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The surgical outcome and recurrence rate of tenosynovial giant cell tumor in the elbow: a literature review

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Background: Tenosynovial giant cell tumor (TSGCT) is a rare proliferative disorder of the synovium. Because of its aggressive nature and recurrence potential, treatment of TSGCT involves surgical resection with or without synovectomy. There is currently a paucity of literature describing the surgical management of TSGCT at the elbow. The aim of this study was to evaluate clinical outcomes and recurrence rates following open and arthroscopic excision of TSGCT in the elbow.

Methods: Electronic databases were searched for relevant articles relating to surgical management of TSGCT of the elbow. We included all patients who received surgical treatment for TSGCT, with no age limitations. We excluded any nonsurgical treatment studies. Seventy-seven articles were identified for screening, and a total of 27 patients from 24 studies were included for the review.

Results: The patients' mean (standard deviation [SD]) age was 40.3 (21.7) years, and the most common presenting symptoms included pain (18/27, 66.7%), swelling (19/27, 70.4%), and decreased range of motion of the elbow (9/27, 33.3%). The majority of patients underwent open excision with or without synovectomy (23/27, 85.1%). Of those undergoing open procedures, 16 (16/23,69.6%) had diffuse TSGCT and 14 (14/23, 60.9%) remained symptom free for a mean (SD) follow-up of 38.9 (25.4) months. Four patients (4/27, 14.8%) were treated arthroscopically, all of whom had diffuse disease.

Conclusions: Our review found that open synovectomy appears to be an effective treatment for both localized and diffuse TSGCT in the elbow, and arthroscopic synovectomy is emerging as a method of surgical management for diffuse TSGCT. However, because of the limited number of patients undergoing surgery for TSGCT, further studies are needed to make a definite conclusion.

Level of evidence: Level IV; Systematic Review

Keywords: Tenosynovial giant cell; PVNS; elbow; arthroscopy; synovectomy; TSGCT

Tenosynovial giant cell tumor (TSGCT), previously known as pigmented villonodular synovitis (PVNS), is a rare idiopathic condition of the synovium.[13] The overall incidence is 1.8 per million.[10,23] The less common localized form is nodular, affecting a discrete area of a joint, bursa, or tendon sheath. However, the majority of TSGCT cases are the diffuse form, involving the intra-articular synovium of a large joint. The knee is the most commonly affected, followed by the hip, ankle, shoulder, and elbow.[10,23] TSGCT is usually found in adults between the third and fourth decade of life and is no more prevalent in men than women.[23]

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Table I Search strategy of PubMed and MEDLINE databases

	Total articles
PubMed, January 27, 2019	
1. "Synovitis, Pigmented Villonodular" OR "Giant Cell Tumor of Tendon Sheath" OR "Noonan like syndrome"	1115
2. "Elbow" OR "Elbow Joint"	38,793
3. "1" AND "2"	40
MEDLINE, January 27, 2019	
1. "Synovitis, Pigmented Villonodular" OR "Giant Cell Tumor of Tendon Sheath" OR "Noonan like syndrome"	1648
2. "Elbow" OR "Elbow Joint" OR "elbow.mp"	17,094
3. "1" AND "2"	37
Total from PubMed and MEDLINE	77

Elbow involvement is rare, but prompt recognition and treatment is necessary to preserve upper extremity function. Because of the aggressive and destructive nature of TSGCT, early surgical synovectomy is the mainstay of treatment.[22,26] Available surgical options include either open or arthroscopic surgery at the elbow,[25] but the outcomes have not yet been described in the literature. The current study examines the clinical outcomes and recurrence rate of open and arthroscopic excision of elbow TSGCT.

Methods

Search strategy

The research question and the inclusion and exclusion criteria for individual studies were established before searching databases. Online databases (PubMed and MEDLINE) were used to find literature related to surgical management of TSGCT of the elbow joint. Key words included "Synovitis, Pigmented Villonodular" OR "Giant Cell Tumor of Tendon Sheath" OR "Noonan like syndrome" AND "Elbow" OR "Elbow Joint." Searches were conducted on January 27, 2019, yielding a total of 77 articles from the 2 databases, without applying any restriction on language or date of publication (Table I).

The following inclusion criteria were used: (1) all levels of evidence, (2) male and female patients with no age limitation, (4) studies on humans, (5) involvement of TSGCT of the elbow, and (6) surgical treatment with or without radiotherapy. We excluded any nonsurgical treatment studies (eg, conservative treatment, radiotherapy, targeted therapy, technique articles without outcomes, cadaveric studies, and review articles).

Study selection

The combined results of all searches produced 77 articles. In the first reviewing stage, titles and abstracts were screened, in addition to the titles and abstracts of crossover references. Twenty-four studies satisfied all inclusion and exclusion criteria. An independent reviewer (S.Z.) performed a full-text review of the 24 eligible studies, which presented treatment data for a total of 27 patients with TSGCT of the elbow (Fig. 1).

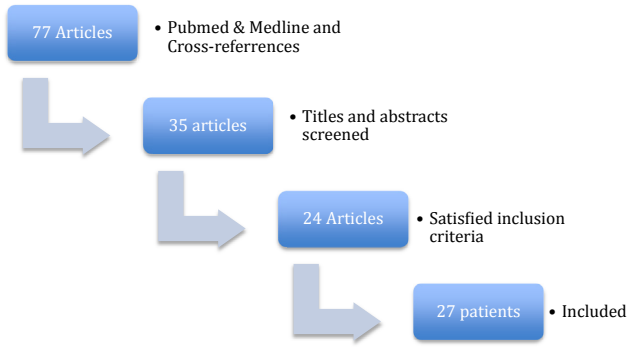

Figure 1 Flow chart illustrating the article screening process.

Data abstraction

One reviewer (H.F.) abstracted relevant study data from the final pool of included studies and recorded them on a spreadsheet. These included study and publication information (author, year of publication, study design, level of evidence, and sample size); patient and disease data (age, sex, disease stage, primary tissue affected, and clinical features); surgical details (procedures performed, technique, and approach); and complications, recurrence, and follow-up.

Results

A total of 27 patients (of 24 studies) were included (Table II). There was 1 case series (which reported on 4 patients) and 23 case reports. In terms of demographics, 16 patients were female (16/27, 59.3%), 10 were male (10/27, 37.0%), and the sex of 1 patient was not reported. The mean (standard deviation [SD]) age of the patients was 40.3 (21.7) years.

The mean (SD) duration of symptoms experienced before consultation was 33.2 (29.3) months. The most common presenting symptoms included pain (18/27, 66.7%), swelling (19/27, 70.4%), and decreased range of motion of the elbow (9/27, 33.3%). On physical examination, most patients presented with a palpable mass (20/27, 74.0%), but tenderness was only reported in 4 cases (4/27, 14.8%).

Before removal, 11 patients (11/27, 40.7%) underwent conformational biopsy of their lesion. Thirteen patients (13/27, 48.1%) had postoperative confirmation by

Table II Studies included for review

Study	Level of evidence	N	Age, yr/sex	Type	Clinical presentation	Management	Follow-up, mo	Recurrence
Torisu et al[39]	IV	1	48 M	Diffuse	P, S, L	Open synovectomy	24	No
Pandey et al[28]	IV	1	7 F	Diffuse	S	Open wide excision	72	No
Deo et al[8]	IV	1	25 M	Localized	S, L	Open wide excision	NR	NR
Pignatti et al[30]	IV	1	54 M	Diffuse	P, L	Open excision	NR	NR
Ekman et al[11]	IV	1	57 M	Diffuse	P, S, L	Arthroscopic synovectomy	72	No
Pimpalnerkar et al[31]	IV	1	56 M	Diffuse	P, S	Open synovectomy	12	No
DiCaprio et al[9]	IV	1	31 F	Diffuse	P, S, L	Open synovectomy	38	Yes
Martin et al[20]	IV	4	48 F	Localized	NR	Open mass excision	72	Yes
			28 M	Localized		Open olecranon bursectomy	92	No
			56 M	Localized		Open olecranon bursectomy	53	No
			48 M	Localized		Open mass excision	25	Yes
Aydingoz et al[2]	IV	1	6 F	Diffuse	S	Open marginal excision	NR	NR
Akkaya et al[1]	IV	1	56 F	Diffuse	P, S	Open subtotal synovectomy	16	No
Geiger et al[12]	IV	1	61 F	Diffuse	P, S	Open synovectomy	NR	NR
Sekiya et al[35]	IV	1	6 F	Diffuse	P, S, L	Open subtotal synovectomy	24	No
Jerome et al[14]	IV	1	16, NR	Diffuse	P	Open synovectomy	48	No
Su et al[38]	IV	1	8 F	Diffuse	P, L	Arthroscopic synovectomy	NR	NR
Wyatt et al[40]	IV	1	55 F	Diffuse	P, S	Open synovectomy	60	No
Chida et al[7]	IV	1	29 F	Diffuse	S	Open synovectomy	30	No
Koca et al[16]	IV	1	41 F	Diffuse	P, S	Arthroscopic synovectomy + radiosynovectomy	20	No
Kohyama et al[17]	IV	1	53 M	Localized	S	Open marginal excision	24	No
Koto et al[18]	IV	1	20 F	Localized	P, S	Open marginal excision	60	No
Mitton et al[21]	IV	1	70 F	Diffuse	P, S	Open wide excision	6	No
Lu et al[19]	IV	1	82 F	Diffuse	P, S	Open wide excision and synovectomy	24	Yes
Ramos et al[32]	IV	1	43 M	Diffuse	P, L	Arthroscopic synovectomy	NR	NR
Savvidou et al[33]	IV	1	68 F	Diffuse	P, S	Open marginal excision	24	No
Caruso et al[5]	IV	1	16 M	Diffuse	P, S, L	Open debridement	NR	NR

M, male; *F*, female; *P*, pain; *S*, swelling; *L*, limited range of motion; *NR*, not reported.

pathology, and 3 patients (3/27, 11.1%) were diagnosed through magnetic resonance imaging (MRI).

Patients underwent either open or arthroscopic excision of the lesion with or without synovectomy. The majority of patients underwent open excision (23/27, 85.1%). Of the patients undergoing open procedures, 16 (16/23, 69.6%) had diffuse TSGCT. The other 7 patients (7/23, 30.4%) had localized TSGCT. Of the patients who underwent an open procedure, 14 (14/23, 60.9%) remained symptom free for a mean (SD) follow-up of 38.9 (25.4) months. Complications of the procedure included recurrence (4/23, 17.4%), slight loss of elbow supination/extension (2/23, 8.7%), and discomfort with crepitus on use of the elbow (1/23, 4.3%). Four patients (4/23, 17.4%) had recurrences within a mean (SD) of 14.0 (9.4) months, 3 of whom underwent re-excision. Two recurrences were detected based on the return of initial presenting symptoms, whereas the presentation of the other 2 recurrences was not described. Five patients did not have follow-up data.

Four patients (4/27, 14.8%) were treated arthroscopically, all of whom had diffuse disease. One arthroscopic patient was also treated with adjuvant radiosynovectomy (rhenium-186), and she remained recurrence free for 20 months. The second arthroscopic patient was found to have synovial chondromatosis at the 6-year follow-up. During the second operation, the lesions found were felt to be dissimilar to those in the initial removal and, thus, was not classified as a case of recurrence. Two patients did not have follow-up data.

Discussion

Etiology

The etiology underlying TSGCT has yet to be clearly elucidated. It is unclear whether it is an inflammatory process, as historically described, or if it could have an underlying neoplastic origin.[37] Findings of chromosomal translocations in patients with TSGCT have led to a recent trend in the literature favoring the theory of a neoplastic

origin.[24,34] However, others argue that although the diffuse form of TSGCT could resemble a neoplastic origin, the localized form is more likely to be a reactive granulomatous form.[29] More research is necessary to elucidate the etiology of this synovial condition, as these findings may also assist in directing treatment options.

Clinical features

The current study found that the most common presenting complaints of TSGCT in the elbow were pain, swelling, and limited range of motion. Among the current cases, the majority of patients had diffuse elbow TSGCT. Three patients had localized disease, all of whom presented with different symptoms, ranging from elbow pain, weakness in the hand, to limited range of motion.

A review by Stephan et al[25] found that diffuse TSGCT has a tendency to present with more concerning symptoms than the localized form. We were unable to confirm this finding when reviewing the cases of patients with elbow TSGCT. This may have been due to the limited sample of patients with localized disease, or the retrospective nature of this study.

Diagnosis

The diagnosis of TSGCT requires a combination of imaging modalities and biopsy.[25] Plain radiographs are often the first-line imaging modality for the presenting signs and symptoms. Soft tissue swelling is a common radiographic finding of elbow TSGCT, but it is not sufficient diagnostically because many other joint abnormalities, such as soft tissue sarcoma, also can have similar signs.[6] In the current study, bone erosion of the radius, ulna, and humerus was commonly found in both diffuse and localized elbow TSGCT.

The most sensitive imaging modality for TSGCT is MRI.[25] Compared with plain radiographs, MRI has superior tissue contrast, which allows visualization of synovial proliferation, joint effusion, bone erosion, and hemosiderin deposits.[6] In both T1- and T2-weighted sequences including gradient echo, hemosiderin deposits are described to be low-signal areas. Although these deposits can also be found in hematoma, pseudoaneurysms, and hemorrhage,[4] the combination of hemosiderin deposits and either villonodular soft tissue masses (localized) or frondlike synovial thickening (diffuse) on MRI is nearly pathognomic of TSGCT.[15,25] However, it is important to note that in practice, localized TSGCT is better defined and thus more straightforward to diagnose than its diffuse counterpart.[15] The mass of diffuse TSGCT is centered in the synovium, which can be mistaken as inflammatory arthritis.[15]

Biopsy remains the criterion standard for diagnosis of TGSCT. Macroscopically, diffuse TSGCT appears as globally thickened, yellow-brown synovium with masses of villi or nodularity.[3] The localized form appears to be less pigmented and only affects a portion of the synovial surface.[3] Microscopically, a mixture of inflammatory mononuclear cells, hemosiderin-laden macrophages, and osteoclast-type multinucleated giant cells is found in both diffuse and localized disease.[25] On pathology, the current study did not find a clear distinction between localized and diffuse TSGCT of the elbow.

Treatment

Although TSGCT is considered a nonmalignant entity, its nature to be locally aggressive and a high potential for recurrence warrant treatment with wide excision.[36] Sufficient removal of the lesion is necessary to reduce the possibility of recurrence.[37] However, extensive open excision at the elbow joint in particular may increase rates of postoperative pain, joint stiffness, and morbidity.[37] To limit these complications, arthroscopic excision has been introduced for management of TSGCT.[25] In comparison to open excision, arthroscopic procedures were found to be effective for localized disease in the knee, hip, shoulder, and ankle.[25] However, for diffuse TSGCT, arthroscopic procedures have been found to be effective only for the knee.[25]

The recurrence rate for open synovectomy in diffuse TSGCT of the knee is between 8.0% and 22.6%.[25] The current study found a recurrence of 17.4% for open synovectomy procedures of the elbow. Two patients had slight loss of elbow supination/extension found on physical examination, but they did not report these findings. One patient reported discomfort and crepitus on use of the elbow. Aside from these minor complications, the open synovectomy procedure was well tolerated.

Because of the limited sample size and the short follow-up, it was not possible to statistically compare the rate of recurrence between the open and arthroscopic procedures. We report that there were no recurrences in the 2 patients who were treated through arthroscopy. However, only 1 of these patients was treated solely with arthroscopy—the second patient received adjuvant radiosynovectomy with rhenium-186.

The efficacy of adjuvant treatment with either external-beam radiation therapy or radiosynovectomy is unclear. Although low recurrence rates have been reported,[27] severe complications, including skin necrosis, joint stiffness, and potential malignant transformation, should not be overlooked.[27] These serious side effects must be discussed with patients before administration of radiation therapy.

Limitations

The current study is not without limitations. Because of the retrospective nature of reviewing case reports and case series, we were unable to ensure consistent methodologies

between authors. As it is a rare entity, the study of TSGCT in the elbow would ideally follow a common protocol for diagnosis, treatment approach, and follow-up. Additional prospectively designed studies will need to be pursued in the future to establish the optimal management of elbow TSGCT.

Conclusion

Over the years, the diagnostic precision of elbow TSGCT has become well established through MRI and tissue biopsy. However, there remains uncertainty regarding the optimal treatment regime for this benign but aggressive tumor. Our review found that open synovectomy appears to be an effective treatment for both localized and diffuse TSGCT in the elbow, and arthroscopic synovectomy is emerging for surgical management of diffuse TSGCT. However, because of the limited number of patients undergoing surgery for TSGCT, further studies are needed to reach a definite conclusion.

Disclaimer

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