Received: 04.11.2013 / Accepted: 20.12.2013
DOI: 10.5137/1019-5149.JTN.9594-13.1

Case Report

Primary Diffuse-Type Tenosynovial Giant Cell Tumor of the Spine: A Report of 3 Cases and Systemic Review of the Literature

Omurganın Primer Difüz Tip Tenosinovyal Dev Hücreli Tümörü: 3 Olgu Bildirimi ve Literatürün Sistematik Gözden Geçirilmesi

Kai WANG[1*], Bin ZHU[1*], Shaomin YANG[2], Zhongjun LIU[1], Miao YU[1], Xiaoguang LIU[1]

[1]*Peking University Third Hospital, Department of Orthopaedics, Beijing, People's Republic of China*
[2]*Peking University Health and Science Center, Department of Pathology, Beijing, People's Republic of China*

[*]*Kai Wang and Bin Zhu contributed equally to this study and shared the first authorship.*

Corresponding Author: Liu XIAOGUANG / E-mail: xiaoguangliu1966@163.com

ABSTRACT

Three patients with spinal primary diffuse-type tenosynovial giant cell tumor (DTGCT) received surgical treatment in our department between 2002 and 2012. All 3 patients were female and aged 23, 33, and 44 years. The mean time from symptom onset to diagnosis was 17 months (range, 5–24 months). One case involved the C1 right lateral mass and C2 vertebral body, the second involved the C1–2 left lateral masses and C2 vertebral body, and the third involved the C5–7 left lateral mass and C6 vertebral body. All patients underwent computed tomography-guided biopsy to confirm the diagnosis of tenosynovial giant cell tumor. Gross total resection was achieved in all patients, including 2 piecemeal resections and 1 en-bloc resection. The mean follow-up time was 6 years (range, 1–11 years), and there was no sign of recurrence in the patients. Seventy cases have been identified so far in the English literature. The male to female ratio is 1:1.38. The mean patient age is 38.5 ± 17.9 years. The tumor distribution includes 32 cases in the cervical spine, 14 in the thoracic spine, 22 in the lumbar spine, and 1 in the sacrococcygeal region. The recurrence rate for patients who underwent gross total resection was 7.7%, and tumor progression was observed in 66.7% of patients who underwent subtotal resection. Above all, DTGCT is a rare primary spinal neoplasm. Preoperative image-guided biopsies play an important role in the diagnosis and treatment strategy. Gross total resection is the best treatment strategy and can reduce the recurrence rate.

KEYWORDS: Tenosynovial giant cell tumor, Diffuse type, Spinal neoplasms, PVNS, Systemic review

ÖZ

Bölümümüzde 2002 ile 2012 arasında spinal primer difüz tip tenosinovyal dev hücreli tümörü (DTGCT) olan üç hastanın cerrahi tedavisi yapıldı. Bu üç hasta 23, 33 ve 44 yaşlarındaydı ve hepsi kadındı. Belirtilerin başlangıcından tanıya kadar ortalama süre 17 aydı (aralık, 5–24 ay). Bir olguda C1 sağ lateral kitle ve C2 vertebral cisim tutulması vardı. İkinci olguda C1–2 sol lateral kitleleri ve C2 vertebra cismi tutulmuştu. Üçüncü olguda C5–7 sol lateral kitleleri ve C6 vertebra cismi tutulmuştu. Tüm hastalarda tenosinov yal dev hücreli tümör tanısını doğrulamak için bilgisayarlı tomografi kılavuzluğunda biyopsi yapıldı. Tüm hastalarda gros total rezeksiyon yapıldı. Hastaların ikisinde parça parça ve 1'inde ise en blok halinde rezeksiyon ile gerçekleştirildi. Ortalama takip süresi 6 yıldı (aralık, 1–11 yıl) ve hastalarda bir nüks bulgusu yoktu. İngilizce literatürde şimdiye kadar 70 olgu tanımlanmıştır. Erkek/kadın oranı 1:1,38'dir. Ortalama hasta yaşı 38,5 ± 17,9 yıldır. Tümör dağılımı servikal omurgada 32 olgu, torasik omurgada 14 olgu, lomber omurgada 22 olgu ve sakrokoksigiyal bölgede 1 olgu şeklindedir. Gros total rezeksiyon yapılan hastalarda nüks oranı %7,7 bulunmuş ve subtotal rezeksiyon yapılan hastaların %66,7'sinde tümör ilerlemesi gözlenmiştir. Genel olarak DTGCT nadir bir primer spinal neoplazmdır. Preoperatif tomografi kılavuzluğunda biyopsiler, tanı ve tedavi stratejisinde önemli bir rol oynamaktadır. En iyi tedavi gros total rezeksiyondur.

ANAHTAR SÖZCÜKLER: Tenosinovyal dev hücreli tümör, Difüz tip, Spinal neoplazmlar, PVNS, Sistematik gözden geçirme

INTRODUCTION

Diffuse-type tenosynovial giant cell tumors (DTGCT) are a type of aggressive benign tumor thought to originate from tendon sheaths, bursae, or diarthrodial joint synovium (5). The etiology of DTGCT remains unknown, and DTGCT was previously named extra-articular PVNS because it shares similar histological characteristics with pigmented villonodular synovitis (PVNS) (39). DTGCT occurs mainly in middle-aged women and usually involves large load-bearing joints such as the knee, ankle, and hip, whereas clinical case reports of DTGCT involving the spine are rare. We retrospectively analyzed a case series of 3 patients diagnosed with spinal DTGCT. The common characteristics, imaging and pathological manifestations, diagnosis and

treatment strategies, and follow-up data from these cases were retrospectively reviewed. Additionally, case reports in the English literature were systematically reviewed.

CASE REPORTS

Case 1

Patient 1 was a 23-year-old woman who experienced neck pain and discomfort over a 2-year period. No limitations in the cervical range of motion (ROM) were noted. The neurological examination was completely normal.

Imaging: The computed tomography (CT) examination revealed a 6×4×6 cm osteolytic lesion on the C1 right lateral mass and the C2 vertebral body (Figure 1A). Magnetic resonance imaging (MRI) demonstrated a lobulated mass that originated in the C1 right lateral mass, with significant paravertebral expansion. Heterogeneous isointensity was identified on the T1-weighted images, and high-signal intensity was identified on the T2-weighted images (Figure 1B).

Treatment: A single-stage combined anterior and posterior piecemeal total resection was performed, and stability reconstruction was achieved via a combination of anterior and posterior internal fixation (nail-plate system and occipitocervical fusion with iliac graft). The intraoperative blood loss was 1500 mL, and the surgery duration was 420 min. After 2.5 years, the posterior internal fixation was removed because of unbearable axial neck pain. The postoperative pathological examination confirmed the diagnosis of DTGCT.

Follow-up examination: The follow-up time was 11 years. The CT images taken at the last follow-up examination showed achievement of solid bone fusion and no sign of recurrence (Figure 1C).

Case 2

The second patient was a 33-year-old woman who experienced neck pain and right hand numbness over a 2-year period. The neurological examination indicated a medial-side sensory loss in the forearm and ring finger.

Imaging: CT images showed a giant osteolytic and expansive bone-destructive lesion centered in the C6 left lateral mass and lamina; the C5 and C7 lateral masses were also involved, and the cortical bone was eroded (Figure 2A). MRI revealed a lobulated mass, with significant expansion to the spinal canal and paravertebral areas. High-signal intensity was identified on the T1-weighted images, and low-signal intensity with patchy high-signal intensity was identified on the T2-weighted images (Figure 2B).

Treatment: A single-stage combined anterior and posterior piecemeal total resection was performed. The C6 vertebral body and lamina and the C5–7 left lateral masses were resected. Stability reconstruction was achieved via a combination of anterior and posterior internal fixation (nail-plate fixation from C3–T2 in the posterior approach and a titanium mesh fusion with iliac bone; Figure 2C). The intraoperative blood loss was 800 mL, and the surgery duration was 300 min.

Follow-up: The follow-up time was 6 years. The patient's right hand and forearm numbness significantly improved. The CT images at last follow-up examination showed no sign of recurrence (Figure 2D).

Case 3

The third patient was a 44-year-old woman who experienced neck pain and discomfort over a 2-year period. Her cervical range of motion (ROM) was significantly limited. The neurological examination was normal.

Imaging: Preoperative CT images showed an osteolytic lesion located at the odontoid, the C2 left vertebral body, and the C1–2 left lateral mass, whereas marginal sclerosis was visible (Figure 3A). MRI revealed a lobulated mass with high-signal intensity on the T1-weighted images and low-signal intensity on the T2-weighted images. Obvious enhancement was observed on the gadolinium diethylenetriamine penta-acetic acid (Gd-DTPA)-enhanced (administered intravenously) T1-weighted images (Figure 3B).

Treatment: A single-stage combined anterior and posterior en-bloc resection was performed. The tumor capsule was intact. Stability reconstruction was achieved via a posterior

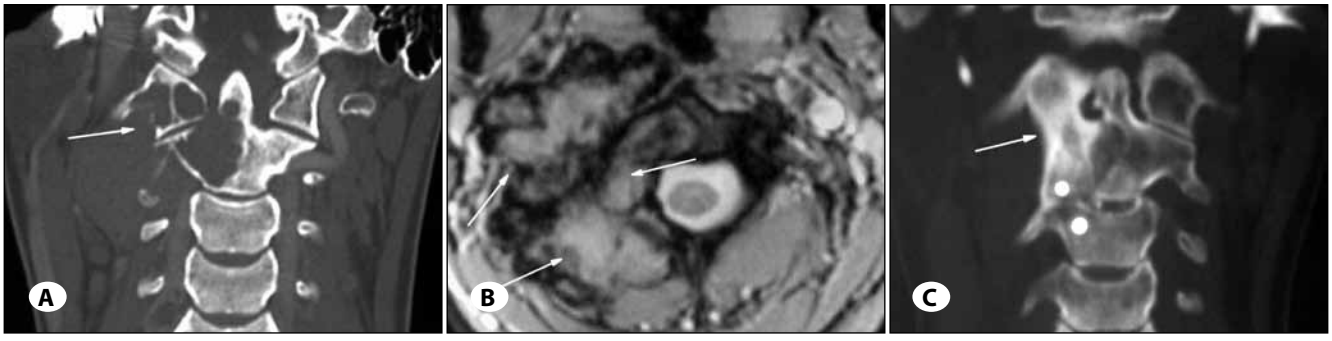

Figure 1: A) Coronal plane computed tomography (CT) image showing an osteolytic lesion of the C1 right lateral mass and the C2 vertebral body (arrow). **B)** Lobulated high-signal intensity mass on T2-weighted magnetic resonance imaging (arrow). **C)** CT image taken 11 years after surgery showed solid bone fusion in the C1–3 region (arrow).

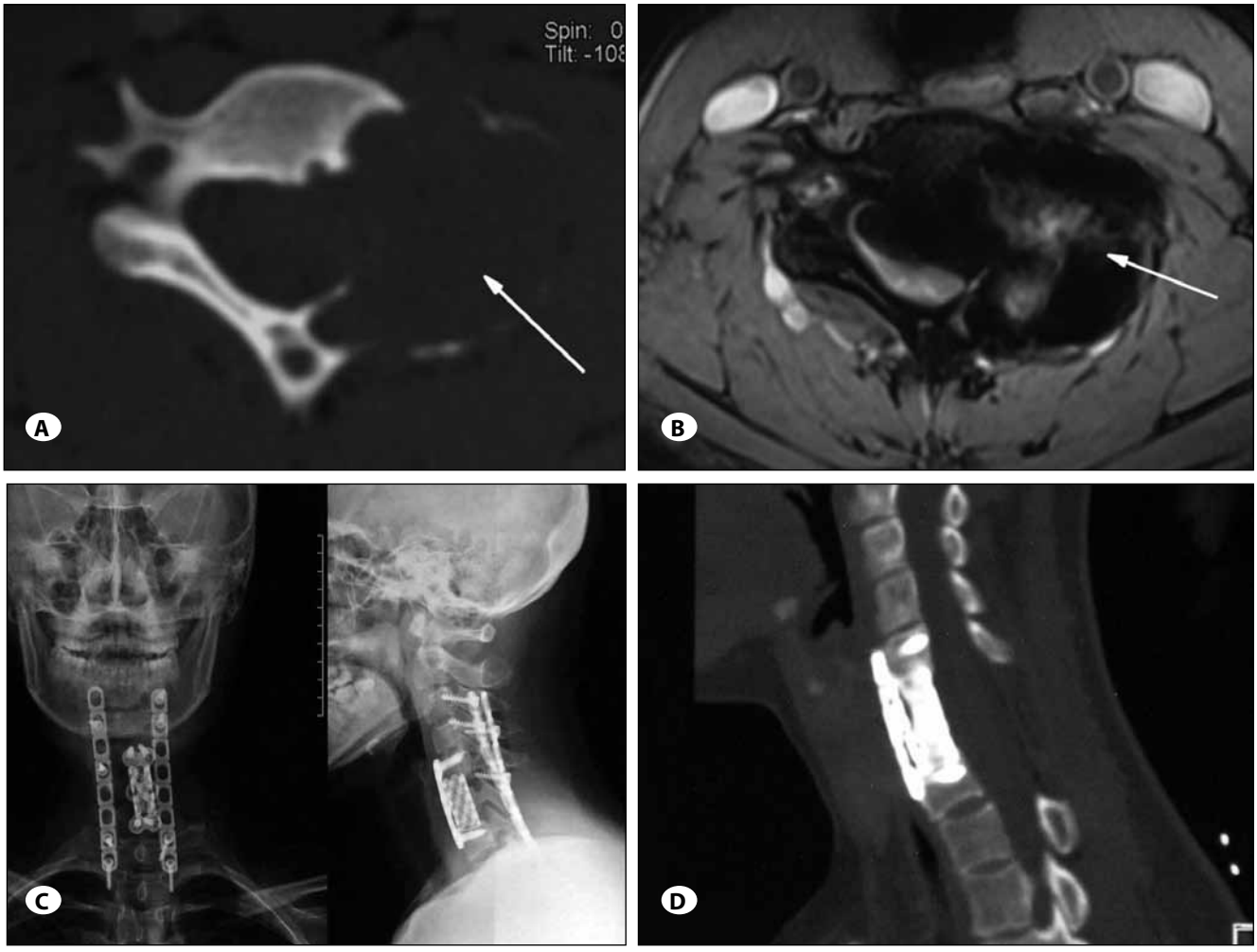

Figure 2: A) Computed tomography (CT) images show C6 left accessory-centric osteolytic and expansive bone destruction (arrow). **B)** Magnetic resonance imaging showed a low-intensity signal with patchy high-density signals on the T2-weighted images; the spinal cord was severely compressed (arrow). **C)** Postoperative antero-posterior and lateral plain graphs showed the internal fixation positioning. **D)** CT image taken 6 years after surgery showed no recurrence.

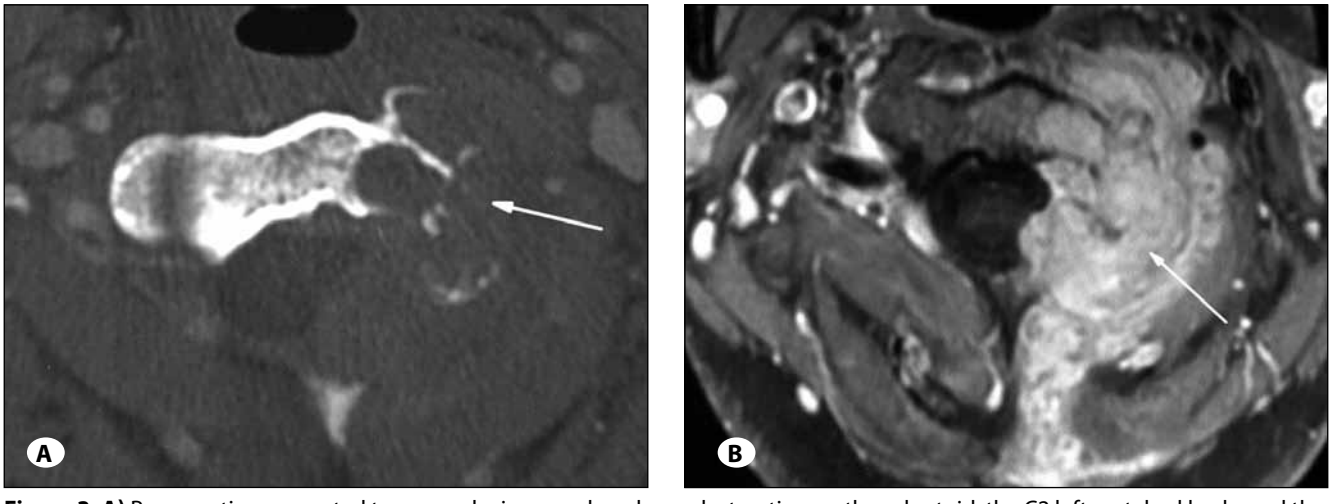

Figure 3: A) Preoperative computed tomography images show bone destruction on the odontoid, the C2 left vertebral body, and the left accessories of C1–2. **B)** A lobulated tumor mass with obvious gadolinium (Gd)-DTPA enhancement was observed on T1-weighted magnetic resonances images with intravenous Gd-DTPA administration.

internal fixation (screw-rod system, occipitocervical fusion with iliac graft). The intraoperative blood loss was 1500 mL, and the surgery duration was 447 min.

Follow-up examination: No recurrence was noted at the 1-year follow-up examination.

Pathological Analysis

Pathologically, all 3 patients were diagnosed with DTGCT, with the following features that differentiated these tumors from other lesions, especially giant cell tumors of the bone. The tumors were non-circumscribed with infiltrative growth into the surrounding tissues. The clefts appeared as synovial-like and pseudoalveolar spaces, and villous patterns could be detected. Mononuclear histiocyte-like cells with vesicular nuclei were the predominant cell type present. Some of the mononuclear cells contained cytoplasmic hemosiderin. Small numbers of osteoclastic giant cells were irregularly distributed throughout the lesions and were more frequently observed around hemorrhagic foci. Sheets of foam cells and scattered lymphocytes were also detected. No necrosis or other obvious features indicative of malignancy were present in the lesions (Figure 4A,B). The immunohistochemical analysis of the 3 patients is listed in Table I.

Table I: Immunohistochemical Analysis of the 3 Patients

	Patient 1	Patient 2	Patient 3
S100	(-)	(-)	(+)
Cytokeratin (mix)	(-)	(-)	(-)
CD45	(-)	(+)	(+)
CD68	(+)	(+)	(+)
Desmin	(-)	(-)	(±)
Vimentin	(+)	(+)	(±)

Systemic review of case reports in the English literature

Search strategy: Two reviewers independently searched the Ovid MEDLINE (1950–present), EMBASE (1980–present), and bibliographies for case reports of DTGCT/PVNS involving the spine. The searches were restricted to English language reports.

The following search terms were used: "diffuse-type tenosynovial giant cell tumor," "DTGCT," "pigmented villonodular synovitis," "PVNS," "giant cell tumor of tendon sheath," "GCTTS," "spinal neoplasms," "spine," and "spine tumor." Any disagreement between the reviewers was resolved by a discussion with the other reviewer. Cases were included if they met the following criteria: a pathological diagnosis of PVNS or DTGCT (giant cell tumor of tendon sheath; GCTTS) and tumors involving the axial skeleton (from atlas to coccyx).

Data extraction: The following information was collected from each case, using a standardized form: author and publication year, tumor location, sex and age, pathological diagnosis, treatment strategy, and follow-up data.

Searching results: After independently reviewing the titles and abstracts identified in the initial search according to the inclusion criteria, 87 cases were included in the full-text screening. Twelve repeatedly reported cases (13, 28), 2 cases of uncertain PVNS diagnoses (43), and 3 non-English reports (19,34,37) were excluded. A total of 70 cases were included in the meta-analysis (Table II) (1,3,4,6~18,20,22,23,25~29, 31~33,35,36,38,40~42,44,46~49).

Meta-Analysis

There were 29 men and 40 women (the gender was unavailable in 1 case) (46), and the male to female ratio was 1:1.38. The mean age of all patients was 38.5 ± 17.9 years. The tumor distribution included 32 cases involving the cervical spine, 14 cases involving the thoracic spine, 22 cases involving the lumbar spine, and 1 case involving the sacrococcygeal region; the location of 1 case was unavailable (13).

Regarding the imaging manifestations, 50 patients (71.4%) reported that the tumors originated from the facet joint, 46 (65.7%) reported spinal canal tumor expansions (epidural or intradural), and 18 (25.7%) reported remarkable paravertebral tumor expansion of the tumor. Regarding the pathological diagnoses, 11 patients were diagnosed with DTGCT/GCTTS, 4 with PVNS/DTGCT, 53 with PVNS, and the other 2 cases with malignant PVNS.

Our systemic review mainly focused on the treatment strategies and prognosis for spinal PVNS/DTGCT. Among the 70 reported cases, imaging data from follow-up examinations performed beyond 1 year were available in 35 cases. Twenty-six patients (26/35, 74.3%) underwent gross total resection (GTR) of the tumors (24 underwent GTR, 1 underwent GTR combined with radiation therapy, and 1 underwent GTR combined with chemotherapy and radiation therapy). No tumor recurrence was observed at the last follow-up examination in 24 cases (24/26, 92.3%), and recurrence was observed in 2 cases (cases 16 and 18; 2/26, 7.7%). Nine patients (9/37, 24.3%) underwent subtotal resection of the tumor, and tumor progression was observed in 6 cases (6/9, 66.7%; includes 2 cases of malignant transformation and metastasis).

DISCUSSION

Tenosynovial giant cell tumors (TGCT) are aggressive soft tissue tumors that belong to a group of lesions that originate from the bursa or the tendon sheath synovium (30). These lesions are classified into 3 types based on their location and encapsulation: nodular tenosynovitis, a localized extra-articular type; PVNS, a diffuse, non-encapsulated intra-articular type; and GCTTS or TGCT, a diffuse, extra-articular growth type. These various tumor types share similar histological findings. TGCT can further be classified as focal-type (FTGCT) or diffuse-type (DTGCT). FTGCT primarily occurs in the fingers and displays clear boundaries, whereas DTGCT occurs in large load-bearing joints such as the knee, hip, ankle, shoulder and elbow and lacks clear boundaries or displays capsular invasion (45); however, DTGCT, PVNS, and GCTTS were intermixed in the literature.

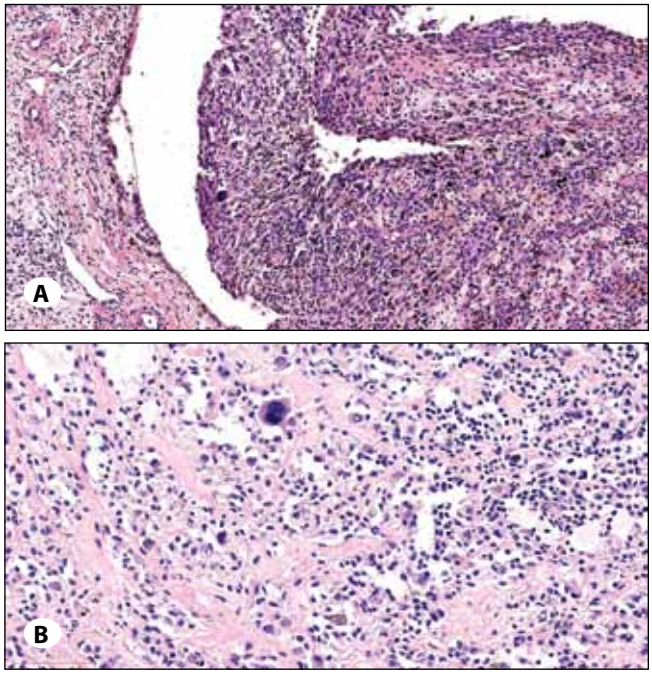

Figure 4: A) The tumors comprise an admixture of mononuclear histiocytoid cells, multinucleate giant cells, foam cells, siderophages, and inflammatory cells. Synovial-like spaces are present (hemotoxylin/eosin staining (HE, x10 magnification). **B)** A villous growth pattern was also detectable (HE, x20).

DTGCT or PVNS involving the spine is extremely rare, and no rigorous reviews were reported in the literature. We report herein 3 cases of spinal DTGCT at our institute and have performed a systematic review of the literature based on the current best evidence. Some scholars believe that primary spinal DTGCT originates from facet joint synovial tissue and mainly grows extra-articularly rather than in the interior of the joint space (11), mostly in the cervical and lumbar vertebrae and rarely in the thoracic spine (13, 15). Our study confirmed these beliefs, as women were more vulnerable to this type of tumor, 71.4% of spinal DTGCT/PVNS cases were reported to originate from facet joints, and 65.7% of these tumors expanded into the spinal canal. Overall, 77.1% of these tumors involved the cervical and lumbar spine.

DTGCT has no specific clinical and radiological manifestations; approximately 50% of patients have focal soft tissue mass shadows, which are occasionally accompanied by manifestations such as bone destruction, periosteal reactions, and calcification within the lesion (21, 28). CT can identify pathological changes of the facet joint, bone destruction, and eroded adjacent vertebral bodies. MRI often shows equal or higher signals than muscle on T1-weighted images, whereas the features on T2-weighted images vary and can be characterized by hypointense, isointense, or hyperintense signals. The lack of specific MRI features can lead to confusion when distinguishing between giant cell tumors of the bone and aneurysmal bone cysts (24).

Pathological manifestations provide the main diagnostic evidence. Histologically, DTGCT is characterized by the presence of monocytes and varying amounts of multinucleated giant cells, foam cells, chronic inflammatory cells, and hemosiderin (13). In the present study, all patients underwent preoperative CT-guided biopsies, followed by histological and immunohistochemical analyses. For all 3 cases, the DTGCT diagnoses were preoperatively confirmed. Immunohistochemically, the biopsy specimens from all 3 cases were positive for vimentin and CD68, suggesting that monocytes comprised the majority of reactive cells in these DTGCT. These findings were consistent with the pathological characteristics of TGCT. Therefore, preoperative CT-guided biopsy is an important method for preoperative diagnoses and treatment strategy decision-making.

GTR was the best treatment strategy, according to our systematic review. The recurrence rate of patients who underwent GTR was only 7.7%, and tumor progression was observed in 66.7% of patients who underwent subtotal resection. Additionally, 2 cases of malignant transformation were observed in patients who underwent subtotal resection. In the present study, all 3 patients received extra-capsular total resections, and no tumor recurrence was observed during the long-term follow-up period. Recently, the intraoperative application of 300-mCi radiotherapy-assisted surgery has been shown to significantly reduce the postoperative recurrence rate of DTGCT of the knee joint (5). Furthermore, some scholars attempted to treat TGCT in the articulations of the appendicular skeleton with imatinib-targeted therapy (2). However, no such treatments were reportedly applied to spinal DTGCT.

CONCLUSION

In summary, primary spinal DTGCT is rarely reported. Diagnosis mainly depends on histological and immunohistochemical analyses. CT-guided biopsy is an important diagnostic method. GTR is the best treatment strategy and can reduce the recurrence rate.

Table II: Common Characteristics of Primary Spinal DTGCT (Including PVNS) Reported in English Literature

Case no	Author	Gender/ Age	Sites	Pathological diagnosis	Treatment strategy	Follow-up
1	Kleinman (1980)	F/65	C3-6,with SCE	PVNS	STR	Symptom-free with 8 months follow-up
2	Campbell (1982)	F/54	L4-L5,right facet joint ,with SCE	PVNS	GTR	Symptom-free with 32 months follow-up
3	Pulitzer (1984)	F/35	C1-4	PVNS	GTR	NTR with 132 months follow-up*
4		M/23	L5-S1,facet joint,with SCE	PVNS	GTR	No follow-up
5	Weidner (1986)	F/48	L4-L5,left facet joint, with PVE and SCE	PVNS	STR	Tumor progress 4 months later, then ER, TR 4 months later, then RR,NTR with 96 months follow-up*
6		F/34	Left facet joint of L2 and L3, left pedicle of L2,with PVE and SCE	PVNS	STR	Symptom-free with 44 months follow-up*
7	Retrum (1987)	F/81	L5-S1,with SCE	PVNS	GTR	No follow-up
8	Karnezis (1990)	F/37	Left facet joint of C6 and C7,lamina of C7	PVNS	GTR	Symptom-free and NTR with 79 months follow-up*
9	Khoury (1991)	F/61	Left facet joint of L4 and L5, with SCE	PVNS	GTR	No follow-up
10		M/84	Left facet joint of C4 and C5, with SCE	PVNS	GTR	Symptom-free with 12 months follow-up
11	Kuwabara (1992)	F/25	Right facet joint of T8 and T9, with SCE and PVE	PVNS	GTR with Chemotherapy and Radiation	NTR with 84 months follow-up*
12	Mahmood (1992)	F/42	Right facet joint and lamina of C6 and C7,with SCE	DTGCT	GTR	No follow-up
13	Titelbaum (1992)	F/51	Right facet joint of L4-5,with SCE	PVNS (DTGCT)	GTR	No follow-up
14	Clark (1993)	M/23	Extraspinal and intraspinal at T7-8 level, laminal arches of T7-8	PVNS	GTR	NTR with 42 months follow-up*
15	Giannini (1996)	F/37	C5 with facet joint involved	PVNS	GTR	NTR with 120 months follow-up
16		F/29	L5-S1 with facet joint involved	PVNS	GTR	TR 12 months later, then RR,NTR with 23 months follow-up
17		F/37	C7 With facet joint involved and SCE	PVNS	GTR	NTR with 66 months follow-up
18		M/38	C4-5 With facet joint involved and SCE	PVNS	GTR	TR, then GTR,NTR with 55 months follow-up
19		M/40	T11	PVNS	GTR	NTR with 64 months follow-up
20		F/21	T3 With facet joint involved	PVNS	GTR	NTR with 53 months follow-up
21		M/43	C5 With facet joint involved and SCE	PVNS	GTR	NTR with 36 months follow-up
22		M/42	L3	PVNS	STR	Symptom-free with 14 months follow-up
23		M/67	L3 With facet joint involved and SCE	PVNS	-	No follow-up
24		M/26	C6 With facet joint involved and SCE	PVNS	GTR	NTR with 11 months follow-up
25		M/44	C5-6 with SCE	PVNS	GTR	NTR with 6 months follow-up
26	Bui-Mansfield (1996)	M/25	Right pedicle, lamina, and facet joint of C4,with SCE and PVE	GCTTS (DTGCT)	-	No follow-up
27	Gezen (1996)	M/19	T9-T12,epidural mass with lamina eroded	PVNS	STR	No tumor progress with 16 months follow-up(MRI)

Table II: Cont.

Case no	Author	Gender/ Age	Sites	Pathological diagnosis	Treatment strategy	Follow-up
28	Clerc (1999)	M/60	Left pedicles and vertebral bodies of L2 and L3,with SCE and PVE	PVNS/ Malignant PVNS	STR	TR and Meta 20 months later, then STR and chemotherapy, die of sepsis 7 months later
29	Bruecks (2000)	F/13	T7–8 posterior elements and pedicle	PVNS	GTR	No follow-up
30	Dimeco (2001)	F/70	Right facet joint of L3 and L4,with SCE	PVNS	GTR	No follow-up
31	Sampathkumar (2001)	M/71	Right facet joint of L5 -S1,with SCE and PVE	PVNS	GTR	NTR with 36 months follow-up
32	Dingle (2002)	F/27	Left facet joint of C4 and C5	GCTTS (DTGCT)	STR	No tumor progress with 24 months follow-up(CT)
33	Graham (2002)	F/44	Lateral mass of C1 and C2,vertebral body of C2,with PVE	PVNS	GTR and Radiation	Symptom-free with 24 months follow-up
34	Furlong (2003) #	M/17	Sacrococcyx	PVNS	GTR	No follow-up
35		F/21	T4-5,facet joint,with SCE	PVNS	GTR	No follow-up
36		M/23	C2-3,facet joint,with SCE	PVNS	STR	Tumor progress with 60 months follow-up, then GTR and radiation, no further follow-up
37		F/25	L4-5,facet joint, with SCE	PVNS	STR	Tumor progress 24 months later, then GTR,TR at last follow-up
38		F/29	L4,facet joint	PVNS	GTR (en-bloc)	NTR with 48 months follow-up
39		M/31	C5-6,facet joint	PVNS	GTR	NTR with 108 months follow-up
40		F/32	C3-4,facet joint, with SCE	PVNS	GTR	NTR with 36 months follow-up
41		M/32	-	PVNS	STR	Tumor progress with 12 months follow-up, then STR and radiation, residual tumor(stable) with 24 months follow-up
42		M/35	Cervical spine	PVNS	GTR and Radiation	NTR with 84 months follow-up
43		F/39	Cervical spine	PVNS	GTR	NTR with 120 months follow-up
44		F/39	C5-6,facet joint, with SCE	PVNS	GTR	No follow-up
45		F/43	C5-6,facet joint, with SCE	PVNS	STR	No follow-up
46		M/44	Cervical spine	PVNS	GTR and Radiation	NTR with 4 months follow-up
47		F/44	C4-5,facet joint, with SCE	PVNS	STR	No follow-up
48	Parmar (2004)	M/43	Posterior elements of C5 and C6,with SCE and PVE	PVNS	GTR	Symptom-free and NTR with 6 months follow-up
49	Motamedi (2005)$	F/14	L5-S1,facet joint, with PVE	PVNS	-	No follow-up
50		F/7	T2-3,facet joint, pedicle, with SCE and PVE	PVNS	-	No follow-up

Table II: Cont.

Case no	Author	Gender/ Age	Sites	Pathological diagnosis	Treatment strategy	Follow-up
51		F/36	T5-6,facet joint, pedicle, with SCE and PVE	PVNS	-	No follow-up
52		M/30	T5-6,with PVE	PVNS	-	No follow-up
53	Doita (2005)	M/26	left facet joint, pedicle and lamina of T7 and T8,with PVE	GCTTS (DTGCT)	STR	Symptom-free and no tumor progress with 24 months follow-up(MRI)
54	Hansen (2007)	M/17	Left facet joint and lamina of C6-7,with SCE	PVNS (DTGCT)	GTR	No follow-up
55	Oda (2007)	F/53	Lateral vertebral body and facet joint of L5 and S1,with PVE and SCE	Malignant PVNS (DTGCT)	STR	Tumor progress 10 months later, then GTR(en-bloc),TR and Meta 7 months later, then chemotherapy, no further follow-up
56	Finn (2007)	F/82	The odontoid process and right lateral mass of C2	PVNS	GTR	NTR with 24 months follow-up(CT)
57	Oe (2007)	M/43	Left facet joint of L4-5 ,with SCE	PVNS	GTR	Symptom-free and NTR with 36 months follow-up(MRI)
58	Gupta (2008)	F/9	T8-9,facet joint, transverse process, with SCE	GCTTS (DTGCT)	GTR	Symptom-free and NTR with 18 months follow-up
59	Blankenbaker (2008)	M/43	C1,Posterior arch, with PVE	TGCT	GTR	No follow-up
60	Rovner (2008)	F/37	Right L5 lamina and L5-S1 facet joint	PVNS	GTR	NTR with 8 months follow-up(CT)
61	Del (2009)	M/17	Left facet joint and lamina of T9,with SCE	PVNS	GTR	NTR with 72 months follow-up(MRI)
62	Musluman (2009)	F/59	Left facet joint of L4-5 ,with SCE	PVNS	GTR	Symptom-free and NTR with 6 months follow-up(CT)
63	Yener (2010)	F/66	Spinous process, right lamina and facet joint of L2,with SCE	PVNS	GTR	No follow-up
64	Hsieh (2012)	M/39	Right facet joint of L2-3,with SCE	PVNS (DTGCT)	GTR	No follow-up
65	Okutan (2012)	M/65	Lamina and spinous process of C7,with SCE	DTGCT	GTR	NTR with 6 months follow-up(CT)
66	Teixeira (2012)	?/31	Right intervertebral foramina between C1 and C2,with SCE and PVE	GCTTS (DTGCT)	GTR (en-bloc)	NTR with 18 months follow-up(MRI)
67	Siribumrungwong (2013)	F/7	left facet joint at T5-T6,with SCE	PVNS (DTGCT)	GTR	NTR with 12 months follow-up(MRI)
68	Our cases	F/23	Right lateral mass of C1 and the vertebral body of C2,with PVE	DTGCT	GTR	NTR with 132 months follow-up(CT)
69		F/33	Left lateral mass of C5-7 and lamina of C6,with SCE and PVE	DTGCT	GTR	NTR with 72 months follow-up(CT)
70		F/44	Left vertebral body of C2 and the left lateral mass of C1-2	DTGCT	GTR (en-bloc)	NTR with 12 months follow-up(CT)

M: *Male;* ***F:*** *Female;* ***DTGCT:*** *Diffuse-type tenosynovial giant cell tumor;* ***PVNS:*** *Pigmented villonodular synovitis;* ***GCTTS:*** *Giant cell tumor of tendon sheath;* ***SCE:*** *Spinal canal expansion;* ***PVE:*** *Paravertebral expansion;* ***GTR:*** *Gross total resection;* ***STR:*** *Subtotal resection;* ***TR:*** *Tumor recurrence;* ***NTR:*** *No tumor recurrence;* ***Meta:*** *Metastasis;* ****:*** *Data updated by Giannini.et.al;* ***#:*** *1 case previously reported by Bui-Mansfield.et.al. (case 26) was excluded;* ***$:****11 cases previously reported by Furlong.et.al were excluded.*

REFERENCES

1. Blankenbaker DG, Tuite MJ, Koplin SA, Salamat MS, Hafez R: Tenosynovial giant cell tumor of the posterior arch of C1. Skeletal Radiol 37(7): 667-671, 2008
2. Blay JY, El SH, Thiesse P, Garret J, Ray-Coquard I: Complete response to imatinib in relapsing pigmented villonodular synovitis/tenosynovial giant cell tumor (PVNS/TGCT). Ann Oncol 19(4): 821-822, 2008
3. Bruecks AK, Macaulay RJ, Tong KA, Goplen G: November 2000: 13 year old girl with back pain and leg weakness. Brain Pathol. 11(2): 263-264, 2001
4. Bui-Mansfield LT, Youngberg RA, Coughlin W, Chooljian D: MRI of giant cell tumor of the tendon sheath in the cervical spine. J Comput Assist Tomogr 20(1): 113-115, 1996
5. Campbell AJ, Wells IP: Pigmented villonodular synovitis of a lumbar vertebral facet joint. J Bone Joint Surg Am 64(1): 145-146, 1982
6. Clark LJ, McCormick PW, Domenico DR, Savory L: Pigmented villonodular synovitis of the spine. Case report. J Neurosurg 79(3): 456-459, 1993
7. Clerc D, Berge E, Benichou O, Paule B, Quillard J, Bisson M: An unusual case of pigmented villonodular synovitis of the spine: Benign aggressive and/or malignant. Rheumatology (Oxford). 38(5): 476-477, 1999
8. del CBL, Rosales OLM, Arriaga NM, Izaguirre A, Pineda C: Pigmented villonodular synovitis of thoracic facet joint presenting as rapidly progressive paraplegia. J Clin Rheumatol 15(8): 393-395, 2009
9. Dimeco F, Rizzo P, Li KW, Ciceri E, Casali C, Pollo B, Lasio G: Pigment villonodular synovitis of the spine. Case report and review of the literature. J Neurosurg Sci 45(4): 216-219; discussion 219, 2001
10. Dingle SR, Flynn JC, Flynn JC Jr, Stewart G: Giant-cell tumor of the tendon sheath involving the cervical spine. A case report. J Bone Joint Surg Am 84-A(9): 1664-1667, 2002
11. Doita M, Miyamoto H, Nishida K, Nabeshima Y, Yoshiya S, Kurosaka M: Giant-cell tumor of the tendon sheath involving the thoracic spine. J Spinal Disord Tech 18(5): 445-448, 2005
12. Finn MA, McCall TD, Schmidt MH: Pigmented villonodular synovitis associated with pathological fracture of the odontoid and atlantoaxial instability. Case report and review of the literature. J Neurosurg Spine 7(2): 248-253, 2007
13. Furlong MA, Motamedi K, Laskin WB, Vinh TN, Murphey M, Sweet DE, Fetsch JF: Synovial-type giant cell tumors of the vertebral column: A clinicopathologic study of 15 cases, with a review of the literature and discussion of the differential diagnosis. Hum Pathol 34(7): 670-679, 2003
14. Gezen F, Akay KM, Aksu AY, Beduk A, Seber N: Spinal pigmented villonodular synovitis: A case report. Spine (Phila Pa 1976) 21(5): 642-645, 1996
15. Giannini C, Scheithauer BW, Wenger DE, Unni KK: Pigmented villonodular synovitis of the spine: A clinical, radiological, and morphological study of 12 cases. J Neurosurg 84(4): 592-597, 1996
16. Graham EJ, Kuklo TR, Kyriakos M, Rubin DA, Riew KD: Invasive pigmented villonodular synovitis of the atlantoaxial joint: A case report. J Bone Joint Surg Am 84-A(10): 1856-1860, 2002
17. Gupta R, Jambhekar N, Sanghvi D: Giant-cell tumour of the synovium in a facet joint in the thoracic spine of a child. J Bone Joint Surg Br 90(2): 236-239, 2008
18. Hansen MA, Harper C, Yiannikas C, McGee-Collett M: A rare presentation of pigmented villonodular synovitis. J Clin Neurosci 14(4): 386-388, 2007
19. Hermann J, Stadlmaier E, Ch A, Spuller E, Reittner P, Graninger W: Erosive intervertebral joint lesions. A case of pigmented villonodular synovitis. Z Rheumatol 66(2): 152, 154-156, 2007
20. Hsieh YC, Chen WY, Hsieh TY, Chan WP: Pigmented villonodular synovitis of the lumbar spine. J Clin Rheumatol 18(5): 274-275, 2012
21. Karasick D, Karasick S: Giant cell tumor of tendon sheath: Spectrum of radiologic findings. Skeletal Radiol 21(4): 219-224, 1992
22. Karnezis TA, McMillan RD, Ciric I: Pigmented villonodular synovitis in a vertebra. A case report. J Bone Joint Surg Am 72(6): 927-930, 1990
23. Khoury GM, Shimkin PM, Kleinman GM, Mastroianni PP, Nijensohn DE: Computed tomography and magnetic resonance imaging findings of pigmented villonodular synovitis of the spine. Spine (Phila Pa 1976) 16(10):1236-1237, 1991
24. Kitagawa Y, Ito H, Amano Y, Sawaizumi T, Takeuchi T: MR imaging for preoperative diagnosis and assessment of local tumor extent on localized giant cell tumor of tendon sheath. Skeletal Radiol 32(11): 633-638, 2003
25. Kleinman GM, Dagi TF, Poletti CE: Villonodular synovitis in the spinal canal: Case report. J Neurosurg 52(6): 846-848, 1980
26. Kuwabara H, Uda H, Nakashima H: Pigmented villonodular synovitis (giant cell tumor of the synovium) occurring in the vertebral column. Report of a case. Acta Pathol Jpn 42(1): 69-74, 1992
27. Mahmood A, Caccamo DV, Morgan JK: Tenosynovial giant-cell tumor of the cervical spine. Case report. J Neurosurg 77(6): 952-955, 1992
28. Motamedi K, Murphey MD, Fetsch JF, Furlong MA, Vinh TN, Laskin WB, Sweet DE: Villonodular synovitis (PVNS) of the spine. Skeletal Radiol 34(4):185-195, 2005
29. Musluman AM, Cavusoglu H, Yilmaz A, Dalkilic T, Tanik C, Aydin Y: Pigmented villonodular synovitis of a lumbar intervertebral facet joint. Spine J 9(8): e6-9, 2009
30. Myers BW, Masi AT: Pigmented villonodular synovitis and tenosynovitis: A clinical epidemiologic study of 166 cases and literature review. Medicine (Baltimore) 59(3):223-238, 1980
31. Oda Y, Takahira T, Yokoyama R, Tsuneyoshi M: Diffuse-type giant cell tumor/pigmented villonodular synovitis arising in the sacrum: Malignant form. Pathol Int 57(9): 627-631, 2007
32. Oe K, Sasai K, Yoshida Y, Ohnari H, Iida H, Sakaida N, Uemura Y: Pigmented villonodular synovitis originating from the lumbar facet joint: A case report. Eur Spine J 16 Suppl 3: 301-305, 2007

33. Okutan O, Solaroglu I, Ozen O, Saygili B, Beskonakli E: Tenosynovial giant cell tumor in the cervico-thoracic junction. Turk Neurosurg 22(6): 769-771, 2012
34. Olivier E, Defives T: Spinal locations of pigmented villonodular synovitis: Case report and review of the literature. Rev Chir Orthop Reparatrice Appar Mot 89(6): 549-557, 2003
35. Ottaviani S, Ayral X, Dougados M, Gossec L: Pigmented villonodular synovitis: A retrospective single-center study of 122 cases and review of the literature. Semin Arthritis Rheum 40(6): 539-546, 2011
36. Parmar HA, Sitoh YY, Tan KK, Teo J, Ibet SM, Hui F: MR imaging features of pigmented villonodular synovitis of the cervical spine. AJNR Am J Neuroradiol 25(1): 146-149, 2004
37. Pechlivanis I, Tannapfel A, Tuttenberg J, Harders A, Schmieder K: Pigmented villonodular synovitis involving the thoracic spine: A case report. Z Orthop Unfall 147(2): 220-224, 2009
38. Pulitzer DR, Reed RJ: Localized pigmented villonodular synovitis of the vertebral column. Arch Pathol Lab Med 108(3): 228-230, 1984
39. Ravi V, Wang WL, Lewis VO: Treatment of tenosynovial giant cell tumor and pigmented villonodular synovitis. Curr Opin Oncol 23(4): 361-366, 2011
40. Retrum ER, Schmidlin TM, Taylor WK, Pepe RG: CT myelography of extradural pigmented villonodular synovitis. AJNR Am J Neuroradiol 8(4): 727-729, 1987
41. Rovner J, Yaghoobian A, Gott M, Tindel N: Pigmented villonodular synovitis of the zygoapophyseal joint: A case report. Spine (Phila Pa 1976) 33(18): E656-658, 2008
42. Sampathkumar K, Rajasekhar C, Robson MJ: Pigmented villonodular synovitis of lumbar facet joint: A rare cause of nerve root entrapment. Spine (Phila Pa 1976) 26(10): E213-215, 2001
43. Savitz MH, Katz SS, Goldstein H, Worcester D: Hypertrophic synovitis of the lumbar facet joint in two cases of herniated intervertebral disc. Mt Sinai J Med 49(5): 434-437, 1982
44. Siribumrungwong K, Tangtrakulwanich B, Nitiruangjaras A: Unusual presentation of giant cell tumor originating from a facet joint of the thoracic spine in a child: A case report and review of the literature. J Med Case Rep 7(1): 178, 2013
45. Somerhausen NS, Fletcher CD: Diffuse-type giant cell tumor: Clinicopathologic and immunohistochemical analysis of 50 cases with extraarticular disease. Am J Surg Pathol 24(4): 479-492, 2000
46. Teixeira WG, Lara NA Jr, Narazaki DK, de Oliveira C, Cavalcanti C, Marins LV, Cristante AF, Teixeira MJ, de Barros Filho TE: Giant-cell tumor of the tendon sheath in the upper cervical spine. J Clin Oncol 30(26): e250-253, 2012
47. Titelbaum DS, Rhodes CH, Brooks JS, Goldberg HI: Pigmented villonodular synovitis of a lumbar facet joint. AJNR Am J Neuroradiol 13(1): 164-166, 1992
48. Weidner N, Challa VR, Bonsib SM, Davis CH Jr, Carrol TJ Jr: Giant cell tumors of synovium (Pigmented villonodular synovitis) involving the vertebral column. Cancer 57(10): 2030-2036, 1986
49. Yener U, Konya D, Bozkurt S, Ozgen S: Pigmented villonodular synovitis of the spine: Report of a lumbar case. Turk Neurosurg 20(2): 251-256, 2010