time of recurrence is 38.7 months from the time of surgery. The earliest observed recurrence was at 13 months. In all cases of recurrence, satellite nodules were noted during the first surgery. Reoperation was done in six patients and two patients refused surgery. They are being followed up.



**Fig. 6** Encasement of neurovascular structures is seen on axial section on magnetic resonance imaging (MRI) in a different patient.

Pearson's correlation coefficient was calculated for four factors with recurrence, and the *p*-value was derived after the *t*-test. The results of the test are outlined in ► **Table 3**. We observed a significant correlation of recurrence with proximity to distal interphalangeal joint (DIPJ) and type II tumors. Encasement of neurovascular structures and pressure erosion of bone, however, did not show statistical significance in recurrence (► **Table 3**). All the patients had well-healed scars, a normal range of motion, and no sensory deficit at their last follow-up.

**Discussion**

There is a large volume of literature on GCTTS, yet there are challenges faced by hand surgeons. The tumor can penetrate the joints and bones or completely encase the neurovascular structures. So complete removal of tumor tissue and preservation of vital tissues is a challenge. In their systematic review, Williams et al reported a recurrence rate of 7 to 44%.<sup>11</sup> Some studies showed a recurrence rate as high as 45%.<sup>8,9</sup> Our overall rate of recurrence was 16% at an average of 38.7 months. In our study, we tried to investigate the correlation between the rate of recurrence and the known risk factors.<sup>2</sup>



**Table 3** Proposed risk factors versus number of recurrences

Risk factor	Recurrence	No recurrence	Correlation coefficient	p value
Location of tumor at the DIPJ or thumb IPJ	6	4	0.59	0.0007
Pressure erosion of bone	2	7	0.07	0.63
Encasement of the neurovascular structures	4	28	0.15	0.28
Type II tumors	8	5	0.73	0.0003

Abbreviations: DIPJ, distal interphalangeal joint; IPJ, interphalangeal joint.

GCTTS appear as soft-tissue shadows on X-ray. The presence of bony erosion is not uncommon. The importance of ultrasound in the diagnosis of GCTTS was described by Middleton et al.<sup>4</sup> GCTTS have a characteristic sonographic appearance and occur in predictable locations. Characteristic description is a solid-appearing hypoechoic mass with detectable blood flow, adjacent to the flexor tendons of the fingers.<sup>4</sup> MRI was done selectively for proximal palmar tumors and in tumors encasing the digital nerves and vessels.

Reilly et al reported that recurrence was higher with tumors localized at the DIPJ and thumb interphalangeal (IP) joints and with dorsally localized tumors.<sup>3</sup> Intraosseous invasion and bone erosions were shown to be a risk factors for recurrence.<sup>2,3</sup> However, some studies show that bone erosion is due to the pressure effect of the tumor and poses no risk of recurrence.<sup>7</sup> Fotiadis et al, in their systematic review, reported that a specific finger or phalanx is not associated with an increased risk of recurrence.<sup>12</sup> Al-Qattan type II lesions were associated with a higher recurrence rate.<sup>13</sup> Neither cellularity nor mitoses could be considered significant prognostic factors for recurrence.<sup>2,12</sup> In our study, significant recurrence is seen in tumors close to the DIPJ and Al-Qattan type II tumors with multiple satellite nodules.

Darwish et al reported that most tumors recurred in the thumb.<sup>10</sup> However, Williams et al, in their series, reported 3 of 34 recurrences localized to the thumb. They showed that the recurrence rate reached 32% when the flexor, extensor tendons, and the joint capsule were involved.<sup>11</sup> In a review by Fotiadis et al, an average recurrence rate of 15.1% was noted with the usage of loupes and 14.4% without magnification, which was not a significant difference. They also stated that recurrence was not associated with a specific finger or phalanx, yet there was a slightly higher incidence in tumors close to the DIPJ.<sup>12</sup> Although the use of radiotherapy for the prevention of recurrence was promoted by some authors, due to small numbers, it is not significant.<sup>9</sup> A gene called nm23 has been identified to inhibit infiltration in normal cells. GCTs that are nm23 negative are more aggressive and associated with a greater recurrence rate.<sup>14</sup> We did not perform nm23 testing in our cases due to cost factors and it will not be altering the management plan.

The presence of degenerative arthritis increases the chances of accumulation of histiocytes in the joints. It increases the difficulty in surgical excision and hence the risk of recurrence.<sup>6</sup> None of our patients had arthritis in the joints close to the tumor.

At times, the tumors grow by displacing or encasing the neurovascular structures. This may also result in incomplete tumor excision.<sup>6</sup> As they are encapsulated tumors, often there will be a plane between the tumor and the neurovascular structures, and the tumor can be dissected in toto. When that is not possible, we open the tumor and carefully dissect around the nerve or vessel end ensure complete tumor excision. We had not faced any inadvertent intra-operative injury to digital or palmar nerves and vessels.

We attribute the low rates of recurrence in our series to our experienced hand surgeons and the use of 5x loupe magnification in all cases. Magnification aided in complete tumor excision from the joints, periosteum, and neurovascular structures. A meta-analysis by Siegel et al states that recurrence can occur any time between 1 and 244 months, but it is mostly seen within the first 5 years.<sup>15</sup> In that aspect, the mean follow-up period being only 3.66 years in our series might also have contributed to the low recurrence rate. On the contrary, a follow-up of 3 years or more is considered sufficient to rule out future recurrences.<sup>7</sup>

There are a few more limitations to our study. Although we acclaim magnification and experience of surgeons as a reason for complete excision, we do not have a comparison group that got operated on in lesser magnification or by a less experienced surgeon to establish a relation. The recurrence rate is small, and confident statistical conclusions could not be drawn in a few aspects. None of our patients had degenerative joint disease near the tumor. So its relation to recurrence could not be evaluated, although it is a known risk factor. Other risk factors like increased cellularity, mitotic activity, and nm23 status of the specimen could not be evaluated for recurrence. We did not give postoperative radiotherapy as the benefit is not conclusive in the literature.<sup>9</sup>

An appreciation for these relative risk factors should enable surgeons to plan appropriately and educate their patients regarding the risk of recurrence.

## Conclusion

The GCTTS of the hand, although benign, has a high propensity to recur. Although many risk factors for recurrence have been described in the literature, in our study, we found strong correlations with two factors, which are Al-Qattan type II tumors with multiple satellite nodules and proximity of the tumor to the IP joints. Magnification and meticulous

dissection ensuring complete excision resulted in minimal recurrence in our series.

#### Author Contributions

P.V.K. contributed to the study design, collection of data, data analysis, and writing of the manuscript. S.S.A. contributed to the study design, data analysis, operative procedure, and patient care. I.A. contributed to the study design, collection of data, data analysis, operative procedure, and patient care. I.K.S. and O.P. contributed to writing of the manuscript. Y.R.K. contributed to data collection.

#### Conflict of Interest

None declared.

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