JBUON 2018; 23(5): 1546-1551
ISSN: 1107-0625, online ISSN: 2241-6293 • www.jbuon.com
E-mail: editorial_office@jbuon.com

ORIGINAL ARTICLE

Giant cell tumors of the tendon sheath of the hand: an 11-year retrospective study

Christos Koutserimpas[1], Grigorios Kastanis[2,3,4], Argyrios Ioannidis[5], Dimitrios Filippou[6], Konstantinos Balalis[4]

[1]Department of Orthopedics and Traumatology, "251" Hellenic Air Force, General Hospital of Athens, Athens, Greece; [2]Department of Orthopaedics and Traumatology, General Hospital of Sitia, Sitia, Crete, Greece; [3]Department of Orthopaedics and Traumatology, "Venizeleio" General Hospital of Heraklion, Heraklion, Crete, Greece; [4]Department of Orthopaedics and Traumatology, University Hospital of Heraklion, Heraklion, Crete, Greece; [5]Second Department of General Surgery, "Sismanoglion" General Hospital, Athens, Greece; [6]Department of Anatomy and Surgical Anatomy, Medical School, National and Kapodistrian University of Athens, Athens, Greece

Summary

Purpose: *Giant cell tumor of the tendon sheath (GCTTS) is a slowly progressing soft tissue tumor. The present retrospective study recorded and evaluated cases of GCTTS of the hand.*

Methods: *A cohort of patients suffering from GCTTS of the hand and treated surgically were studied in terms of diagnosis, therapy, recurrence, as well as in terms of functional outcome with the use of the QuickDASH score.*

Results: *A total of 36 patients (13 men; 23 women) with a mean age of 38.8 years (±standard deviation/SD=8.7) were evaluated. According to Al- Qattan classification 10 cases of type Ia, 11 cases of type Ib, 6 cases of Ic and 9 cases of IIa were found, while the mean tumor diameter was 2.6 cm (SD=1.1). The mean follow up was 21 months (SD=12). The mean QuickDASH Score was 6.3 (SD=6.7). Furthermore, a total of 31 patients (86%) characterized their outcome as satisfactory. Recurrence was observed in 4 patients, while none of them had initially undergone radiotherapy.*

Conclusions: *This study has shown a direct correlation between the QuickDASH Score results and the objective level of satisfaction in cases of GCTTS treated surgically. The present study cohort had 11.11% recurrence rate during a mean follow-up of 21 months. It is of note that none of these cases had initially undergone radiotherapy. It is of utmost importance to carefully select the patients that meet the criteria for postoperative radiotherapy.*

Key words: *giant cell tumor, hand surgery, tendon sheath*

Introduction

GCTTS is a slowly increasing soft tissue tumor with a growth period that varies from months to years [1]. GCTTS, also called pigmented villonodular tenosynovitis, is a lesion composed of pleomorphic cell population, including lipid-laden foamy cells, multinucleated giant cells and round or polygonal stromal cells, often with deposits of hemosiderin in a collagenous stroma [1,2].

The nature of this lesion is controversial, since some authors believe it has a neoplastic nature, whereas others consider it a non-neoplastic tumor. There are a few theories regarding the pathogenesis of GCTTS, such as trauma, infection, vascular disorders, lipid metabolism disorders, osteoclastic proliferation, immune mechanisms, inflammation, neoplasia and metabolic disorders. The dominant

Correspondence to: Argyrios Ioannidis, MD. 2nd Department of General Surgery, "Sismanoglion" General Hospital, 1, Sismanogliou street, 15126, Amarousion, Athens, Greece.
Tel: +30 210 6859159, E-mail: agis.ioannidis@gmail.com
Received: 14/02/2018; Accepted: 09/03/2018

pathogenic hypothesis is however, a reactive or regenerative hyperplasia associated with an inflammatory process [3,4].

GCTTS has two types: diffuse and localized. The localized type is more commonly observed in the hand and rarely in the joints. The diffuse type is larger than the localized type, may be localized within the joint, and may lead to a limited range of motion [5].

Solitary tenosynovial nodules in the digits of the hand have been reported in 7-45% of patients. It is the second most common tumor of the hand after ganglions, while it appears at the sheath of the flexor tendon in 85% of the cases. It affects patients between 30 and 50 years old, with a higher frequency in women [6].

Therapeutically these lesions related to tendon sheaths should be carefully surgically removed, in order to prevent recurrence (Brunner approach) [7].

According to Al Qattan, GCTTS can be intraoperatively classified into two main types, depending on whether the entire tumor was, or was not, surrounded by one pseudocapsule. Each type is then sub-classified according to the thickness of the capsule, lobulation of the tumor, the presence of satellite lesions, and the diffuse or multicenteric nature of the tumor (Table 1) [8].

The Quick Disabilities of the Arm Shoulder and Hand (DASH) is a shortened version of the DASH Outcome Measure. Instead of 30 items, the QuickDASH uses 11 items to measure physical function and symptoms in people with any or multiple musculoskeletal disorders of the upper limb. The QuickDASH was designed to include disability questionnaires. As such, scaling was ranked from 0 indicating least disability to 100 indicating most disability [9].

The present retrospective study, with a mean follow up of 21 months, recorded and evaluated 36 cases of GCTTS of the hand in terms of diagnosis, therapy, recurrence, as well as in terms of functional outcome with the use of the QuickDASH score.

Methods

Between January 2005 and December 2015, 36 consecutive patients suffering from GCTTS of the hand, treated surgically from a single experienced hand surgeon with radical resection of the tumor (with palmar he zig-zag volar-digital incision Brunner approach, cleaning of bone erosions, and restoration of the tendon sheath) were studied.

Physiotherapy included initially passive and later active mobilization with the limit of pain from the second postoperative day for all patients. Gender, age, personal medical history, clinical appearance, location of the tumor and the cause of the mass were evaluated and recorded. Additionally, recurrence, as well as complications were recorded during the follow-up.

All lesions were classified according to Al-Qattan classification. The QuickDASH score was performed postoperatively, evaluating the function during the follow-up. At their follow-up, patients completed a visual analogue scale of 0 to 100, where 0 indicated "not satisfied" and 100 was "very satisfied". Those defining their level of satisfaction as ≥50 were categorized as being satisfied and those <50 were categorized as dissatisfied. This method has already been used for estimating patient's satisfaction in hip and knee arthroplasty [10,11].

Statistics

Mean values and standard deviations (SDs) were calculated using Microsoft Excel 2007 (Microsoft corporation, Redmond, Washington). A p value <0.05 was considered as statistically significant.

Results

A total of 36 patients (13 men; 23 women) with a mean age of 38.8 years (SD=8.7) were surgically treated for a GCTTS of the hand from 2005-2015. The tumor was palmar-located in all cases, while in 24 cases at the predominant side (right side for right handed and left side for left handed). Seven patients were complaining of numbness and 12 had a mechanical block. The duration of symptoms ranged from 2 months to 4 years.

Table 1. Al Qattan classification of tendon sheath giant cell tumors of the hand

Type I: The entire volume is surrounded by a pseudo - membrane
a. Solitary nodule in a thick whitish membrane. b. Solitary nodule in a thin film. c. Lobar lesion surrounded by a common pseudo membrane.
Type II: The entire volume is not surrounded by a membrane
a. A main node (in a pseudo-membrane), which is accompanied by a separate satellite lesion. b. Widespread type with many granular tumors without membrane. c. Polycentric type with many separate lesions in the same finger.

The identified cause was trauma in 12 cases (33.34%), rheumatic arthritis in 2 (5.55%), while the rest were of unknown cause (22; 61.11%). The mass was located at digit (D) 2 in 14 cases or 38.8%, at D3 and D4 in 7 cases or 19.4% each and at D5 in 8 or 22.4%.

Plain radiographs showed soft tissue swelling in all cases and bone erosion in 8 patients. These patients received radiotherapy after surgery.

According to Al-Qattan classification the tumors were categorized by the same experienced surgeon intraoperatively as 10 cases of type Ia, 11 cases of type Ib, 6 cases of Ic and 9 cases of IIa. Macroscopically the mean tumor size was 2.6 cm (SD=1.1), while histologically all lesions fitted the classical description of pigmented villonodular tenosynovitis, with polygonal cells with round nuclei and faintly eosinophilic cytoplasm interspersed among giant cells. Granules of hemosiderin were also present in the cytoplasm of the stromal cells (Figures 1 and 2).

The mean patient follow-up was 21 months (SD=12). Surgical results were evaluated according to the QuickDASH Score (for function) at follow-up. The mean QuickDASH Score was 6.3 (SD=6.7). Furthermore, a total of 31 patients (86%) characterized their outcome as satisfactory.

Postoperative complications were observed in 4 (11.11%) patients: 1 case of neuroapraxia, 1 case of infection and 2 cases of stiffness. Recurrence was recorded in 4 patients, distally from the initial tumor, at a 5, 12, 16 and 27-month follow-up period, while 3 of them underwent a second procedure and radiotherapy. None of the patients with recurrence had, initially, undergone radiotherapy (Table 2).

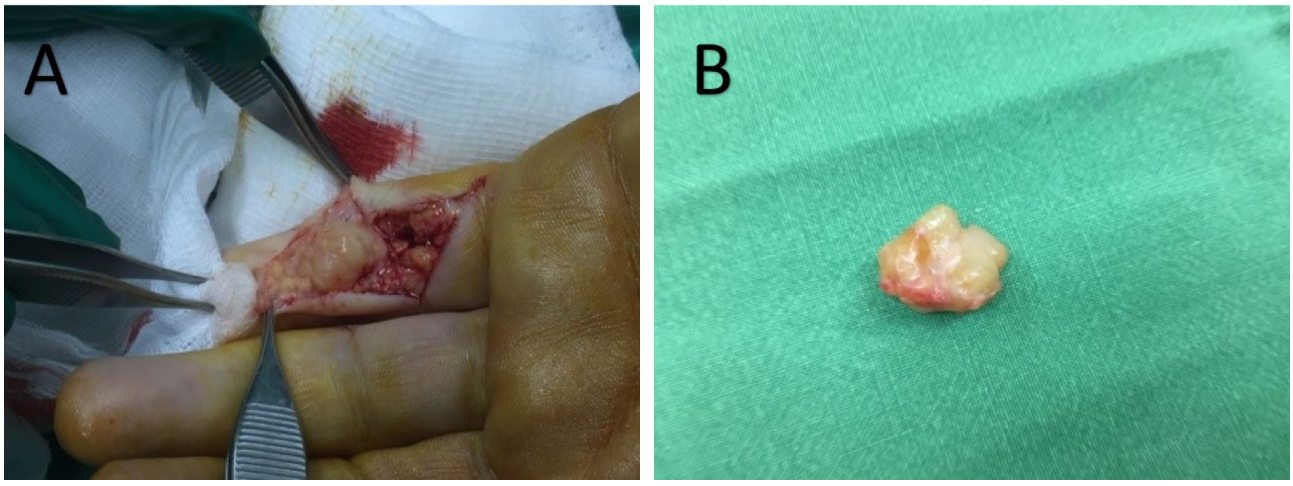

Figure 1. A: Male 34 years old (case 2), with block of small digit. Surgical specimen after resection. **B:** Lobar lesion surrounded by a common pseudo membrane, categorized as Ib according to Al-Qattan classification.

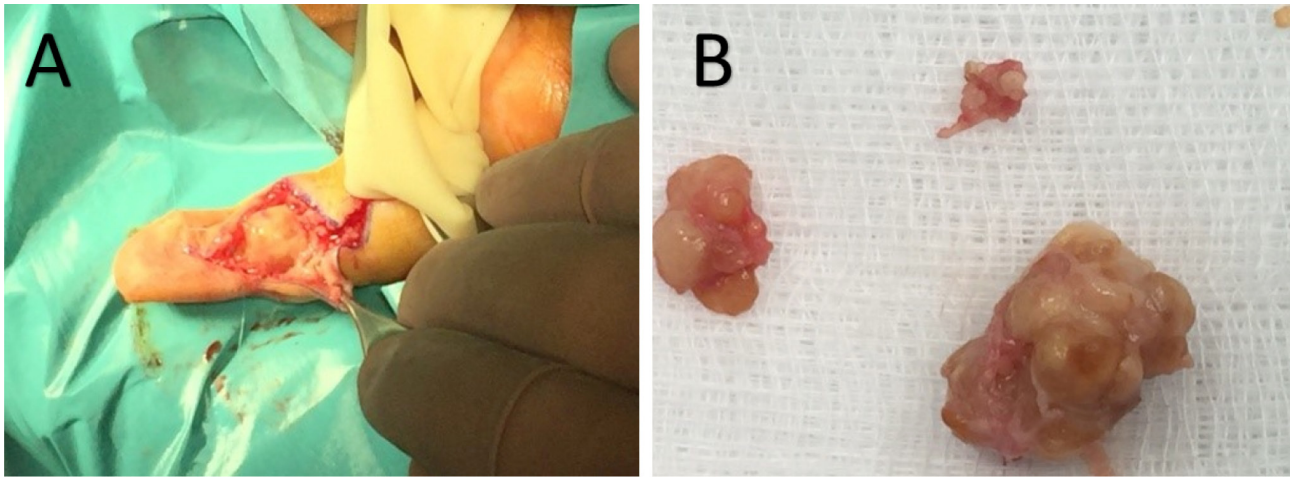

Figure 2. A: Female 39 years old (case 31), with block of small digit. **B:** Surgical specimen after resection. Lobar lesion surrounded by a common pseudo membrane, categorized as IIc according to Al-Qattan classification.

Table 2. Demographics and clinical findings of the study population

n	*Gender*	*Age, years*	*Site*	*Al-Qattan classification*	*Tumor diameter (cm)*	*Quick DASH Score*	*Follow-up (months)*
1	F	38	Index finger	Ic	3.6	22.7	36
2	M	34	Little finger	IIa	2.5	18.2	48
3	M	45	Middle finger	Ib	2.3	11.4	18
4	F	38	Index finger	Ia	5	0	10
5	F	56	Little finger	Ic	2.2	15.9	24
6	M	36	Index finger	IIa	4.2	2.3	26
7	F	13	Ring finger	Ib	2.3	4.6	14
8	M	27	Middle finger	Ia	3.2	0	2
9	F	31	Ring finger	Ib	2.1	2.3	17
10	M	33	Index finger	Ic	2.4	0	30
11	F	44	Ring finger	Ib	1.7	6.8	8
12	M	40	Index finger	Ib	2.9	4.6	58
13	F	52	Little finger	Ia	1.4	22.7	10
14	F	30	Index finger	IIa	2.7	2.3	16
15	M	32	Ring finger	Ic	1.8	6.8	12
16	F	34	Middle finger	Ib	3	4.6	36
17	F	44	Ring finger	IIa	0.4	2.3	32
18	F	39	Middle finger	Ib	2.6	11.4	12
19	F	46	Ring finger	Ia	1.6	0	23
20	M	35	Index finger	Ia	3.7	2.3	8
21	F	38	Middle finger	IIa	3.4	0	24
22	M	45	Index finger	Ia	4.2	4.6	24
23	F	23	Little finger	Ib	3.4	2.3	18
24	M	34	Index finger	Ic	1.1	18.2	26
25	F	42	Little finger	IIa	1.4	2.3	14
26	F	46	Little finger	Ib	0.3	0	36
27	M	45	Ring finger	Ia	2.4	0	24
28	F	50	Index finger	Ia	3.6	4.6	30
29	M	46	Middle finger	IIa	4.2	13.6	18
30	F	48	Index finger	Ia	2.8	2.3	18
31	F	39	Ring finger	IIa	3.2	0	7
32	M	34	Index finger	Ib	2.7	4.6	19
33	F	46	Middle finger	Ia	1.4	6.9	10
34	F	28	Index finger	Ib	2.8	13.6	23
35	F	47	Little finger	Ic	2.2	2.3	24
36	F	40	Index finger	IIa	2.6	11.4	6

M: males, F: females

Discussion

GCTTS was first described in 1952 as fibrous xanthoma. Since then and based on pathological findings, it was referred to as fibrous histiocytoma of synovium, pigmented nodular tenosynovitis, tenosynovial giant cell tumor, localized nodular tenosynovitis, benign synovioma, and fibrous xanthoma of synovium [12].

The GCTTS or pigmented villonodular tenosynovitis is a benign tumor, the second most common at the hand after ganglionic cysts. The differential diagnosis of the tumor include neurofibroma, pyogenic granuloma, desmidoma, and malignant fibrous histiocytoma. The diagnosis of GCTTS is based mainly on clinical examination [13].

The present study population consisted mainly of females (approximately 2/3 of the sample) with a mean age of 38.8 years. The GCTTS of the hand presents as a slow-growing clinical tumor and is generally observed in patients aged 30–50, with a female-to-male ratio of 3:2 [3,14]. The female to male ratio (2:1) and the age of the patients

in the present study are in concordance with those reported in the literature [1,3,6].

Regarding the functional follow-up of the present patients, the mean DASH Score was 6.3. This can be interpreted as satisfactory, since it is lower than 10.1, which is the score of the general population, as reported in a large survey by Hunsaker et al. [15].

In addition, the majority of patients (31; 86%) objectively defined their outcome as satisfactory at the mean 21 months follow-up. Only 5 (14%) of them were categorized as "not satisfied", since they defined their level of satisfaction as less than 50. It is of note that those had the higher scores in QuickDASH Score (22.7, 22.7, 18.2, 18.2 and 15.9). Due to the small number of patients, we cannot come to statistically significant conclusions. However, the study has shown a direct correlation between the QuickDASH Score results and the objective level of satisfaction in patients treated surgically for GCTTS of the hand.

Regarding recurrence, the present study had an 11.11% rate during the mean 21 months follow-up. It is of note that none of these cases had, initially, undergone radiotherapy. In the literature recurrence rates are reported between 10 and 45% of the cases [16].

The radical careful resection of the tumor is a very important factor in minimizing recurrence, which, although always mentioned, is not enough highlighted. Carefully removing the tendon sheath and cleaning bone erosions, as well as using the microscope for the surgical resection have been shown to reduce recurrence [16]. Postoperative radiation therapy has also been proven to decrease the recurrence rate [16].

It is of utmost importance to decide which patients are good candidates for postoperative radiotherapy. There are many authors that recommend postoperative radiation in high risk cases [17-19]. These include a tumor located at the distal interphalangeal joint of the finger or the thumb, poor surgical technique (incomplete excision), bone erosion (from X-rays), high mitotic activity (histologically), proximity to the arthritic joint, and type II and nm 23 negative tumors [20-24].

Conflict of interests

The authors declare no conflict of interests.

References

1. Darwish FM., Haddad WH. Giant cell tumor of tendon sheath: experience with 52 cases. Singapore Med J 2008;49:879-82.
2. Jaffe HL, Lichtenstien HL, Elsutro CJ. Pigmented villonodular synovitis, bursitis and tenosynovitis. Arch Pathol 1941;31:731-65.
3. Hamdi MF, Touati B, Zakhama A . Giant cell tumour of the flexor tendon sheath of the hand. Musculosketal Surg 2012;96:29-33.
4. Cavaliere A, Sidoni A, Bucciarelli E. Giant cell tumor of tendon sheath: immunohistochemical study of 20 cases. Tumori 1997;83:841-6.
5. Suresh SS, Zaki H. Giant cell tumour of tendon sheath: case series and review of literature. J Hand Microsurg 2010:67-71.
6. Garg B, Kotwal PP. Giant cell tumor of the tendon sheath of the hand. J Orthop Surg (Hong Kong) 2011;19:218-20.
7. Ozalp T, Yercan H, Kurt C et al. Giant cell tumor of tendon sheath of the hand. Acta Orthop Traumatol Turc 2004;38:120-4.
8. Al-Qattan M. Giant cell tumours of tendon sheath: classification and recurrence rate. J Hand Surg 2001;26:72-5.
9. Aasheim T, Finsen V. The DASH and the QuickDASH instruments. Normative values in the general population in Norway. J Hand Surg Eur 2014,39:140-4.
10. Baker PN, van der Meulen JH, Lewsey J, Gregg PJ. The role of pain and function in determining patient satisfaction after total knee replacement. Data from the National Joint Registry for England and Wales. J Bone Joint Surg Br 2007;89:893-900.
11. Judge A, Arden NK, Kiran A et al. Interpretation of patient-reported outcomes for hip and knee replacement surgery: identification of thresholds associated with satisfaction with surgery. J Bone Joint Surg Br 2012;94:412-8.
12. Adams EL, Yoder EM, Kasdan ML. Giant cell tumour of the tendon sheath: experience with 65 cases. Eplasty 2012;12:423-30.
13. Monagham H., Salter DM., Al-Nafussi. Giant cell tumors of the tendon sheath (localized nodular tenosynovitis): clinicopathological features of 71 cases. J Clin Pathol 2001;54:404-7.
14. Messoudi A, Fnini S, Labsaili N, Ghrib S, Rafai M, Largab A. Giant cell tumours of the tendon sheath of the hand: 32 cases. Chir Main 2007;26:165-9.
15. Hunsaker FG, Cioffi DA, Amadio PC, Wright JG, Caughlin B. The American Academy of Orthopaedic Surgeons Outcomes Instruments – Normative Values from the General Population. J Bone Joint Surg 2002;84-A(2):208-15.
16. Choughri H., Moussaoui A., Nonnenmacher J. Giant cell tumor of the tendon sheath with intraosse-

ous phalangeal invasion: a case report and review of the literature. Eur J Orthop Surg Traumatol 2005;15: 151-6.

17. O' Sullivan B, Cummings B, Catton C, Bell R, Davis A. Outcome following radiation treatment for high-risk pigmented villonodular synovitis. Int J Radiat Oncol Biol Phys 2005;63:S50.
18. Coroneos CJ, O'Sullivan B, Ferguson PC, Chung PW, Anastakis DJ. Radiation therapy for infiltrative giant cell tumor of the tendon sheath. J Hand Surg Am 2012;37:775-82.
19. Kotwal PP, Gupta V, Malhotra R. Giant-cell tumor of the tendon sheath. Is radiotherapy indicated to prevent recurrence after surgery? J Bone Joint Surg Br 2000;82:571-3.
20. Grover R, Grobbelaar AO, Richman PI et al. Measurement of invasive potential provides an accurate prognostic marker for giant cell tumor of tendon sheath. J Hand Surg Br 1998;23:728-31.
21. Kitagawa Y, Ito H, Yokoyama M et al. The effect of cellular proliferative activity on recurrence and local tumor extent of localized giant cell tumor of tendon sheath. J Hand Surg Br 2004;29:604-7.
22. Williams J, Hodari A, Janevski P, Siddiqui A. Recurrence of giant cell tumors in the hand: a prospective study. J Hand Surg Am 2010;35:451-6.
23. Ikeda K, Osamura N, Tomita K. Giant cell tumor in the tendon sheath of the hand: importance of the type of lesion. Scand J Plast Reconstr Surg Hand Surg 2007;41:138-42.
24. Rao AS, Vigorita VJ. Pigmented villonodular synovitis (giant-cell tumor of the tendon sheath and synovial membrane). A review of eighty-one cases. J Bone Joint Surg Am 1984;66:76-4.