Recurrent Pigmented Villonodular Synovitis and Multifocal Giant Cell Tumor of the Tendon Sheath: Case Report

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Intra- and extra-articular giant cell tumor of tendon sheath (GCTTS) and pigmented villonodular synovitis (PVNS) are histologically similar, usually benign tumors that can be characterized by synovial involvement (GCTTS) or overgrowth (PVNS). These tumors are most often found in the knee and digits of the hand. Although recurrence is a common feature of both conditions, multifocal lesions are rare. We present an unusual case of multifocal, recurrent, bilateral GCTTS/PVNS involving both upper and lower extremities. Recurrent right ankle and right index finger masses, in addition to masses on the right small finger and left thumb, were excised over a 14-year period. *(J Hand Surg Am. 2015;40(3):537–541.*

Key words Giant cell tumor of tendon sheath, pigmented villonodular synovitis, multifocal, recurrent.

Giant cell tumor of the tendon sheath (GCTTS) and pigmented villonodular synovitis (PVNS) are benign, locally invasive tumors involving synovium, tendon sheaths, and bursae. Patients may present with a discrete mass or with joint swelling, pain, or locking or catching.[1] The knee and digits of the hand are the most common locations of lesions (Table 1)[2–20] and the tumor is characterized by the site of involvement and its cell types.[1]

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Received for publication September 16, 2014; accepted in revised form November 7, 2014.

No benefits in any form have been received or will be received related directly or indirectly to the subject of this article.

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0363-5023/15/4003-0020$36.00/0
http://dx.doi.org/10.1016/j.jhsa.2014.11.010

Traditionally, PVNS has been a catchall term for tumors with synovial or synovial cell–like involvement. The associated terminology and treatment options for the spectrum of lesions associated with these conditions have evolved since Jaffe et al's landmark report.[21] The World Health Organization has separated PVNS from GCTTS by describing them as 2 distinct types of giant cell tumor.[22] This distinction between PVNS and GCTTS is based on tumor behavior.[22] PVNS, also termed *diffuse-type PVNS* or *diffuse-type giant cell tumor*, in this new taxonomy is defined as "destructive proliferation of synovial-like mononuclear cells, mixed with multinucleated giant cells, foam cells, siderophages and inflammatory cells."[22] PVNS is further typified by its diffuse nature, demonstrated by infiltration throughout the affected area beyond the joint and into the surrounding tissues.[22] GCTTS, "a family of lesions arising from the synovium of joints, bursae and tendon sheaths," is distinguished from PVNS by its localized nature.[22] GCTTS is further subdivided by tumor location, being either intra-articular (previously termed *localized PVNS*) or extra-articular (previously termed *nodular tenosynovitis*).[3] Extra-articular GCTTS is commonly seen in the tendon sheaths of the hand.[11]

TABLE 1. Multiple GCTTS/PVNS Studies or Case Report: Locations of Occurrence, Adult or Child Designations, and Time Intervals Between Recurrence of GCTTS/PVNS

Author/Year	GCTTS/ PVNS	Adult/ Child	Large Joints	Digits	Other Location	Time Interval Between Recurrence
Greenfield et al 1950[2]	PVNS	Adult	Bil knees			0
Phalen et al 1959[3]	GCTTS	Adult		Different hand		NA
Byers et al 1968[4]	PVNS	Adult	Bil ankles			0
Gehweiler et al 1969[5]	PVNS	Adult	Bil knees			5 y
Leszczynski et al 1975[6]	PVNS	Child	Knee, bil ankles			NA
Crosby et al 1977[7]	PVNS	Adult		Different hand		0
Eisenberg et al 1978[8]	PVNS	Adult	Bil hips			10 mo
Raja et al 1981[9]	PVNS	Adult	Knee, ankle			17 mo
Walls et al 1985[10]	PVNS	Child	Knee		Bil posterior tendons joined at the medial knee	11 mo
Ushijima et al 1986[11]	GCTTS	Adult		Ring finger	Great toe	NA
Jamieson et al 1990[12]	PVNS	Adult	Bil wrists			0
Kay et al 1996[13]	PVNS	Child	Wrist		pes anserine tendon	10 mo
Hitora et al 2002[14]	GCTTS	Adult		Same tendon		4 y
Dalal et al 2003[15]	GCTTS	Adult		Bil thumbs		2 y
Tavangar et al 2005[16]	PVNS	Child	Bil knees, elbow			NA
Park et al 2006[17]	GCTTS	Adult		Same tendon		7 mo
Altaykan et al 2009[18]	GCTTS	Adult		Same tendon		3 y
Singh et al 2011[19]	GCTTS	Adult		Same tendon		0
Zeinstra et al 2013[20]	GCTTS	Adult		Same tendon		0

NA, not available; bil, bilateral.

The vast majority of reported synovial tumor cases involve unifocal lesions. Multifocal lesions are rare and have been described most often in children.[16] We report a case of histologically confirmed, multifocal, bilateral, and recurrent extra- and intra-articular giant cell tumor of the tendon sheath in an adult involving both the upper and lower extremities.

CASE REPORT

A 36-year-old woman initially presented at another institution with a soft tissue mass near her right ankle. An operative report described an extensive multilobulated soft tissue mass arising from and surrounding the sheath of the posterior tibial tendon. The pathology report from an excisional biopsy indicated PVNS. Two years later, the patient underwent surgery at the same institution for recurrent swelling at the prior excision site. The operative report described extensive friable tissue surrounding the posterior tibial tendon proximally, extending throughout the tendon sheath, and distal to the tendon. In addition, the tumor had infiltrated the flexor digitorum longus and flexor hallucis longus tendons and was found extensively around the proximal portion of the metatarsals and dorsal to the talonavicular joint extra-articularly. Because of the extensive area of involvement and a histologic similarity to the first tumor, this recurrence was diagnosed as PVNS. Adjuvant treatment was not administered and the mass did not recur.

Six years later, the patient presented to a second institution for evaluation of masses on her right index and small fingers. Intraoperative examination revealed a solid and well-defined mass localized over the volar neck of the middle phalanx of the index finger and a poorly defined mass dorsal to the small finger distal interphalangeal joint. Bony erosion of the index middle phalanx was observed at surgery without articular extension. After excisional biopsy,