



Giant Cell Tumor on Tendon Sheath of the Hand: The Incidence of Recurrence, Functional Outcomes and a Literature Review

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Abstract

Giant Cell Tumor on Tendon Sheath (GCTTS) of the hand is a benign nodular tumor originated from tendon sheath of the tendons and the joints and is the second most common soft tissue tumor of the hand after ganglion cyst. The aim of this study is to evaluate 79 cases of giant cell tumor of tendon sheath of the hand and to analyze the postoperative functional outcomes and recurrence rate of the tumor in a mean period of five years follow up. **Material and Methods:** Fifty five subjects were females and twenty four males with an average age of 38,8 years-old (range 23-65 years old) were included in this study. The most frequent location of tumor was the index finger (32/79 patients, 40,5%). The majority of patients was presented in outpatient's clinic for a painless swelling on volar side of the digit, while only 13(16,45%) patients presented for painful mass and numbness of the digit in 7(8,9%) cases. All cases were categorized by Al-Qattan classification system with type I lesion in 51(64,5 %) cases and type II in 28(35,5 %) cases. Dimension of tumors macroscopically varied from 0,4cm to 5 cm. In 12 cases we reconstructed the A2 pulley to avoid bowstring deformity and eventual postoperative loss of hand function. In 12 cases we reconstructed the A2 pulley to avoid bowstring deformity and eventual postoperative loss of hand function. 8 patients with bone erosion underwent postoperatively complementary local radiotherapy. A standard protocol of rehabilitation was performed in all patients. **Results:** Results were evaluated according to complications, range of motion, Quick Dash Score and functional satisfaction of patients in a mean follow-up of 49 months. Two major complications presented in this study: 5 cases with transitional neuroapraxia and 6 cases with recurrence of the tumor with a mean of postsurgical period 14,8 months. Finally at last assessment the mean Quick Dash Score was 4,5 (range from 0-11,4) the range of motion was fully recovered in 73 patients while patient's satisfaction rated from "very satisfied" in 58(73,4%) cases, "satisfied" in 17(21,5) and "dissatisfied" in 4(5,1%) patients. **Conclusions:** Giant cell tumor of tendon sheath of the hand is a common tumor which presents a high incidence of recurrence in different populations of patients. The goal of treatment is to reduce the recurrence rate and restore a functional hand. The operation procedure should be well planned preoperatively, the tumor must excised en block, dissection must be under surgical loop and must be emphasized to remove satellite lesion while when patients present increased risk factors or inadequate excision of the mass, postoperative radiotherapy is performed.

Keyword: Giant cell tumors, tendon sheath, hand tumor, recurrence

Introduction

Giant Cell Tumor on Tendon Sheath (GCTTS) in the hand is a benign nodular tumor which originates from the tendon sheath of the tendons and the joints ^[1]. It is the most common soft tissue tumor of the hand after ganglion cyst ^[2]. GCTTS belongs to a rare group of proliferative disorder of the synovial joints and tendon sheaths and for these reason it has been referred to the literature by a diversity of titles such as benign syn-ovioma, fibrous xanthoma, localized villonodular synovitis, sclerosing hemangioma, fibrous histiocytoma of synovium, and pigmented villonodular

tenosynovitis ^[3]. GCTTS is a painless tumor with a slow-growing period for clinical manifestation which varies from months to years ^[4]. Generally it appears in patients between 30 and 50 years of age and affects more commonly women than men ^[5,6].

Initial classification of the tumor described from Byers et al in two major types, the localized nodular type (common in hand) and the diffuse type (common in other joints) ^[25]. The first type is a well delineated lobulated swelling in majority of cases, extrarticular and benign which appears most common in volar surface on sheaths flexor tendon of the hand ^[2]. The diffuse type is an infiltrative mass in plurality of cases, intra-articular with

multiple satellite lesion with preference for large joints and may produce limited range of motion [6,7]. Al-Qattan (2001) has analyzed the postoperative recurrence of the tumor and proposed a new classification system for GCTTS consisted of two main types depending on whether the entire tumor was, or not surrounded by one pseudocapsule, as assessed during surgical procedure. Type I is a single tumor which is round or multi lobulated and type II is appeared as two or more distinct tumors which are not joined together. Each type was sub-classified according to the thickness of the capsule, lobulation of the tumor, the presence of satellite lesions, and the diffuse or multicentric nature of the tumor [12].

Although pathogenesis of the tumor is not clear, many etiological factors have been described in literature such as infection, vascular disorders, osteoclastic proliferation, immune mechanisms, inflammation, neoplasia and metabolic disorders, sequel of bleeding after trauma [4,8]. The most common accepting theory is that GCTTS originates from a reactive or regenerate hyperplasia associated with an inflammatory process [9,13].

Primary step in diagnosis of GCTTS is clinical examination and patient medical history. Standard radiography is useful when tumor produces erosion in cortical area of the phalange or invades medullary canal [10]. Ultrasonography or magnetic resonance imaging(MRI) are important tools both in diagnosis of the lesion and in management of the preoperative planning [11].

The aim of this study is to evaluate 79 cases of giant cell tumor of tendon sheath of the hand and to analyze the postoperative functional outcomes and recurrence rate of the tumor in a mean period of five years follow up.

Material & Methods

The study was performed at the Orthopaedic department of General Hospital of Heraklion-“Venizeleio «for 7 years from 2012 to 2019. The Institutional Ethical Committee approved the study. Every soft tissue tumor of the hand was inserted into a database using a conventionalize protocol. The protocol format included demographics elements, localization of the tumor, preoperative imaging examinations, surgical details (Al-Qattan classification system), and following-up data. At the final examination the complication rates were assessed (postoperative recurrence, infection, and digital neuropathy), range of motion, functional outcomes and satisfaction of patients.

Table I. Anatomical distribution of the GCTTS

	MCP Joint	Proximal Phalanx	PIP Joint	Middle Phalanx	DIP Joint	Distal Phalanx	Thumb IP Joint	Thumb Proximal Phalanx
Volar	6	25	5	13	11	4	1	1
Dorsal			1	4	1		1	
Lateral		3	2	1				
Total	6	28	8	18	12	4	2	1

All patients were performed radiographic examination which revealed in 8 cases bony erosions. Sixty five patients underwent magnetic resonance imaging with the results showing a low signal on T1WIs and T2WIs, while ultrasonography in 14 cases showed a hypoechogenic image.

The operative excision was performed with 4,5x magnification loupes under brachial tourniquet and regional anesthesia. Surgical approach of the mass depended on the location so volar digital Brunner approach performed when the tumor was volarly, midlateral in lateral location of tumor and dorsal incision in dorsal localization. Extreme attention was given to excise the tumor en block without destroying the capsule, with scope to avoid

Seventy nine patients were included in this study. Fifty five were females and twenty four males with an average age of 38,8 years-old (range23-65yrs). The mean duration of symptoms before excision ranged from eight months to ten years. The most frequent location of tumor was in index finger(32/79 patients, 40,5%),while the other distribution of the tumor was in thumb(3/79 patients, 3,8%), middle (19/79 patients, 24%) ring(14/79 patients, 17,8%), and little finger(11/79 patients, 13,9%) (Fig.1). The majority of patients was presented in outpatient’s department for a painless swelling on volar side of the digits, while only 13(16,45%) patients presented for painful mass and numbness of the digit in 7(8,9%) cases. The dominant hand was affected in 48 cases (60,75 %). Among causes, 17(21,5 %) cases referred a past trauma, 5(6,3%) cases had rheumatic arthritis and the rest 57(72,2%) cases had unknown etiology.



Figure 1: Anatomical distribution of GCTTS in 79 cases: Thumb (3), Index (32), Middle (19), Ring (14), Little finger (11) (grey arrow).

Determining the location of the tumor, depending on the anatomical landmark of the hand was in metacarpophalangeal joint(7,6%), proximal phalanx(35,44%), proximal interphalangeal joint(10,12%), middle phalanx(22,8%), distal interphalangeal joint(15,19%), distal phalanx(5%), proximal phalanx of the thump(1,35%) and interphalangeal joint of the thumb(2,53), while the localization was in volar surface(83,6%), dorsal(8,9%) and lateral(7,6%) (Table I).

damage in digital neurovascular element and tendons and to discover and remove all satellite lesions (Fig. 2). Dimension of tumors macroscopically varied from 0,4cm to 5 cm). In 12 cases we reconstructed the A2 pulley to avoid bowstring deformity and eventually postoperative loss of hand function. All cases were categorized by Al-Qattan classification system in which type I lesion was in 51(64,5 %) cases and type II in 28(35,5 %) cases (Table II). The majority of tumors detected type I(51/79 - 64,5 %) more than type II(28/79 -35,5 %). Microscopically tumors contained giant cells, histiocytes and haemosiderin. Patents with bone erosion underwent complementary local radiotherapy.

Table II: Al Qattan classification in 79 cases

Type	Subtype
I. The entire volume is surrounded by a pseudo-membrane 51(64,5 %) pts	a. Solitary nodule in a thick whitish membrane. 15/79 pts b. Solitary nodule in a thin film. 27/79 pts c. Lobar lesion surrounded by a common pseudo membrane 9/79 pts
II. The entire volume is not surrounded by a membrane 28(35,5 %) pts	a. A main node (in a pseudo-membrane), which is accompanied by a separate satellite lesion. 17/79 pts b. Widespread type with many granular tumors without membrane. 9/79 pts c. Polycentric type with many separate lesions in the same finger 2/79 pts

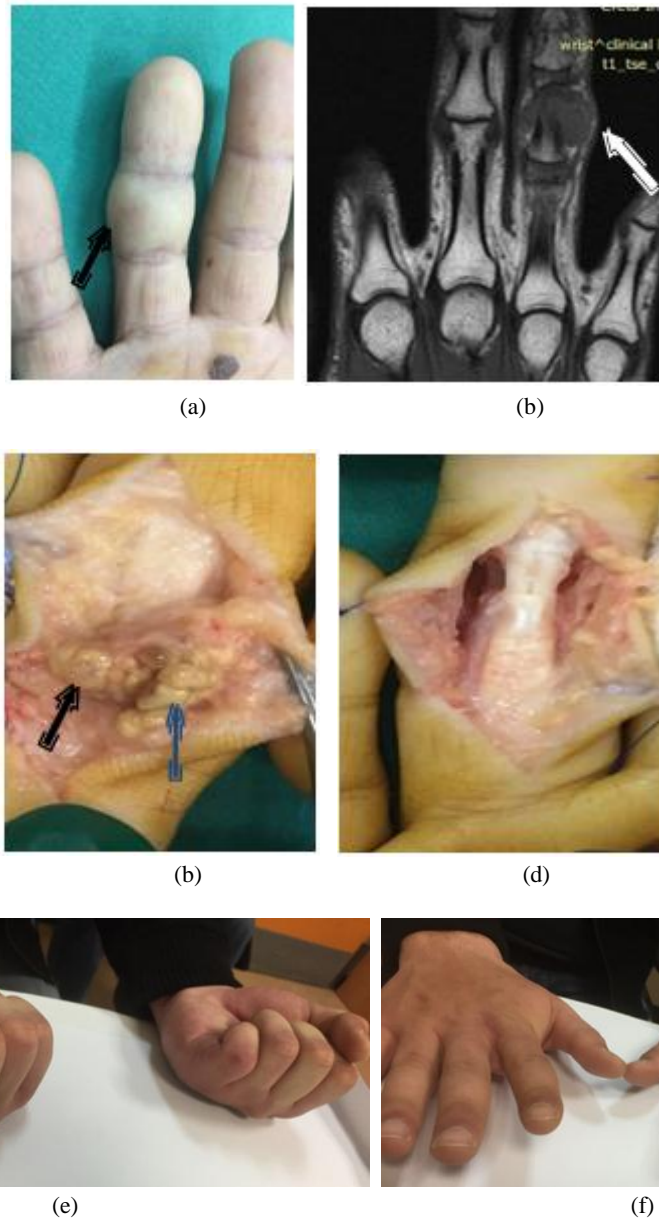


Figure 2: Man 34 years old(a) with a swelling on volar middle phalanx of right ring finger for 3 years (black arrow). MR I(b) appears GCTTS (white arrow). Tumor over flexors tendons (black arrow) with lateral extension (blue arrow) which classified as type IIb (Al-Qattan)(c). Full excision of the tumor (d). At final follow-up (50 months) full range of motion without recurrence (e,f)

Postoperatively sutures were removed at 10-12 days and a standard protocol of rehabilitation was performed in all patients. Physiotherapy treatments were delivered twice a week. The main goal in the first sessions was the decrease of post-operative edema and the prevention of scar tissue formation. Patient was trained to perform scar massage on his own, passive and self assistive finger flexion and extension for 2 weeks and then when pain and edema decreased active range of motion was performed in pain limit. Patients received 20 sessions of physiotherapy treatment in order to eliminate scar formation and enhance the range of movement and

then were assessed every month for six months follow-up to secure that there was no dysfunction.

Results

The Mean follow-up is 49 months (range from 14 months to 6 years). None of patients missed the last re-examination. Results were evaluated according to complications, range of motion, Quick Dash Score and functional satisfaction of patients. We classified complications in two categories, complications among the surgery (infection, skin necrosis, neurovascular damage) and postoperative

complications (recurrence of tumor, stiffness). We observed 5 cases in the first category with transitional neuroapraxia. In three cases the mass was localized in lateral size and in two cases the tumor was localized volarly and has lateral extension. All cases recovered after a mean period of 4 months (range 8 weeks to 6 months). Three cases developed infection which treated with antibiotics for 2 weeks.

Recurrences were present in 6 patients, at 5,9,12,16,20,27 months after the surgery. Five patients underwent a second procedure which accompanied with local radiotherapy. None of these patients had initial bone erosion and were classified as type I in 1 cases and type II in 5 cases. Among localization one was at proximal phalanx, two at proximal interphalangeal joint, one at middle phalanx and 2 at distal interphalangeal joint. Four of these

cases had lateral extension of the tumor and three of them were in contact with digital neurovascular bundle. None of these patients developed new recurrence of the tumor.

At final examination the mean Quick Dash Score was 4,5 (range from 0-11,4). The range of motion was fully recovered in 73 patients while in 6 patients presented a mean restriction at the flexion 6°, 8°, 10° of distal interphalangeal compared with the contralateral hand. The other two cases presented a restriction of 5°, 7° of proximal interphalangeal joint. Three of them suffered from rheumatic arthritis and two cases had second operation because of recurrence of the tumor. Finally, the patient's satisfaction recorded as "very satisfied" in 58(73,4%) cases, "satisfied" in 17(21,5) and "dissatisfied" in 4(5,1%) patients (Fig 3).

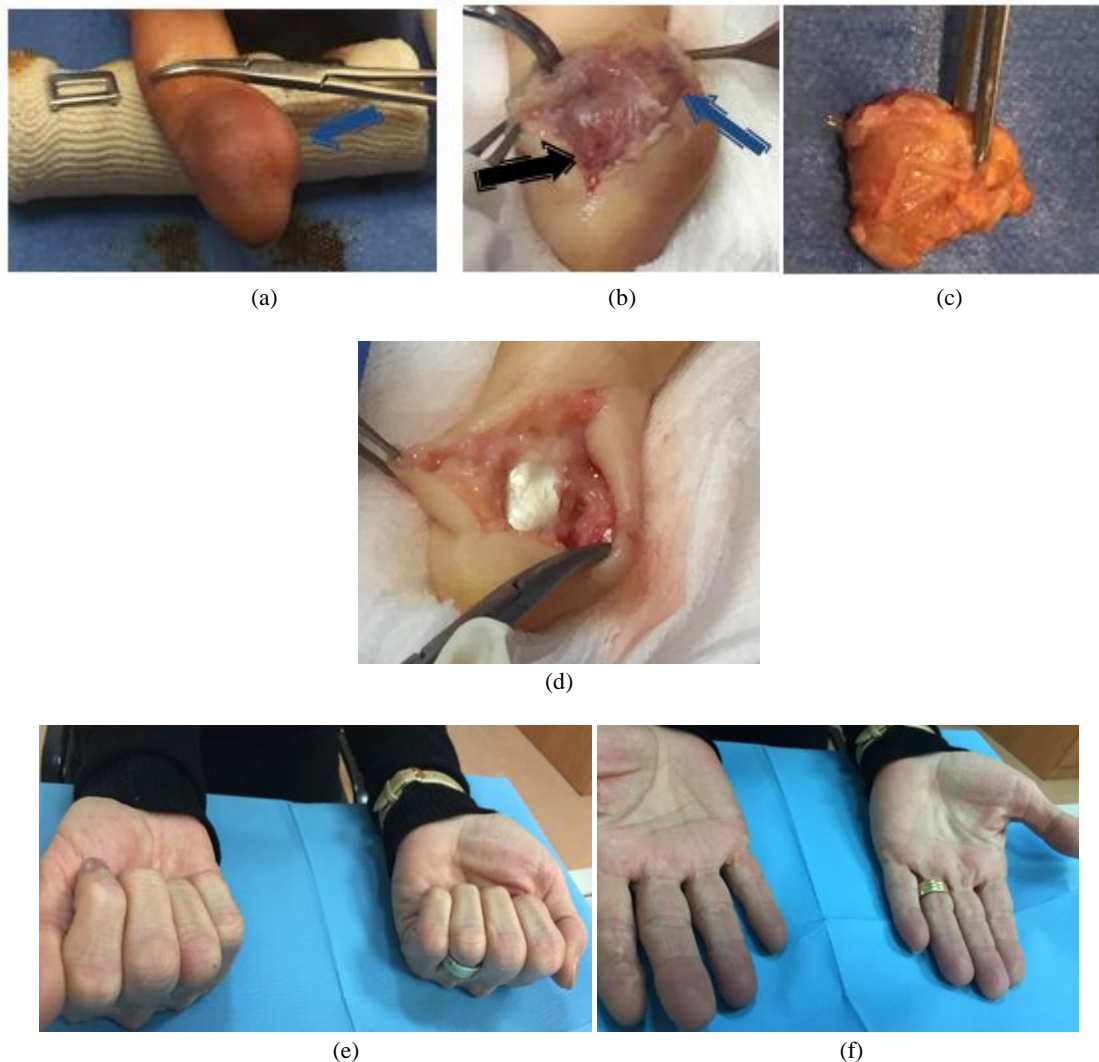


Figure 3: Female 58 years old with a painful swelling (blue arrow) on volar distal phalanx of right index finger for ten years(a). Tumor localized (b) on flexor profundus tendon (black arrow) of index finger over attaching to the distal phalanx with lateral extension(blue arrow). GCTTS type Ia (Al- Qattan)(c). Full excision of the tumor (d). At final follow-up (52 months) full range of motion without recurrence (e,f)

Discussion

From the first description in the literature by Chassaignac (1852) as "malignant tumor of tendon sheaths" to its final name from Targett (1897) as "giant cell tumor of tendon sheath" tumor has been described by many titles with the result that there is not clear definition of the disease [11,14,15]. It is the second most common type of soft tissue tumor of the hand after ganglion cyst, and appears in general as solitary benign painless, slow-growing tumor. In rare cases tumor can produce severe restriction of motion [18]. In

literature, this lesion has also been reported in wrist, shoulder, hip, knee and foot [17]. GCTTS originates from the tenosynosheath, bursa and joint synovium [16]. Histopathologically the tumor presents fusiform cells, fibrotic tissue elements, histiocytes with cholesterol, multinuclear giant cell and haemosiderin [19]. Initial histological characteristics of cellularity and mitosis were considered important elements to postoperative recurrence of tumor finally reported that is not affected by these factors [12]. Rao et al (1984) found in their own study high incidence of recurrence in tumors with increased mitotic activity and suggested that there

was no relation between the initial number of mitosis and recurrence but all recurrence lesion showed increased mitotic activity [18]. Instead Monaghan et al in 51 patients found that mitotic and apoptotic features are common in GSTTS but they do not indicate the recurrence of the tumor [2]. Recent advances in molecular oncology by Groves et al showed that giant cell tumors nm23 negative are more aggressive and have increased recurrence rate [20]. Lorea et al (2004) in their study reported that there is no correlation between nm23 H1 expression and risk for recurrence [28].

Giant cell tumor of tendon sheaths of the hand can occur in any age predominant between the third and fifth decade of life with a great incidence of 2 per 100,000 per year between 40-50 years especially in female [8]. In majority of cases it is diagnosed in a single joint while the incidence of poliarticular comprise is less than 1% [21]. Preoperative imaging examination must be performed with target to decreased differential diagnosis and to discover other lesions (bone erosion, subchondral cyst) [13]. Ultrasonography usually distinguish a hypoechogenic mass but has also been described a hyperechogenic lesion [22]. Additional MRI shows a low signal intensity on both T1-T2 weighted images of the lesion but principal role of the specific examination is early diagnosis of GSTTS, accurate size and extend of the lesion and to differentiate between villonodular synovitis, synovial chondromatosis and synovial sarcoma [23]. Lautenbach et al(2013) in 84 cases of GCTTS suggest that none of the soft tissue imaging methods(U/S - MRI) prevail in diagnosis or for the preoperative planning [24]. On the contrary Wang et al(2017) investigate the value of the MRI in both types of GCTTS in 38 patients and suggest that the specific imaging examination is an optimal modality for preoperative assessment of tumor size, extent and invasion of adjacent joint and Tenosynovial space [26]. We postulate that these examination offer element about the lesion (size, extension, bony erosion) but the final diagnosis offers histopathological confirmation.

Although wide marginal surgical excision of the tumor is the gold standard of treatment for both types of GCTTS the important risk of this lesion is recurrence and joint damage and particular(especially) caution must need in management of diffuse type of tumor because of increased rate of recurrence [27]. Local recurrence of the tumor has been reported in literature varied from 4,7% to 45% [5,12,13,27,29,30]. Fotiadis et al(2011) proposed the risk factors of GCTTS recurrence, location at the distal interphalangeal joint of the finger or the thumb, incomplete excision, bone erosion, cellularity and mitotic activity on histopathological examination, proximity to the arthritic joint, tumors which are nm 23 negative and type II tumors [5]. Reilly and Grover suggest that bone erosion might a factor for recurrence [13,20]. Kitagawa and Al- Qattan do not support this theory and defend that bony erosion caused from the pressure effect of the tumor and is not invasion [12,32]. In 8 patients in our study with bony erosion none appeared recurrence, but all of them underwent radiotherapy.

Anatomical location like interphalangeal joints of the finger and thumb localization is a factor for recurrence [29]. Reilly et al reported when tumors localized in DIP joint and thumb IP joint, had an incidence of recurrence 79% and those with dorsal localization 34%, while Williams et al reported 3 cases in 34 patients with GSTTS localized into the thumb [13,33]. Ozben et al presented in their study recurrences at the PIP and DIP joint but none after excision of 11 tumors from the thumb and in dorsally localized tumors [30]. In our recurrence rate only 4 cases were localized in interphalangeal joints (2 in PIP and 2 in DIP joint) but none of the cases with localization in thumb presented recurrence.

Kitagawa et al (2004) investigated the involvement of tumor with digital neurovascular band associated with increased rate and supported that vicinity of tumor with neurovascular elements makes the complete excision of the tumor difficult and maybe a factor of recurrence [32]. Di Grazie et al reported 3 recurrences in 7 cases with neurovascular involvement, while Ozben et al referred in their study neurovascular intricacy did not increase recurrence risk [29,30]. Three patients from our recurrence had involvement with digital neurovascular bundle.

Al-Qattan suggests that type II tumors had increased recurrence rates [12]. This theory is supported by other investigators as well and in our study 5 patients were classified as type II [4,5,29]. On the contrary Ozben supported that type II GCTTS don't show an increased recurrence rate [30].

Ikeda and Kitagawa suggested parameters to decreased recurrences such as the use of a tourniquet, use of magnification devices like loupe or operating microscope, and meticulous surgical dissection and thorough exploration and clearance of all the satellite lesions [25,32]. If the tendon was involved with tumor, must be sacrificed and need reconstruction [25]. Debridement and absolute curettage are needed in lesions involving the adjacent bone and bone cement or graft must be used, while in cases with involvement of the skin an elliptical excision of the involved area followed by primary closure or skin grafting when the defect is larger is recommended [32,35].

Postoperative Radiotherapy has been suggested after inadequate excision and in patients with increased risk factors of GCTTS recurrence [34]. Kotwal et al. reported no recurrence in 14 patients with postoperative radiotherapy and suggested the use of 20 Gy in daily doses of 2 Gy, On the contrary other studies in the literature reported 2.3-75% recurrences with postoperative radiotherapy [5,27,36,37]. In literature options regarding postoperative radiotherapy are general equivocal [5,29,30,36]. In our series all the cases with bone erosion and five patients from the recurrence ?? who underwent radiotherapy the tumors were not relapsed.

Conclusions

Giant cell tumor of tendon sheath of the hand is a common tumor. Magnetic resonance imaging is a diagnostic tool defining the location, extension, growth characteristic of the lesion and managing the preoperative planning. The tumor presents a high incidence of recurrence in different populations of patients. For this reason the risks factors of recurrences must be decreased with excised a block the tumor, dissection must be under surgical loop to discover and remove satellite lesion, protect the neurovascular elements, reconstruct the pulley system and when patients present increased risk factors or inadequate excision of the mass to perform postoperative radiotherapy. Patients should be informed about the high percentage of recurrence of the tumor even in complete excision and that should be considered in postoperative reexaminations.

Ethical Approval

Our institution does not require ethical approval for reporting individual cases or case series.

Consent Form

Verbal informed consent was obtained from the patient for their anonymized information to be published in this article.

Conflict of interest

The authors declare that have no conflict of interest.

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