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Giant Cell Tumor of Tendon Sheath: Prospective Study of 20 Patients

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Abstract

Introduction: Giant cell tumor of the tendon sheath (GCTTS) is a solitary benign soft- tissue tumor. It is the second most common tumor of the hand. GCTTS is most commonly found in the distal interphalangeal (DIP) joint of hand and very common in 4th decade of life. Our diligent search through available literature could not find any consensus on the etiology, prognostic factors and treatment modalities of GCTTS. We did prospective study of twenty patients with GCTTS of hand who underwent complete excision of the tumour, which was first of such kind of study. *Methods:* A prospective non-randomized study of twenty patients with GCTTS of hand, who underwent excision of the tumor was done. ompilation of the parameters were done which included, age and gender of patient, presentation of tumor, tumor size and location. Pre-operative X-ray of hand and wrist, ultrasonography and/or MRI whenever required was done. FNAC findings and post operative histopathological reports were recorded carefully. All the patients were followed up at months 3 and 6 and year 1 post-operatively, and recurrence rate was carefully noted. *Results:* In our study of 20 patients, 13 were female and 7 were female. The mean age of patients was 44 years. Patients presented, an average 14 months after initial onset of symptoms. Size of tumor on clinical examination was mean diameter of 3.4 cm in its greatest dimension. Most common location of the tumor was the palmar aspect of the hand on the thumb followed by little finger. Four patients had neurovascular bundle involvement who presented with predominant pain and swelling. All patients underwent surgical excision without adjuvant radiotherapy. No surgical complications were noted in any of the cases.In follow up study up to the 1 year, 2 patients had recurrence.

Keywords: Wrist Thumb Radiotherapy; Adjuvant Giant Cell Tumors Fingers Tendons.

Introduction

Giant cell tumor of the tendon sheath (GCTTS) is a solitary benign soft- tissue tumor. It is the second most common tumor of the hand after ganglion cysts [1]. Chassaignac in 1852 described it first time as fibrous xanthoma [2]. It's been referred by multiple names since then, such as localized nodular tenosynovitis, pigmented villonodular proliferative synovitis, sclerosing hemangioma, benign synovioma, proliferative synovitis, xanthoma, xanthogranuloma, xanthosarcoma, myeloid endothelioma, fibrohemosideric sarcoma, giant cell fibrohemangioma, pigmented villonodular tenosynovitis, fibroma, myeloma, myeloxanthoma, and fibrous histiocytoma [1,3].

Giant cell tumor of the tendon sheath is most commonly found in the distal interphalangeal (DIP) joint of hand [4,5]. Presentation of this tumor is more common in fourth and fifth decades of life with women preponderance [2,3].

Our diligent search through available literature could not find any consensus on the etiology, prognostic factors and treatment modalities of giant cell tumors of the tendon sheath.

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We did prospective study of twenty patients with GCTTS of hand who underwent complete excision of the tumour. This study was undertaken to record the etiology, epidemiology, different presentations, gross and microscopic characteristics and treatment modalities of GCTTS of hand.

Materials and Methods

A prospective non-randomized study of twenty patients with GCTTS of hand, who underwent excision of the tumor was carried out in our hospital between May 2014 and May 2017. Demographic details of all patients were recorded as per protocol.

Detail history and compilation of the parameters were done which included, age and gender of patient, presentation of tumor, tumor size and location. Pre-operative X-ray of hand and wrist , ultrasonography and/or MRI whenever required was done. Pre-operative FNAC findings and post operative histopathological reports were carefully compiled. Modality of treatment were recorded. All the patients were followed up at months 3 and 6 and year 1, and recurrence rate was carefully noted.

Data was complied in a database using Microsoft Excel. Data analysis was used to analyze various factors and associations.

Results

In our study of twenty patients, 13 were female and 7 were female. The mean age of patients was 44 (range: 24–76 years). Among twenty patients, 66% patients were >40 years of age and remaining 34% were less than 40 years of age.

Table 1: Distribution of presentation at the time of diagnosis

Complaints	Number of patients
Swelling	20
Associated pain	4
Associated numbness	2
Bony erosion	7
Degenerative joint disease	2
Intra-osseous invasion	1
Multiple swellings	0

Table 2: Distribution of location of tumor

Location	Number of patients
Thumb	9
Index finger	4
Middle finger	1
Ring finger	1
Little finger	5

Patients in our study presented, an average 14 months after initial onset of symptoms (range: 2 – 72 months). All patients presented with gradually progressive swelling over hand and 4 of them also had an associated pain [Table 1].

We found that, none of our patient gave any history of trauma. Numbness or tingling and degenerative joint disease were observed in 2 patients each [Table 1] Bony erosion were noted in 7 patients.

The affected hand was dominant in 82% of cases and it was right in all of them. Size of tumor on clinical examination at the time of presentation was mean diameter of 3.4 cm in its greatest dimension (range: 1.8 – 7.1 cm).

The tumor was most often located on the palmar aspect of the hand in our study and it was observed in 14 patients whereas, 6 patients had tumor location on dorsal aspect of hand [Figure 1]. We did not encounter any tumor specifically located on radial or ulnar aspect of hand.

The most common digital location of tumor was the thumb followed by little finger, which was seen in 9 and 5 patients respectively [Figure 1,2]. The most common area of tumor was DIP joint, in 12 patients. [Table 2].

Pre-operative X-ray of hand and MRI in our study revealed that 7 patients had bony erosion and 1 also had an intra-osseous invasion of tumor [Figure 3]. We noticed that in all these 8 cases, size of tumor at the time of presentation was more than 5 cm diameter in its greatest dimension.

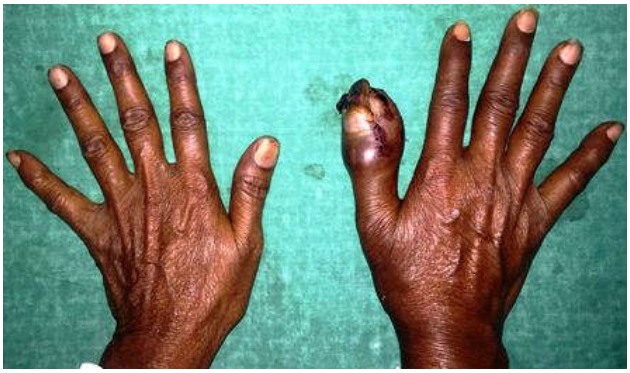

Fig. 1: Right Thumb GCTTS

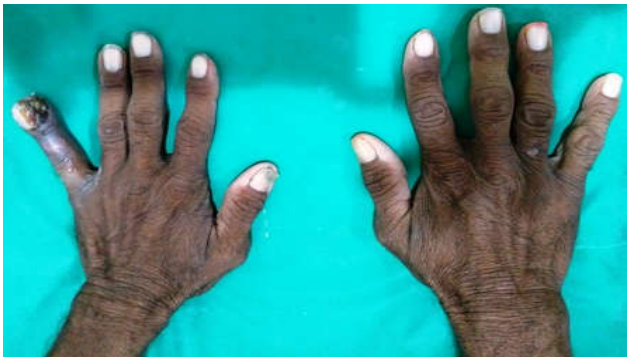

Fig. 2: Left Little finger GCTTS

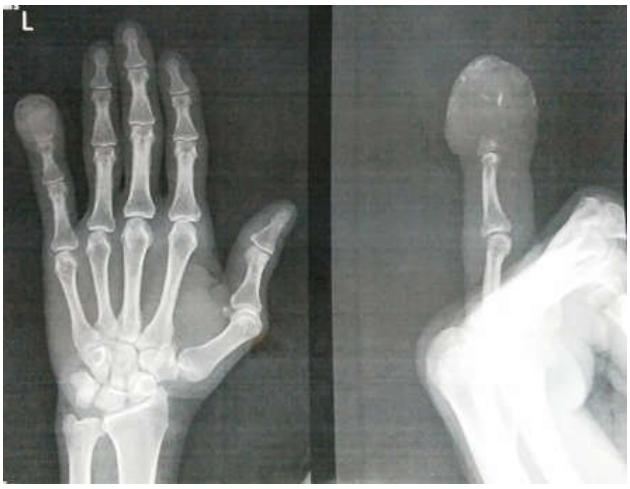

Fig. 3: Left Little finger GCTTS with bony erosion and destruction

Pre-operative FNAC of swelling showed typical mitosis in 15 patients. Smears were evident of cluster of epithelial cells with eosinophilic cytoplasm and round to oval nuclei with mild pleomorphism. It also showed, multinucleated giant cells with abundant foamy macrophages suggestive of GCTTS. None of the FNAC report were suggestive of any calcification, cartilage debris or necrosis. Four patients had neurovascular bundle involvement who presented with predominant pain and swelling.

All patients underwent surgery under brachial plexus block. Modified racket incision was taken whenever it was possible. Dorsal digital nerves and digital artery were sacrificed with flexor and extensor tendons in 6 cases. Disarticulation was required in 6 cases at DIP joint due to extensive involvement of soft tissue, flexor and extensor tendons and osseous erosion of distal phalynx. Primary skin closure was done in all cases. No surgical complications were noted in any of the cases.

In our follow up study up to the 1 year, we found that 2 patients had recurrence. One patient experienced recurrence after 6 months whereas, in 2nd case it was noticed on first follow up it self at 3 months. Both patients underwent subsequent removal of the tumor.

Discussion

Giant cell tumors of the tendon sheath (GCTTS) is a solitary benign soft- tissue tumor closely resembling with giant cell tumors of the bones [6]. Chassaignac was the first one to describe it as fibrous xanthoma [2]. Salm and Sissons however, were the first ones to name this entity as "Giant-cell tumors of soft tissues" in 1972 [7]. The exact etiology and pathogenesis of GCTTS is still unclear, but there has been a better understanding of the pathogenesis and its association with recurrence in recent years.

In our study of 20 patients, we found that GCTTS was 65% more common in female than male and mean age of patients was 44 years. Adams et al in their study of 65 patients found that, tumor was common in female and it was seen in 35 female patients with mean age of 49 years [8]. According another study, tumor was most commonly diagnosed in the fourth and fifth decades of life [9,10] with women affected more commonly than men [2,3,5].

We found that mean duration of symptoms in our cases was, 14 months (range: 2 – 72 months). In a review outcomes of 106 patients with GCTTS, Garg et al observed the mean duration of presentation was 21 months (range: 2–96 months) [11]. According to the one of largest series of GCTTS in children, patients presented an average of 6 months to 2.5 years after initial onset, as this tumor is slow-growing in nature [12].

We found that, patients with GCTTS presented with varied complaints such as swelling, associated numbness/tingling, tenderness and even bony erosion or destruction at times. Gradually progressive swelling over hand was however, the main complaint which gathered the attention of patient towards it and forced them to visit the doctor, in our as well as in other available studies [7-12]. Our observation was that, patients with large sized swelling predominantly more than >3 cm and with bony erosion or destruction also complained associated pain due to involvement of neurovascular bundle.

The affected hand was dominant in 67% of cases and was the right hand in 65% of cases in one study [8]. We found that, affected hand was dominant in 82 % of cases and it was right in all of them. Size of tumor in our study varied from 1.8 cm to 7.1 cm in diameter in its greatest dimension with mean being 3.4 cm. Another studyfound that the mean diameter of the tumor was 1.5 cm (range: 0.5 - 3.5 cm) [8].

Seventy percentages of the tumors in our study was located on the palmar aspect of the hand whereas, it was located on dorsal aspect only in 30% patients. Garg et al. found similar findings in their 77 patients, where tumor was located on palmar aspect in 86% cases [11]. Another large study mentions tumor being located on the palmar aspect of the hand in 49% and on dorsal aspect in 35% patients [8]. This observation from different studies can lead to conclusion that palmar aspect of hand being the most common location of GCTTS.

We noticed that, there is a vast difference in observations about the most common digital location of the GCTTS. The most common digital location in our study was the thumb followed by little finger, which was seen in 45% and 25% patients respectively. The most common area of tumor was DIP joint, in 60% patients. Admas et al however, noted that the index finger (37%) was the most common area in which the tumor was located.[8] Another study also

found that index finer was the most common location of tumor and DIP was the most common involved joint is their patients [7-9,11-13].

Peculiar location of the GCTTS makes preoperative diagnosis tricy. The variability of a preoperative diagnosis of GCTTS in available literature shows a discrepancy in diagnostic techniques and/or knowledge of tumor presentation. Different available literature mentions pre-operatvie X-ray, MRI and FNAC of the tumor for diagnosis, but none of them gives the consensus about the same. The diagnosis was made with the help of X-ray and/or MRI in 98 patients and by histopathological examination in only 8 patients, in Garg et al study of their 106 cases [11]. We performed pre-operative X-ray of hand and MRI as well as FNAC in all our patients for better understanding of the pathogenesis of GCTTS.

Glowacki et al. mentioned that GCTTS had no characteristic radiographic appearance and indicated only a soft tissue mass in the majority of their cases [5]. Reilly et al even found normal findings in 32% of their patients [14] We also found similar findings on X-ray, other than in 8 cases where we could visualize the bony erosion and destruction. Few studies also performed ultrasonography in their study, findings of which showed solid, homogeneous, hypoechoic mass with increased vascularity on doppler studies [15].

Magnetic resonance imaging usually reveals decreased signal intensity on T1- and T2-weighted images [5]. We revealed that bony erosion and intra-osseous invasion of tumor was better visualised by MRI than X-ray and it was the most definitive imaging study. We emphasize to perform MRI in all cases of GCTTS, whenever and wherever the cost-effectiveness is not an issue. It will also help to understand the exact extend of the tumor and better planning of surgery.

Darwish et al found that preoperative pathological diagnosis of GCTTS can be made best by using fine-needle aspiration biopsy [16]. In our study, pre-operative FNAC of tumor showed typical mitosis in 15 patients. Smears revealed cluster of epithelial cells with eosinophilic cytoplasm and round to oval nuclei with mild pleomorphism in all 20 cases. It also showed, multinucleated giant cells with abundant foamy macrophages suggestive of GCTTS in 17 cases. All these findings were comparable to other studies [8,11,16].

Recurrence rates of GCTTS have ranged from 0% to 44% according Monaghan et al. [17] whereas, some other studies mentions recurrence rate between 25 to 45% after excision [2,18,19]. One study postulates that, balance of minimal excision of a benign process with maintenance of function in a highly important location such as a hand can be one of the reason for recurrence after pure surgical intervention [8]. Adjuvant therapies such as phenol treatment [4] and radiation therapy [11,19] have been described to lower the recurrence rates in some of the available literature. We couldn't find exact consensus for radiotherapy usage in such patients and hence, we decided to undergo only surgery in all our 20 cases and recurrence rate in our study was 10% up to 1 year follow up.

Garg et al found that adjuvant radiotherapy with total dosage of 15–25 Gy in a fraction of 1.5 Gy daily, would prevent the recurrence and hence they delivered postoperative radiotherapy in their high risk patients [11]. They defined the case as high risk for recurrence whenever, histopathological report was evident of cells in mitosis. Our diligent search could not find any other such study supporting the role of radiotherapy in GCTTS.

We performed surgery in all our 20 cases, under brachial plexus block. We had to sacrifice dorsal digital nerves and digital artery were with flexor and extensor tendons in 6 cases. Our primary goal while operating in these cases was to excise the whole tumor with minimal disruption to adjacent structures and preservation of anatomical structures. Adams et al, rightly mentioned that recurrence is very closely related to the incomplete excision of the tumor or satellite lesions [8]. Salvaging tissue and adequately removing tumor margins has to be the main aim while operating on GCTTS.

Distal joint location of the tumor and direct invasion of the tumor intothe extensor tendon, flexor tendon, or joint capsule has increase risk of recurrence [20]. The likely reason for this being difficult excision of such tumors and hence, operating surgeon has to be more aggressive in removing them.

Conclusion

After reporting first case of GCTTS and reviewing the available literature, we decided to undertake a first such kind of prospective study of GCTTS and its presentations in our institute. In our diligent search we could only find retrospective studies about such tumors. Although, our data was inclusive of only 20 patients we carefully compiled the clinical history, suspected etiology and applied the same diagnostic modalities and treatment for all 20 cases in our study.

We observed that, patients with GCTTS tended to present late as only initial complain was progressive swelling in such cases. Those presenting with associated pain with swelling either had bony erosion/destruction or there was an involvement of neurovascular bundle. Such variants of GCTTS needs prompt attention and speedy diagnosis and aggressive surgical approach is needed in these cases.

Pre-operative diagnosis of GCTTS can be difficult and challenging for surgeon. Our experience in or 20 patients and from available literature we must say that, we can not rely on X-ray for confirmatory

diagnosis. MRI seems to be the mainstay of diagnosis according us and it is must before any further intervention.

Treatment of GCTTS ,irrespective of its size and nature remains the topic of discussion. Some schools of thought like us believes that, meticulous surgical excision of tumor with safe margin is sufficient and prevent the recurrence whereas, some authors delivered radiotherapy post-operatively and found that recurrence rate was lesser. However, we couldn't find role of pure neo-adjuvant radiotherapy in GCTTS as well as consensus about treatment modalities.

We think that further prospective research is necessary which could entail a better knowledge about GCTTS its etiology, diagnosis and best available treatment to prevent recurrence, till then surgeon should emphasise on curative surgery for GCTTS.

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