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Localized-Type Tenosynovial Giant Cell Tumors of the Upper Extremity: A 20-Year Retrospective Study with 139 Patients

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Abstract

Objective: Localized-type tenosynovial giant cell tumor is a rare, benign tumor that originates from synovial joints, tendon sheaths, and bursae. It has 2 forms: diffuse and localized. The localized form is most commonly found in hand and knee joints. The aim of this study is to present and evaluate the behavior of the tumor, its recurrence rates, its distribution by gender and localization, and the demographic features of affected individuals.

Methods: In this study, charts of patients operated on localized-type tenosynovial giant cell tumors located at the upper extremity between 2000 and 2020 were retrospectively evaluated.

Results: A total of 139 patients underwent surgery for a localized-type tenosynovial giant cell tumor of the upper extremity, with 99 of them being female and 40 being male. The average age of the patients was 45.9 years. Among these cases, 114 (82%) were localized in the fingers, 13 (9.8%) were in the palmar and dorsal regions, 6 (4.3%) were in the wrists, 4 (1.88%) were in the elbows, and 1 (0.7%) was in the shoulder. The first finger was the most frequently affected location, accounting for 24.5% of cases, and the tumor was more commonly seen on the right hand. This study provides information on the tumor's location, its correlation with relapse, and any comorbidities that may be present.

Conclusion: Localized-type tenosynovial giant cell tumors are rare neoplasms. This study concludes that overuse of the extremities and female sex may be risk factors for developing the tumor.

Keywords: Hand tumor, soft tissue tumor, plastic surgery, hand surgery, microsurgery, orthopedic surgery

Introduction

Localized-type tenosynovial giant cell tumors (LTGCT) are the second most common soft tissue tumor after ganglion cysts. Initially thought to be reactive, they are now considered neoplastic. Localized-type tenosynovial giant cell tumors can also be referred to as pigmented villonodular synovitis and fibrous xanthoma. These tumors usually present as slow-growing, painless, and nontender masses, with rare instances of malignant transformation.[1] The diagnosis of LTGCTs is based on a combination of clinical, radiological, and histological findings. Imaging studies, such as X-rays, ultrasound, magnetic resonance imaging (MRI), and computed tomography (CT) scans, can help identify the location, extent, and characteristics of the tumor. A biopsy and histological analysis are needed to confirm the diagnosis. Treatment options for LTGCTs depend on various factors, including the size, location, subtype, and symptoms of the tumor. Surgical excision is the primary treatment for these tumors.

Pathologically, they can be classified as nodular or diffuse. The nodular type is commonly found on the volar side of the hand, particularly around the fingers, while the diffuse type is typically seen around large joints such as the knee, hip, wrist, and elbow.[2] A LTGCT are more common in women and are usually observed between the ages of 30 and 50,[3] with local recurrence rates ranging from 5%-30% in different series.[4]

In this study, we present a series of 139 patients with localized-type tenosynovial giant cell tumors localized in the upper extremity. To the best of our knowledge, this is one of the largest series of upper extremity LTGCT.

Methods

In this study, we retrospectively investigated the pathology reports of patients who underwent surgery for upper extremity masses and were diagnosed with localized-type tenosynovial giant cell tumors between January 2000 and December 2020. We recorded patient charts, demographic information, tumor behavior, recurrence rates, gender distribution, localization, and follow-up periods. The study was conducted with the approval of the institutional review board, and ethical clearance was obtained

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on September 23, 2020 (Approval Number: 58170629-604.01 .01-123664). Statistical analysis was performed using Statistical Package for the Social Sciences Statistics software, version 24.0 (IBM Corporation, Armonk, NY, USA). Associations between categorical variables were tested with $\chi 2$ analysis.

Results

The study included 139 patients diagnosed with localized-type tenosynovial giant cell tumors, with 99 of them being female and 40 being male (Table 1). The average age of the patients was 45.9 years. The location of the tumor was mainly on the right upper extremity (91 of 139 cases). Among the 139 cases, 100 were located in the first, second, and third fingers. Hypertension was the primary comorbid disease, affecting 66% of the patients. Diabetes mellitus was the second most common comorbid disease, affecting 40 out of 139 patients. Other comorbid diseases included hypothyroidism and coronary artery disease. Additionally, 25 out of 139 patients had a history of smoking. The most common location of the tumor was the third finger (36 of 139 cases), followed by the second (33 of 139) and first (31 of 139) fingers, respectively (Figure 1). While the tumor was rarely observed in the palmar region (6 of 139) and wrist (6 of 139), the rarest location of the tumor was the elbow (4 of 139) (Figure 2).

In our study of 139 patients who underwent surgery for primary lesions, recurrence was detected in 7 cases. Six of these patients were female, and only 1 was male. It was observed that in the majority of relapse cases (4 out of 7), the primary lesions were excised from the fourth finger. In 2 female patients, relapse occurred in lesions on the third finger, while in 1 female patient, the relapse was observed in a lesion located in the palm. Although it seems that female gender is a risk factor for recurrence, statistical analysis did not reveal any statistical significance ($P = .72$).

Table 1. Demographic Data, Localizations, and Accompanying Comorbidities of Patients

Characteristics	Patients, n	Rate, %
Sex		
Male	40	28.8
Female	99	71.2
Location		
Right	91	65.5
Left	48	34.5
Finger		
1	31	22.3
2	33	23.7
3	36	25.9
4	6	4.3
5	8	5.8
Smoker	25	18
Comorbidities		
Hypertension	91	66.6
Diabetes mellitus	40	28.8
Hypothyroidism	3	2.1
Chronic obstructive pulmonary disease	6	4.2
Other	24	16.8
Recurrence	7	5.0

Discussion

Localized-type tenosynovial giant cell tumor, previously known as giant cell tumor of the tendon sheath, was first described as fibrous xanthoma in 1952 and later referred to as fibrous histiocytoma of the synovium.[4] Our study included 139 patients with LTGCT, and around 70% of them were female, with an average age of 45.9 (ranging from 9 to 80 years). Previous studies have also reported that LTGCT grows slowly and is usually seen in individuals aged between 30 and 50, which is consistent with our findings.[5] Grazia et al found that the average age of patients with LTGCT was 45, and 65% of their patients were female. Our study yielded similar results, including the localization of the tumor, which was more common in the first 3 fingers, and recurrence rates of 4.7%.[6] However, our study had the advantage of a larger

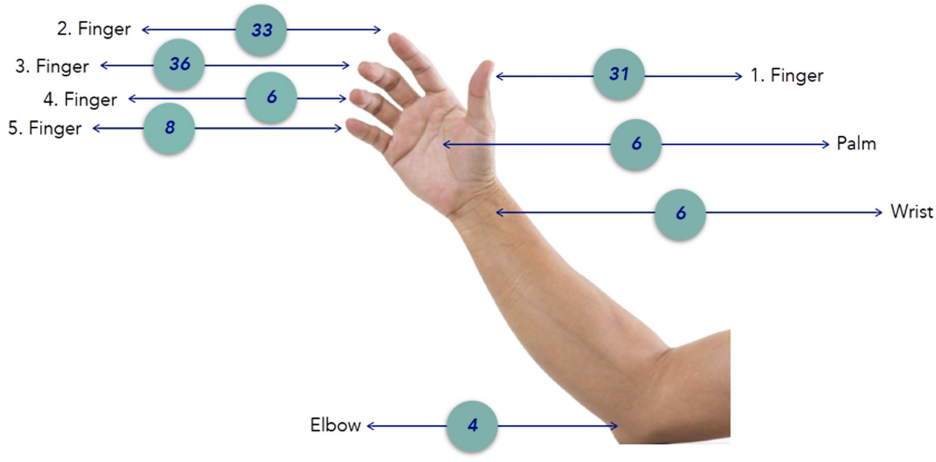

Figure 1. Schematic distribution of tumors of 139 patients diagnosed with localized-type tenosynovial giant cell tumors of the upper extremity.

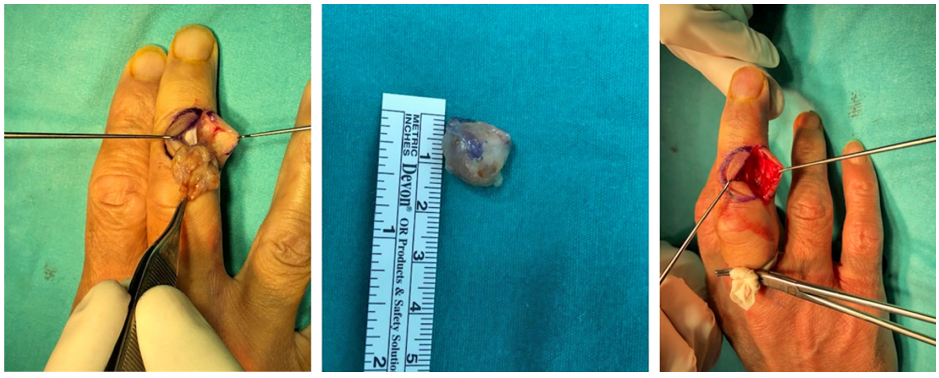

Figure 2. Intraoperative view of a patient with a pathologically confirmed localized-type tenosynovial giant cell tumor on the third finger of the left hand.

sample size than previous studies, which had examined smaller patient samples.[6,7]

Our study revealed that the tumor was commonly located on the right side with a 2:1 frequency and most commonly localized in the fingers. Nearly 80% correlation was found between the dominant hand of the patients and the side where the tumor was most common, which is consistent with previous studies.[8-10] Furthermore, when examining the distribution of LTGCT in more detail, we observed an obvious excess in the dominant fingers as well as in the dominant hand. LTGCT are also more commonly found in the first, second, and third fingers rather than in the other 2 fingers. These findings suggest a tendency for LTGCT to occur on the more used side. Therefore, it can be concluded that the emergence of LTGCT, which is actually a connective tissue disease, on the overused side may be related to the excess of degenerated connective tissue over time.

The primary approach to treating localized tenosynovial giant cell tumors is surgical resection with meticulous care. Previous studies have suggested that a thorough cleaning of the tendon sheath, the removal of bone erosions, and the use of magnifying glasses during surgery can reduce the likelihood of recurrence. Radiotherapy has also been shown to be effective in decreasing recurrence rates, as demonstrated by Sullivan et al and Kotwall et al, who reported recurrence rates of 4% and 25% in their respective studies.[11,12] In our study, the recurrence rate was 5.04%, which is considered acceptable. We believe that the higher recurrence rates of the tumor in the fourth digit are simply due to the low number of samples, since there were only 6 cases, of which 4 recurred. Although postoperative radiotherapy has been shown to significantly reduce recurrence rates, none of the patients in our study underwent this treatment.[13] Therefore, we believe that the efficacy of radiotherapy remains controversial given our low recurrence rate without using this treatment.

There are also limitations to our study, with the biggest one being its retrospective nature. Our center is a tertiary health-care center, where patients are generally referred to from various cities. Combined, a retrospective study with such heterogeneity limits surveillance of both patients and the tumor. Although our series is one of the largest, the sample size is still not sufficient for a risk factor analysis. Larger series with a prospective design are needed.

Conclusion

Overall, this study analyzed 139 patients with pathologically verified LTGCT, providing insights into their location, correlation with relapse, and comorbidities. The study identified overused extremities, female gender, and connective tissue disorders as risk factors for this condition. Furthermore, the study found that recurrence rates in localized-type tenosynovial giant cell tumors are considerably high, and the efficacy of using radiotherapy as a treatment method remains controversial.

Ethics Committee Approval: Ethical committee approval was received from the Ethics Committee of İstanbul University-Cerrahpaşa (Approval no: 58170629-604.01.01-123664, Date: September 23, 2020).

Informed Consent: Written informed consent was obtained from the patients who agreed to take part in the study.

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Declaration of Interests: The authors have no conflict of interest to declare.

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