

## Review Article

# Giant Cell Tumors of Sheaths and Tendons: About 48 Cases with Review of The Literature

I. Boujguenna<sup>1\*</sup>, S. Berrada<sup>1</sup>, A. Fakhri<sup>1</sup>, Y. Chaouqui<sup>2</sup>, Y. Najeb<sup>2</sup>, CH. Bouyaali<sup>3</sup>, N. Cherif Idrissi El Ganouni<sup>3</sup>, H. Rais<sup>1</sup>

<sup>1</sup>Department of Pathological Anatomy, FMPM-UCAM-CHU Mohamed VI, Marrakech Morocco

<sup>2</sup>Department of Traumatology and Orthopedics, FMPM-UCAM-CHU Mohamed VI, Marrakech Morocco

<sup>3</sup>Department of Radiology, FMPM-UCAM-CHU Mohamed VI, Marrakech Morocco

\*Corresponding author: I. Boujguenna, Department of Pathological Anatomy, FMPM-UCAM-CHU Mohamed VI 40000 Marrakech Morocco. Email: imane.boujguenna1992@gmail.com

**Citation:** Boujguenna I, Berrada S, Fakhri A, Chaouqui Y, Najeb Y, et al. (2019) Giant Cell Tumors of Sheaths and Tendons: About 48 Cases with Review of The Literature. Ann med clin Oncol 2: 113. DOI: 10.29011/AMCO-113.000113

**Received Date:** 11 January, 2018; **Accepted Date:** 18 January, 2018; **Published Date:** 24 January, 2019

### Abstract

The Synovial Duct Giant Cell Tumor (TCGS) of tendons is a rare tumor defined according to WHO 2013 as a benign fibro-histiocytic tumor. It develops at the expense of the synovial joint, the serous bursa and the tendinous sheath. The most common location is the tendon sheath of the finger flexors (85%). It mainly affects the young adult with a female predominance. It manifests clinically in two localized and diffuse forms. Its etiology is not yet elucidated. His diagnosis is pathological. Treatment is exclusively surgical with a tendency to recurrence. The purpose of this work is to specify the epidemiological, pathological and evolutionary profile of TCGS with a review of the literature. This is a retrospective study involving 48 cases. of TCGS collected at the pathology anatomy department of Mohammed VI University Hospital of Marrakech over a period of 13 years (January 2004 - December 2018). Our series included 35 women and 13 men with an average age of 50 years (15 to 60 years). 34 patients consulted for a nodule located at the level of the hand, 7 at the foot, 5 at the knee and 2 at the elbow. The duration of evolution of these nodules varied between 8 and 36 months. The clinical examination revealed a nodule of firm consistency, often well limited. Surgical excision was performed in all cases. The pathological study confirmed the diagnosis in all patients. The evolution was marked by the appearance of recurrence in 3 patients. Despite the benign histology of TCGS, its local recurrence capacity (4 to 30%) makes it a tumor with intermediate malignancy. Recurrence depends on the nodular or diffuse nature of the tumor, the histological characteristics and the completeness or non-completeness of the resection, hence the need for postoperative monitoring.

**Keywords:** Giant Cell Tumors; Pathological Anatomy; Sheaths and Tendons

### Introduction

The Synovial Duct Giant Cell Tumor (TCGS) of tendons is a rare tumor defined according to WHO 2013 as a benign fibro-histiocytic tumor. It develops at the expense of the synovial joint, the serous bursa and the tendinous sheath. The most common location is the tendon sheath of the finger flexors (85%). It mainly affects the young adult with a female predominance. It manifests clinically in two localized and diffuse forms (1, 2, 3). Its etiology is not yet elucidated. His diagnosis is pathological. Treatment is exclusively surgical with a tendency to recur (16,20). The aim of

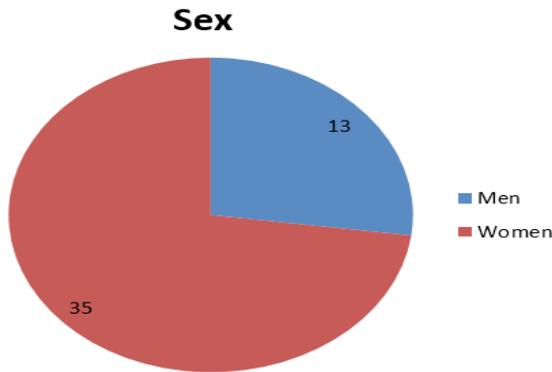
this work is to specify the epidemiological, anatomopathological and evolutionary profile of TCGS with a review of the literature.

### Material and Methods

This is a retrospective study on 44 cases of TCGS collected at the pathology anatomy unit of the Mohammed VI University Hospital of Marrakech over a period of 14 years (January 2004 - December 2018).

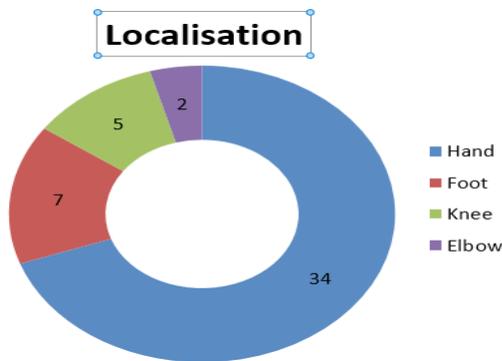
### Results

Our series included 35 women (73%) and 13 men (27%) (Figure 1).



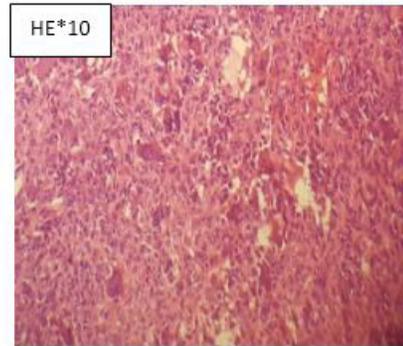
**Figure 1:** Distribution by gender.

The average age of 50 years (15 to 60 years). Thirty-four patients (71%) consulted for a localized nodule in the hand, seven at the foot (14%), five at the knee (11%) and two at the elbow (4%) (Figure 2).

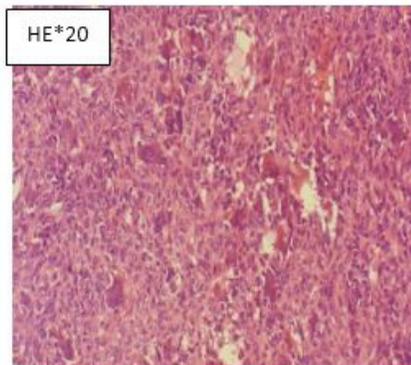


**Figure 2:** Distribution by location.

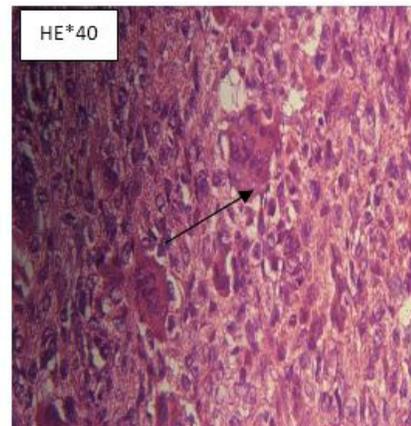
The duration of evolution of these nodules varied between 8 and 36 months. The clinical examination revealed a nodule of firm consistency, often well limited. Surgical excision was performed in all cases. The anatomopathological study confirmed the diagnosis by showing in all patients a poorly limited proliferation made up of two main components: stromal cells and giant cells. The giant cells are large with sometimes more than 20 or 30 nuclei, most often centrally located in an abundant, homogeneous, granular, vacuolated cytoplasm. The mononuclear stromal component is of neoplastic origin. ovoid, medium-sized stromal cells with eosinophilic scant cytoplasm. The nuclei have a fine chromatin with and poorly limited cytoplasm, not atypical with presence of haemorrhagic foci. (Figures 3-5). The evolution was marked by the appearance of recurrence in 3 patients.



**Figure 3:** Proliferation made up of two main components: stromal cells and giant cells (HE\*10).



**Figure 4:** Proliferation made up of two main components: stromal cells and giant cells (HE\*20).



**Figure 5:** Giant cells are large with sometimes more than 20 or 30 nuclei, most often centrally located in an abundant, homogeneous, granular, vacuolated cytoplasm.

## Discussion

The giant cell tumor of the synovial sheath of the hands is a tumor of the young adult. It is a pathology of the 4th and 5th decade [1,2]. Almost all studies have noted a female predominance [2-4] We have found this notion in our series. The diagnosis of the tumor is retained only after a delay often a long time [3,4]. Like most soft-tissue tumors, the etiology of TCGGS in the hand remains unrecognized. It is a digital tumefaction, generally unique, painless in the majority of cases, often palmar, firm, well limited, of variable size, with evolutionary power slow and mobile compared to the superficial plane. The diagnosis is based on imaging and anatomopathological study which confirms the diagnosis by showing a proliferation made up of two main components: stromal cells and giant cells. The giant cells are large with sometimes more than 20 or 30 nuclei, most often of central location in an abundant cytoplasm, homogeneous, granular, vacuole, these cells are osteoclastic, rich in acid phosphatase with a positivity for lysozymes, alpha -antitrypsin, alpha-antichymotrypsin and other histiocytic markers. These giant cells are a priori non-neoplastic but probably correspond to an endoduplication of circulating monocytes which were recruited into the lesion by a mechanism auto or paracrine due to TGF beta. They are of homogeneous and diffuse distribution (except remodeled zones), + heterogeneous and smaller if aneurysmal cyst or chondroblastoma.

The mononuclear stromal component is of neoplastic origin, it is the only proliferative element able to show atypies on the periphery of haemorrhagic or necrotic reworking territories. Its cells are of mesenchymal origin but show a close histogenetic relationship with osteoblasts because focal focal osteoid or bone deposits are found in 1/3 of the cases. These medium-sized, ovoid, eosinophilic low cytoplasmic stromal cells produce type 1 and 3 collagens. The nuclei have fine chromatin, poorly limited cytoplasm, little intercellular collagen, and variable number of mitoses (2 to 20/10 chps). Without pejorative meaning), not atypical [5,6]. This stromal component can be fusiform with storiform fibro-histiocytic tumor type arrangement. Cytological atypia can be seen, especially around haemorrhagic foci.

Abundant vascularization with numerous capillaries or blood lakes (appearance of aneurysmal cyst), changes in the form of cysts, necrosis, foam cells (xanthelasmization) or even fibrosis, possibility of osteogenesis within the tumor (especially in vertebral forms), possibility of periosteal osteogenesis in the form of osteoid foci lined with large osteoblasts, sometimes hypocellular cartilage or coarse calcifications. The immunohistochemical study may show a positivity to focal PS100, actin +, H caldesmone -, receptor positivity estrogen in half of the cases [7-10]. The differential diagnosis arises with numerous benign lesions that may present giant cells in the bone such as fibrous metaphyseal defect, non-ossifying fibroma (immature skeletal subjects, metaphyseal seat,

irregular distribution of multinucleate giant cells, less numerous, compressed in a fibroblastic fundus), chondromyxoid fibroma, chondroblastoma (cellular component of chondroblasts with crenellated nuclear contours and incisures, variable number of giant cells of osteoclastic type, randomly distributed to oval and vesicular nuclei which do not resemble those chondroblasts), Langerhans cell granulomatosis, solitary bone cyst, brown tumor of hyperparathyroidism, solid variant of aneurysmal cyst, osteoid osteoma, osteoblastoma, giant cell repair granuloma, and osteosarcoma rich in giant cells (metaphysis long bones of the adolescent, clear cytological atypia, mitosis ++ and abnormal). Essentially, the diagnosis is that giant cells are uniformly distributed and uniform in giant-cell tumors, whereas in all other lesions there are foci containing many giant cells packed together with extensive areas that are totally lacking, the giant cells being morphologically identical in all of these pathologies. The diagnosis of giant cell tumor must be questioned in the case of a child, a diaphyseal or metaphyseal localization lesion, a multiple or localized lesion in vertebrae other than sacrum, jaw or the small bones of the hands and feet.

In the presence of aneurysmal cyst criteria in an epiphyseal lesion, the specimens are multiplied in search of typical giant cell foci. The solid variant of the aneurysmal cyst resembles the giant cell tumor, but is seen in younger patients (skeletally immature) and has a metaphyseal site [11-14]. The giant cell tenosynovial tumor remains a lesion of unknown nature with a relatively high local recurrence rate of up to 45% in some series [15]. Several recidivism factors have been reported in the literature. Rao and Vigorita [15] found that increased cellularity and high mitotic activity were common in recurrent lesions. However, the authors find no clear relationship between the number of mitoses and the rate of recidivism. On the other hand, Kitagawa et al. [16] and Al-Qattan [17] reported that neither hypercellularity nor the high mitotic index could be considered as prognostic histological factors for recurrence. Recent studies in molecular oncology have suggested that nm 23 (a gene expressed in normal cells and may inhibit infiltration) could be used as a prognostic marker. Negative giant cell tumors are more aggressive and are associated with a higher recurrence rate [18,19] found in the 53 out of 91 patients with TCGGS that the joint from which the tumor originated or that closest to the tumor mass, had clinical and / or radiological evidence of traumatic or idiopathic degenerative lesion.

The authors suggested that the damaged joint could predispose to the accumulation of histiocytes either in the synovial joint or in adjacent tendon sheaths. Under certain conditions, these histiocytes could proliferate, undergo metaplasia and produce swelling with a histological appearance of giant cell tumor of the tendon sheath. Giant cells, fibroblasts or epithelioid cells are generally predominant and constitute a large proportion of the tumor mass. In addition, the synovial joint and tendon sheath

may harbor tumors, which are locally aggressive, but most often painless, with limited growth potential [18]. In spite of this, the presence of a degenerative lesion of adjacent joints increases the difficulty of achieving complete surgical excision of the tumor [16]. Al-Qattan studied 43 TCGGS cases with a four-year follow-up (two-six years). Of 30 encapsulated tumors, no recurrence was noted whereas on 13 non-encapsulated tumors there were five recurrences. The surgical treatment of TCG has a dual purpose: diagnostic and therapeutic. Indeed, it allows to confirm the anatomopathological diagnosis of the lesion on the one hand and the oncological remission of another. The treatment is always surgical which consists of an excision of the tumor in totality. This excision must be meticulous and complete to prevent tumor recurrence. According to Suresh et al, incomplete excision of the tumor, due to insufficient surgical technique, is the most important cause of recurrence [20]. Adequate surgical exposure, meticulous dissection and the use of a microscope during excision are necessary to reduce the rate of recurrence. This rate is significantly reduced if patients receive adjuvant radiotherapy [20].

In our series, recurrence is most likely due to incomplete excision. Finally, it can be said that a rigorous assessment of the relative risk of recurrence of this type of tumor will make it possible to plan appropriate surgery and to inform patients about the risk of recurrence. Radiotherapy has been proposed following the difficult and sometimes incomplete excision of tumors with intra-articular extensions, and even intraosseous or around the collateral pedicles or in the presence of a high mitotic activity on histological examination for prevent recidivism [16]. Kotwal et al reported only 4% recurrence after adjuvant radiotherapy. However, for Ozalp, et al. Excision is suggested even after several recurrences, and radiotherapy has no place even for recurrent tumors [21].

## Conclusion

The Giant Cell Tumour of bone (GCT) is a locally aggressive intraosseous neoplasm of obscure biological behaviour. Although well defined in clinical, radiological and histological terms, detailed information on its biological development is still relatively incomplete. Despite the benign histology of TCGS, its local recurrence capacity makes it a tumor with intermediate malignancy. Recurrence depends on the nodular or diffuse nature of the tumor, the histological characteristics and the completeness or non-completeness of the resection, hence the need for postoperative monitoring.

## References

1. Messoudi A, Fini S, Labsaili N, Ghrib S, Rafai M, et al. (2007) Les tumeurs à cellules géantes des gaines synoviales de la main : à propos de 32 cas. *Chirurgie de la main* 26 : 165-169.
2. Fotiadis E, Papadopoulos A, Svarnas T, Akritopoulos P, Sachinis NP, et al. (2011) Giant cell tumor of tendon sheath of the digits: a systematic review. *Hand (NY)* 6: 244-249.
3. Wang CS, Duan Q, Xue YJ, Huang XM, Wang LL, et al. (2015) Giant cell tumour of tendon sheath with bone invasion in extremities: analysis of clinical and imaging findings. *Radiol Med* 120: 745-752.
4. Lautenbach M, Kim S, Millrose M, Eisenschenk A (2013) Nodular giant cell tumour of the tendon sheath of the hand: analysis of eighty-four cases: diagnostic decisions and outcome. *Int Orthop* 37: 2211-2215.
5. Jones KB, Deyoung BR, Morcuendo JA, Buckwalter JA (2006) Ethanol as local adjuvant for giant cell tumor of bone. *Iowa Orthop J* 26: 69-76.
6. Athanasou NA, Quinn J, Ferguson DJP, Mcgee Jo'D (1991) Bone resorption by macrophage polykaryons of giant cell tumor of tendon sheath. *Br J Cancer* 63: 527-533.
7. Park JW, Beckenbaugh RR, Amrami KK (2006) Calcifying aponevrotic fibroma of the hand: radiologic differentiation from giant cell tumors of the tendon sheath. *J Hand Surg Am* 31: 1024-1028.
8. Watanabe K, Tajino T, Sekiguchi M, Suzuki T (2000) h-Caldesmon as a specific marker for smooth muscle tumors. Comparison with other smooth muscle markers in bone tumors. *Am J Clin Pathol* 113: 663-668.
9. Rosai J (1996) Bone and joints. In: Rosai J, editor. *Ackerman's surgical pathology*. StLouis : Mosby 1996: 1917-2020.
10. Fletcher CD, Unni KK, Mertens F (2002) *Pathology and genetics of tumours of soft tissue and bone*. Lyon: IARC press 2002.
11. Bertoni F, Unni KK, Beabout JW, Ebersold MJ (1992) Giant cell tumor of the skull. *Cancer* 70: 1124-1132.
12. Fain JS, Unni KK, Beabout JW, Rock MG (1993) Nonepiphyseal giant cell tumor of the long bones. Clinical, radiologic, and pathologic study. *Cancer* 71: 3514-3519.
13. Biscaglia R, Bacchini P, Bertoni F (2000) Giant cell tumor of the bones of the hand and foot. *Cancer* 88: 2022-2032.
14. Nascimento AG, Huvos AG, Marcove RC (1979) Primary malignant giant cell tumor of bone: a study of eight cases and review of the literature. *Cancer* 44: 1393-1402.
15. Rao AS, Vigorita VJ (1984) Pigmented villonodular synovitis (giant cell tumor of the tendon sheath of the synovial membrane): a review of eighty one cases. *J Bone Joint Surg Am* 66: 76-94.
16. Al-qattan M (2001) Giant cell tumors of tendon sheath: Classification and recurrence rate. *J Hand Surg Br* 26: 72-75.
17. Kitagawa Y, Ito H, Yokoyama M, Sawaizumi T, Maeda S (2004) The effect of cellular proliferative activity on recurrence and local tumour extent of localized giant cell tumour of tendon sheath. *J Hand Surg Br* 29: 604-607.
18. Grover R, Grobbelaar AO, Richman PI, Smith PJ (1998) Measurement of invasive potential provides an accurate prognostic marker for giant cell tumour of tendon sheath. *J Hand Surg Br* 23: 728-731.
19. Ikeda K, Osamura N, Tomita K (2007) Giant cell tumour in the tendon sheath of the hand: importance of the type of lesion. *Scand J Plast Reconstr Surg Hand Surg* 41: 138-142.
20. Suresh SS, Zaki Hosam (2010) Giant cell tumor of tendon sheath: case series and review of literature. *J Hand Microsurg* 2: 67-71.
21. Ozalp T, Yercan H, Kurt C, Ozdemir O, Co'kunol E (2004) Giant cell tumors of the tendon sheath involving the hand or the wrist: analysis of 141 patients. *Acta Orthop Traumatol Turc* 38: 120-124.