RESEARCH ARTICLE

Total Synovectomy and Bone Grafting/Cementation after Curettage of the Bone Lesion in Diffuse Type of Tenosynovial Giant Cell Tumor: A Retrospective Cohort Study

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Abstract

Objectives: Although the diffuse type of tenosynovial giant cell tumor (D-TGCT) is rare, bone involvement is common in such lesions. However, the optimal management of bone lesions in D-TGCT is not well-described. In this study, we reported the outcomes of total synovectomy, curettage, and bone grafting/cementation in the treatment of D-TGCT with subchondral bone involvement. We also described the prevalence, demographic, and characteristic features of the lesions.

Methods: In a retrospective study, we included 13 patients with D-TGCT of large joints and associated subchondral cyst/cyst-like bone lesions of ≥ 5 mm that were managed with total synovectomy and curettage. Cavities with a bone defect of ≤ 30 mm (n=12) were filled with bone grafts. Cavities of > 30 mm (n=1) were augmented with bone cement. The limb function was evaluated by the Musculoskeletal Tumor Society (MSTS) score.

Results: The study population consisted of 6 (46.1%) males and 7 (53.9%) females with a mean age of 30 ± 7.9 years. The most frequent sites of involvement were the knees and ankle joints (n=5 each, 38.5%). The mean follow-up of the patients was 69.2 ± 32.9 months. The mean MSTS score of the patients was obtained at 98.2 ± 3.2 (range 90-100). The D-TGCT recurred in two patients, both of which were in the synovium. Postoperative complications were three cases of transient pain and one case of knee joint stiffness. While no patient had an osteoarthritic change in preoperative radiographs, two patients had osteoarthritic change (grade II) in the last follow-up, one in the knee and one in the hip.

Conclusion: Curettage and filling the defect with bone graft or cement are adequate treatments for managing bone lesions in D-TGCT.

Level of evidence: IV

Keywords: Bone lesion, Curettage, Diffuse tenosynovial giant cell tumor, Pigmented villonodular synovitis

Introduction

Tenosynovial giant cell tumor is a rare, benign, but potentially aggressive lesion characterized by exuberant proliferation of synovium and pigment deposition (hemosiderin) inside the joints, tendon sheaths, and bursae.[1] According to the World Health Organization classification in 2013, tenosynovial giant cell tumor is divided into two subgroups, namely localized (nodular tenosynovitis) and diffuse. The localized form involves tendon sheaths, bursas, and small joints. Diffuse type, which was first coined by Jaffe et al. in 1941 as pigmented villonodular synovitis (PVNS), mainly affects large joints, such as the knee, hip, ankle, and elbow joints, with an annual incidence of about 1.8 per million.[2] The diffuse type of tenosynovial giant cell tumor (D-TGCT) is more

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aggressive and destructive than the local type, with a recurrence rate between 21% and 50%.[3,4] The treatment of choice for D-TGCT is surgical excision with total synovectomy of the involved joint, either through an open or arthroscopic procedure.[5]

The bone lesion is not uncommon among patients with D-TGCT and has been reported to be present in up to 51% of the patients.[5] It could be presented in the form of marginal pressure erosions and/or subchondral cysts/cyst-like areas surrounded by thin sclerosis.[6,7] Despite the high prevalence of bone lesions in D-TGCT, only small numbers of studies are available regarding its management. Bone invasion in D-TGCT is generally treated with curettage and filling of intraosseous lesions with bone graft.[8,9]

In this study, we evaluated the outcomes of total synovectomy, curettage, and bone grafting/cementation in a series of 16 D-TGCT patients with subchondral cysts/cyst-like bone involvement. We also described the prevalence, demographic, and characteristic features of the lesions.

Materials and Methods

This study was approved by the review board of our institute under the code IR.IUMS.REC.1400.626. A comprehensive agreement for the academic use of the information, such as the type of treatments, treatment progress, or any other data acquired during their treatments, was obtained from the patients by the hospital at the time of their hospitalization, and no identifiable information of the participants was included in the manuscript.

Between 2009 and 2020, medical profiles of the D-TGCT patients who underwent surgical treatment in our university hospital were retrospectively reviewed. Inclusion criteria were D-TGCT with bone involvement, absence of degenerative joint disease at presentation, presence of a subchondral cyst, lesion size of ≥ 5 mm, treatment with synovectomy, curettage bone grafting/cementation, and a minimum follow-up of 24 months. Patients with inadequate medical records and those who were lost to follow-up were excluded from the study. From a total of 75 D-TGCT patients, bone involvement was present in 38 (50.6%) patients. In 5 (6.6%) patients, bone involvement was in the form of marginal pressure erosion, and in the remaining 33 (44%), it was in the form of cyst/cyst-like lesions in the computed tomography (CT) scans. Twenty patients did not meet the study criteria. Finally, 13 patients were identified as eligible for the study [Figure 1].

A sonography-guided core needle biopsy was carried out for five patients with inconclusive imaging diagnoses. The final diagnosis was confirmed by histopathologic evaluation of the extracted specimen in all patients.

Demographic, clinical, and tumor characteristics of the patients were extracted from their medical records. Imaging characteristics of the patients were investigated on the anteroposterior and lateral radiographs, CT scans, and magnetic resonance imaging (MRI) of the lesion.

For knee involvement, a two-stage, extensile open arthrotomy, as described previously,[10] was performed. Accordingly, in a prone position with lazy-s skin incision, we incised two heads of gastrocnemius about 5 mm from their origin. With the protection of the neurovascular bundle, a longitudinal arthrotomy was undertaken.

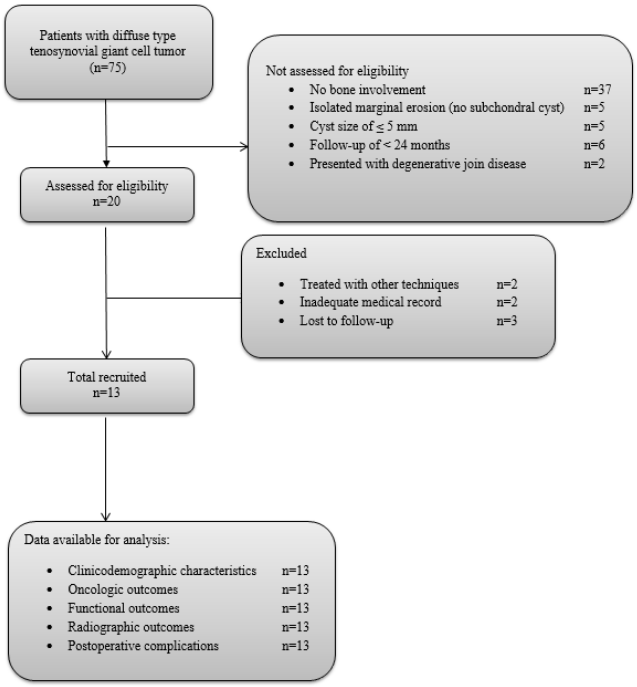

Figure 1. Flow diagram of the study

During the excision of the tumor and synovium, we tried to protect the posterior cruciate and meniscotibial ligaments. After obtaining a full range of motion, the second stage of anterior synovectomy was performed with an anteromedial or anterolateral parapatellar approach, depending on the location of the bone lesion. For the hip, surgical dislocation of the hip was employed, as previously described by Gans et al.11 [Figure 2]. The shoulder was approached through a standard deltopectoral approach. Ankle involvement was managed through simultaneous anterolateral and posteromedial approaches. The bone lesion was managed by extended curettage12 through an extra-articular window [Figure 3]. Cavities with a bone defect of ≤ 30 mm (n=12) were filled with cancellous fresh frozen allograft (mostly from the femoral head). To provide stronger augmentation, cavities of > 30 mm (n=1) were augmented with bone cement [Figure 4].

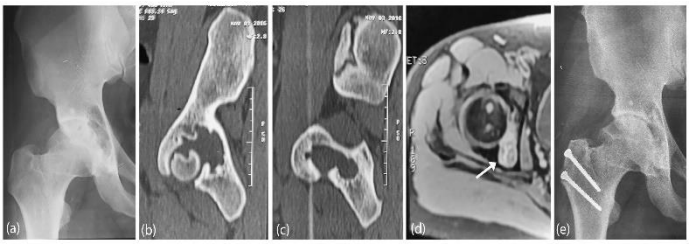

Figure 2. (a) Preoperative anteroposterior hip radiograph of a 33-year-old male with D-TGCT and associated bone cyst in the ilium and femoral neck; (b) Coronal reconstruction CT scan showing the bone cyst in the ilium; (c) Coronal reconstruction CT scan showing the bone cyst in the femoral neck; (d) T2-weighted fact suppressed axial MRI showing the ischium defect (white arrow) before treatment by synovectomy, extended curettage, and bone grafting; (e) Anteroposterior hip radiograph of the four years after the operation showing grade II osteoarthritic change of the hip joint

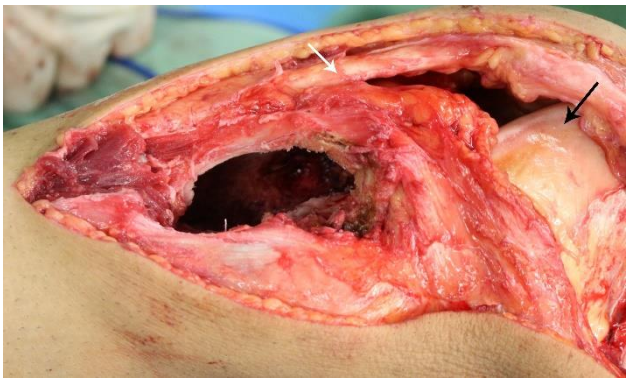

Figure 3. Intraoperative photograph showing extended curettage through creating an extra-articular window in the proximal tibia; Black arrow is showing the lateral femoral condyle and white arrow is showing the patellar tendon.

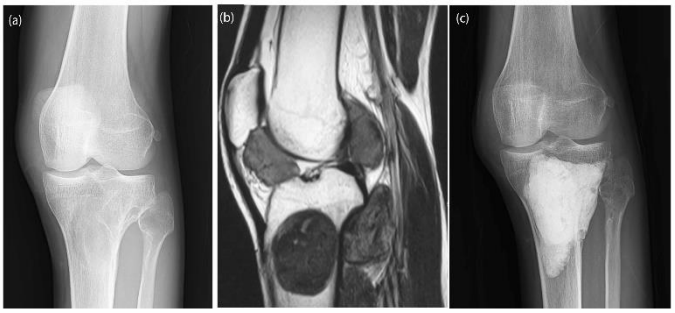

Figure 4. (a) Preoperative anteroposterior knee radiograph of a 40-year-old male with D-TGCT and an associated tibial bone cyst; (b) T1-weighted fact suppressed sagittal MRI showing a large bone defect in the proximal tibia; (c) Anteroposterior knee radiograph of three years after the treatment with synovectomy, extended curettage, and cementation.

Isometric quadriceps exercises were started two days after the operation. Passive mobilization was started after two weeks. Patients with lower extremity involvement were kept on partial weight-bearing for three weeks with two crutches and one crutch for another three weeks. Follow-up visits were performed every three months for the first two years, every six months for the next three years, and yearly thereafter.

The largest diameter of the cavity in the axial CT scan (Siemens SOMATOM Sensation 16 Slice CT scanner, slice thickness of 1.5 mm, Forchheim, Germany) was regarded as the size of the lesion. Functional outcomes of the limb were assessed through a chart review and scored using the Musculoskeletal Tumor Society (MSTS) score for the upper and lower limbs separately. MSTS scoring system includes six subscales with assigned 0-5 points each, making a total score ranging from 0 to 30. The final scores of the patients were presented as a percentage, where a higher score represented a better outcome.[13] the oncologic outcomes of the patients (local recurrence) were assessed clinically. If suspected, further investigation was done by obtaining MRI. Osteoarthritic changes were inspected on final postoperative radiographs and classified according to the Kellgren and Lawrence system.[14] Postoperative complications, including persistent pain and stiffness, were also extracted from the patients' profiles.

All the measurements were done by two fellowship-trained musculoskeletal tumor surgeons who were not involved in the patients' care. In case of a discrepancy between the two evaluators, a third evaluator was asked to check the case to reach a consensus.

Results

Thirteen patients with subchondral cysts/cyst-like bone lesions were included in this study. The study population consisted of 6 (46.1%) males and 7 (53.9%) females with a mean age of 30 ± 7.9 years (range 20-46). The most frequent sites of involvement were the knee and ankle joint (n=5 for each, 38.5%), followed by the hip (n=3, 23%). In 7 (53.8%) patients, both sides of the joint were involved. In the majority of bilateral joint involvements (6 of 7 cases), only one side was surgically managed because the size of the lesion on the other side was smaller than 5 mm. The mean size of the bone lesion at its largest diameter was 14.8 ± 9.5 mm (range 7-40). The main symptoms at presentation were swelling with or without associated pain. The mean symptom duration was 44.5 ± 37 months (range 3-120). The detailed characteristics of the patients are demonstrated in [Table 1].

The mean follow-up of the patients after synovectomy, curettage, and bone grafting/cementation was 69.2 ± 32.9 months (range 24-132). The mean MSTS score of the patients at the last follow-up was 98.2 ± 3.2% (range 90-100). In this respect, 9 patients had a full MSTS score (MSTS=100). The other four patients had MSTS scores of < 100, which was mainly attributed to postoperative complications, such as osteoarthritis, pain, and stiffness.

The D-TGCT recurred in 2 (15.4%) patients during the follow-up period. One of the recurrences was in the knee. The other one was in the ankle. The lesion only recurred in the synovium. These patients were treated with re-synovectomy and radiotherapy. No recurrence was observed in these patients afterward.

Three patients had postoperative pain, which did not resolve until the last follow-up. One patient had postoperative knee stiffness, which was resolved with arthroscopic release and physical therapy. While no osteoarthritic change was present in preoperative radiographs of the included cases, the osteoarthritic change (grade II) was observed in the postoperative radiographs of two patients, one in the knee and one in the hip. No intervention was performed in this regard.

Discussion

In this study, we evaluated the outcomes of synovectomy, curettage, and bone grafting/cementation in a series of 13 patients having D-TGCT with bone involvement. We generally filled the void cavity with fresh frozen allograft.

Table 1. Characteristic features and outcomes of D-TGCT patients with an associated bone lesion

Case	Gender	Age (year)	Involved limb	Symptoms	Involved bone	Lesion size (mm)	Symptom duration (month)	Follow-up (month)	Recurrence	Complication	*Postoperative arthritic change (KL grade)	MSTS
1	Male	40	Knee	Pain and swelling	Tibia	40	6	48	No	No	0	100
2	Male	46	Ankle	Pain and swelling	Talus	10	48	132	Yes	Pain	0	96.7
3	Female	21	Hip	Pain and swelling	Pelvis and femoral head	10 <5	3	60	No	No	0	100
4	Female	21	Ankle	Pain and swelling	Talus	15	24	48	No	No	0	100
5	Male	26	Ankle	Pain and swelling	Talus and tibia	10 <5	24	84	No	No	0	100
6	Male	37	Knee	Pain and swelling	Femur	15	96	60	No	No	II	100
7	Female	20	Knee	Swelling	Tibia and femur	15 <5	6	48	No	No	0	100
8	Female	31	Knee	Pain and swelling	Tibia	10	120	36	No	No	0	100
9	Female	27	Ankle	Swelling	Talus and tibia	10 <5	72	120	No	No	0	100
10	Female	34	Ankle	Pain and swelling	Talus and tibia	10 <5	72	72	No	Pain	0	93.3
11	Female	30	Hip	Swelling	Pelvis	7	12	108	No	No	0	100
12	Male	33	Hip	Pain	Pelvis and femoral head	20 30	48	60	No	Pain	II	90
13	Male	24	Knee	Pain	Tibia and femur	10 <5	48	24	Yes	Stiffness	0	96.7

However, large defects were filled with bone cement to avoid potential pathologic fracture with early initiation of the range of motion, even by accepting the risk of a more complex conversion to total joint arthroplasty, if required.[15] The lesion was successfully treated in 11 patients, while it recurred in the synovium of the two remaining cases. The functional outcomes of the patients were full or nearly full in all patients. Postoperative complications included three cases of transient pain and one case of stiffness. The osteoarthritic change was seen in the final radiographs of two patients with D-TGCT, one in the knee and the other in the hip.

D-TGCT, also termed diffuse PVNS, with the associated bone lesion, has been reported in a small number of earlier studies. The prevalence of PVNS with the associated bone lesion is reported to be 42% in a study by Nishida et al. and 51% in a study by Dorwart et al.[5] In the present study, the prevalence of bone lesion in PVNS was 50.6%, which was comparable with earlier studies. This evidence reveals that bone lesion is not as rare in D-TGCT as stated in earlier studies. PVNS generally shows a higher prevalence in the female population, and this pattern seems to be the same when the bone is involved. Accordingly, four of five patients in a study by Scott[15] and four out of seven patients in a study by Pantazopoulos et al.[16] Were female. Likewise, 7 out of 13 patients in the present study were female. The age range was 20 to 52 years in the study by Pantazopoulos et al.[16] The upper age limit was 38 years in the study by Scott[15]. The age range of patients in the current study was 20 to 46 years. Therefore, it seems that bone involvement of D-TGCT generally occurs in middle-aged patients. Scott[15] reported five cases of PVNS with the associated bone lesion that were located in the knee (n=2), hip (n=2), and elbow (n=1). Pantazopoulos et al.[16] Reported seven cases of PVNS with the associated bone lesion. Four lesions were located in the hip, while the remaining lesions were located in the knees, shoulders, and ankle joints. Similarly, in a study by Nishida et al.[17] (n=24), hip involvement was significantly higher than knee involvement. According to a review study by Dorwart et al, [5] erosive bone lesions were also more frequent in the hip. However, they also pointed out that it was difficult to accurately estimate the location of bone change because most of the published

series had not provided sufficient information in this regard. In the present study, the knee and ankle were the most frequent locations of bone involvement by PVNS (n=5 for each), followed by the hip (n=3). Although bone lesion was reported to be mainly large in the study by Scott, it was less discussed in earlier studies. The mean size of the bone lesion was 14.8 mm in the present study, ranging from 10 to 40 mm. We did not include cysts with bone lesions of < 5 mm in the present study because they were generally managed non-surgically. The main symptom of the patients at presentation was pain or swelling.[15,16] The same symptoms were noticed in our series. The symptom duration ranged from 5 months to 18 years in the study by Scott.[15] Nishida et al.[17] Associated the presence of bone lesions with significantly higher symptom duration so that patients with bone involvement had a mean symptom duration of 6.4 years, whereas this duration was 1.4 years in patients without a bone lesion[17]. In the present study, the symptom duration ranged from 3 months to 10 years. However, it was more than one year in the majority of patients (n=10), confirming the elongation of symptom duration discussed by Nishida et al.[17] Therapeutic strategies for the treatment of PVNS with bone involvement showed a remarkable variation in the earlier studies ranging from a partial to total synovectomy, curettage to wide resection, joint arthrodesis, bone grafting with or without cyst extraction, and combined radiotherapy.[15,16] However, the accumulative evidence suggests total synovectomy and curettage as the most efficient treatment approaches for these lesions[16] as they provide acceptable local control. Only two local recurrences were observed in patients of the present series. Persistent pain, osteoarthritic changes, and joint stiffness were among the postoperative complications in our series. These complications were more or less reported in earlier studies, depending on the type of surgical intervention.[15,16]

Altogether, the results of the present study, in line with the results of earlier studies, revealed that bone cysts of D-TGCT could be effectively managed with curettage and bone grafting/cementation.

In addition, the rate of bone involvement in D-TGCT was not as low as stated in some studies.[6] By contrast, its prevalence was high enough to be used as a diagnostic index in D-TGCT.[5,17]

The present study was not without limitations. The main limitation of the study was its retrospective design and the small number of patients. Therefore, future large-scale prospective studies are required to confirm the present findings.

Conclusion

The associated bone lesion is frequently detected in D-TGCT of large joints and can be adequately managed with curettage bone grafting/cementation. No serious postoperative complication was observed following this method of treatment. Therefore, curettage and bone grafting/cementation can be suggested as the treatment of choice for managing bone lesions in D-TGCT.

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