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Clinical and oncological outcomes after surgical excision of pigmented villonodular synovitis at the foot and ankle

M.T. Korim B.Sc, MBBS, MRCS, FRCS(Tr&Orth)[a,*], D.R. Clarke B.Sc, MBChB, MBBS, MRCS[a], P.E. Allen MBChB, FRCS (Orth)[a], C.J. Richards BM BS, FRCPath[b,c], R.U. Ashford MBBS, MD, FRCS(Tr & Orth)[a,b,d]

[a] Leicester Orthopaedics, University Hospitals of Leicester, Leicester General Hospital, Gwendolen Road, Leicester, UK
[b] East Midlands Sarcoma Service, UK
[c] Histopathology, University Hospitals of Leicester, Leicester Royal Infirmary, Infirmary Square, Leicester, UK
[d] Academic Orthopaedics, Trauma & Sports Medicine, University of Nottingham, Queens Medical Centre, Nottingham, UK

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ABSTRACT

Background: Pigmented villonodular synovitis (PVNS) is a rare benign neoplastic disease of the synovium of joints and tendon sheaths, which may be locally aggressive. It can be broadly classified into localised disease or more diffuse forms, with the latter more prone to recurrence after surgical excision. We describe our experience in the management of foot and ankle PVNS, focusing on the diffuse type.
Methods: Patients with PVNS were identified from a histology database from 2000 to 2010 at the University Hospitals of Leicester. The primary aim was to determine oncological outcomes and evaluate clinical outcomes with the Toronto Extremity Salvage Score (TESS) and the American Academy of Foot and Ankle Surgeons (AOFAS) scores.
Results: 30 patients, 16 males and 14 females with a mean age of 37 ± 15 years, who underwent surgery, were identified. There were 22 nodular PVNS and 8 diffuse PVNS. The diffuse PVNS was more likely to be in the hindfoot (75%, 6/8), of which 50% (3/6) had osteoarthritis at presentation. The localised PVNS was mostly located in the forefoot (91%, 20/22). None of the localised PVNS had a recurrence. The surgical recurrence rate in this series was similar to the pooled recurrence rate from the literature [12.5% (1/8) compared to 12.2% (6/49)]. The mean TESS and AOFAS scores were 86 and 78, respectively.
Conclusions: Diffuse PVNS is more likely to occur in the hindfoot and nodular PVNS is more common in the forefoot. Aggressive synovectomy alone is an effective treatment for diffuse PVNS, with good oncological and clinical outcomes.

1. Background

Giant cell tumours (GCT) of the tendon sheaths and synovium can be classified into two forms: localised (giant cell tumour of the tendon sheath, or nodular tenosynovitis) and diffuse (diffuse-type giant cell tumour or pigmented villonodular synovitis). The World Health Organisation classifies diffuse PVNS as 'diffuse-type giant cell tumour' (Dt-GCT) ICD-O code 9251/0, differentiating it from localised PVNS and Giant cell-rich tumours of the tendon sheath (GCTTS), the latter considered to be the extra-articular form of localised PVNS [1,2]. In this paper Dt-GCT will be used to describe diffuse PVNS and localised PVNS will be used for nodular PVNS and GCTTS.

Dt-GCT was first identified in 1852 as a neoplastic condition, though the term PVNS was first coined by Jaffe et al. [3]. In 1976 it was subdivided into either diffuse or localised by Granowitz and Mankin [4]. Both of these forms are similar histologically [5,6]. Localised PVNS is characterised by focal involvement of the synovium, with nodular or pedunculated masses. Dt-GCT is characterised by involvement of most or all of the joint synovium leading to eventual osseous erosions and subchondral cysts. Patients present with insidious swelling and/or pain which has been present for months or years. Swelling and pain are more pronounced than in localised PVNS and usually are poorly localised. Dt-GCT tends to have a more rapidly destructive course and, as a result, a poorer prognosis [2,5,6]. Dt-GCT has a higher

* Corresponding author at: 33 West Avenue, Leicester LE2 1TS, UK. Tel.: +44 07792746502; fax: +44 01162706001.
E-mail addresses: mtkorim@doctors.org.uk, mtkorim@blueyonder.co.uk (M.T. Korim).

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recurrence rate after synovectomy [1,2,5,7]. External beam radiotherapy and intra-articular instillation of Yttrium-90 have been described as an adjunct to reduce recurrence, but are not widely used because of worries regarding treating a "benign" condition with a treatment that can potentially result in a post-treatment malignancy [7–11].

There are only a few small case series of foot and ankle Dt-GCT treated with surgery alone and functional outcomes after treatment are not well established. We describe our experience on the management of foot and ankle PVNS in Leicester from 2000 to 2010, concentrating on the Dt-GCT. The primary aim was to determine oncological outcome and evaluate clinical outcomes with validated foot scores.

2. Methods

Patients with confirmed PVNS (Dt-GCT and localised PVNS) were identified from the histology database at the University Hospitals of Leicester from 2000 to 2010. A case note and radiological review was then undertaken.

The diagnosis of PVNS was made from the clinical presentation, MRI findings and typical histological findings as assessed by a specialist sarcoma pathologist. The case notes were reviewed for the intra-operative findings, especially for extension into neighbouring structures and completeness of excision. All the available imaging (MRI scans, CT scans and plain radiographs) were reviewed for the location of the PVNS, presence of osteoarthritis as well as involvement of neighbouring tendon sheaths.

All the patients underwent open synovectomy performed by a consultant orthopaedic surgeon. The MRI scan was used to plan the surgical approach in all the Dt-GCT as well as guide the surgeon to the abnormal synovium. No radiotherapy was utilised for any patient in this series. The clinical, radiological and intra-operative findings are illustrated in Table 1.The patients with Dt-GCT were followed at 3 months and then yearly afterwards for 5 years before discharge. Post-operative MRI was arranged in cases of suspected recurrence. The patients with localised PVNS were discharged at the first follow up visit if they were no signs of recurrence.

The outcome of surgery was assessed using recurrence rates and validated foot outcome scores. The Toronto Extremity Salvage Score (TESS) [12] and the American Academy of Foot and Ankle Surgeon (AOFAS) [13] scores were used to assess functional outcomes following surgery. The TESS is a 30 item questionnaire and assesses routine daily activities such as dressing, work and mobilisation. The score is expressed as a percentage and maximum score is 100 [12]. The AOFAS is a validated tool to assess outcomes after foot and ankle surgery and is also scored out of 100. The average score for the normal population is 50 [13].

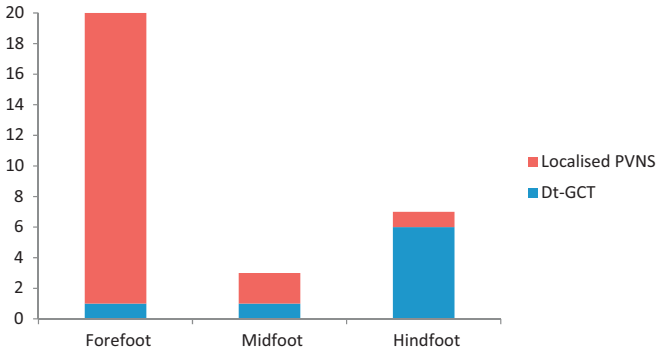

Fig. 1. Type and location of the PVNS in the foot and ankle.

3. Results

There were 30 patients, 16 males and 14 females, with a mean age of 37 ± 15 years at presentation and an average follow up of 4 years (range 2–10 years). Eight were Dt-GCT and 22 localised PVNS. The location of all the PVNS is summarised in Fig. 1.

The Dt-GCT was more likely to occur in the hindfoot; 75% (6/8) of the Dt-GCT occurred in the hindfoot, 12.5% (1/8) in the midfoot and 12.5% (1/8) was in the forefoot. The neighbouring tendon sheaths were involved in 37.5% (3/8) cases. 91% (19/22) of the nodular PVNS occurred in the forefoot, except two involving the midfoot joints and one over the ankle joint

3.1. Clinical outcomes

The mean TESS and AOFAS scores were 85 and 78, respectively, and these scores are illustrated in Fig. 2. This is consistent with good functional outcomes based on 50 being the normative AOFAS score. There is no accepted normal score for the TESS.

3.2. Complications and recurrence

There were no recurrences in the localised PVNS group. The surgical recurrence rate in the Dt-GCT group was 12.5% (1/8) and a further patient had a small re-growth that is regressing in size radiologically, with further treatment unlikely. The former was from the 5th toe IP joint, MTP joint and metatarsal. At the time of primary surgery, the disease was found to be involving both little toe neurovascular bundles from the IP joint up to and including the MTP joint. There was also involvement of the flexor and extensor tendons. An attempt was made to salvage the toe by preserving the medial neurovascular bundle and performing a marginal excision

Table 1
Clinical and radiological parameters of patients with Dt-GCT.

Age/Gender	Follow up (years)	Joint(s) involved	MRI findings and structures involved	Surgical approach & resection	Recurrence
55F	4	Ankle and subtalar joint with talar tilt and OA	Posterolateral disease Peroneal tendons	Anteromedial (complete)	No-Arthrodesis as primary treatment
51F	10	Ankle with OA	Anterolateral disease	Anterolateral (complete)	No
18F	4.5	Ankle, No OA	Posterolateral disease	Posterolateral (complete)	11 mm lesion regressing in size on MRI
26M	9	Intercuneiform joints and naviculocuneiform joint, No OA	Capsule of joints only	Dorsal longitudinal (complete)	No
43F	5	Ankle with OA	Anterior disease	Anterior (complete)	No
57M	4	5th toe IPJ, MTPJ and metatarsal	Extensive disease around the 5th ray, Flexor and extensor tendons	Dorsal longitudinal (subtotal resection preserving NVB)	Recurrence-further surgery
21F	4	Ankle joint, No OA	Anterolateral disease (recurrent)	Anterolateral (complete)	No
34M	2	Ankle, No OA	Anterolateral disease (recurrent)	Anterior (complete)	No