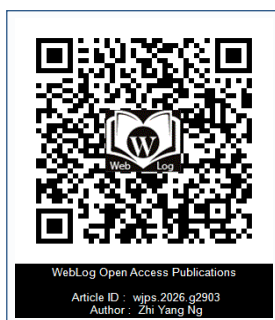




Giant Cell Tumour of the Tendon Sheath in the Distal Phalanx of the Third Toe

Kin Seng Tong, Xavier Chalhoub and Zhi Yang Ng*

Department of Plastic Surgery, Queen Alexandra Hospital, Portsmouth University Hospitals NHS Trust, UK



Abstract

Giant cell tumours of the tendon sheath (GCTTS) involving the distal phalanx of the foot are exceedingly rare, with only a few cases reported. We report a case of GCTTS involving the distal phalanx of the middle toe in a 36-year-old female, who presented with a three-year history of an enlarging mass in her right foot with discomfort in shoe wear. While GCTTS was suspected clinically, ultrasonographic evaluation suggested an epidermoid cyst instead. Histological diagnosis of GCTTS was confirmed following surgical excision. Successful toe preservation was achieved and there was no evidence of recurrence at 10 months' follow-up. This case demonstrates the importance of maintaining a high index of clinical suspicion for GCTTS when faced with an enlarging and relatively asymptomatic mass in the toe. A history of trauma is not always obvious. Imaging oftentimes remain equivocal and surgical excision is preferred as it achieves both diagnosis and treatment.

Introduction

In 2013, the World Health Organization introduced an umbrella term tenosynovial giant-cell tumour (TGCT) for soft tissue tumours that can be further divided into localised or diffuse types [1]. Under this new classification system, giant cell tumours of the tendon sheath (GCTTS) became subsumed under localised TGCTs and could be further subdivided into those with intra- or extra-articular origins although such a distinction is not always clear due to the proximity of related structures. A recent review by Fraser, et al. identified 41 known cases of TGCTs in the forefoot but without further distinction with regard to the exact anatomical location of involvement [2]. Due to some likely overlap between the localized and diffuse types of TGCT, the last report on anatomical site breakdown of GCTTS in the foot identified 49 cases, of which only seven (one fourth, one third, three second, and two big toe) involved the distal phalanx [3]. Overall, the current evidence base remains equivocal for optimal treatment of such lesions, due in large part to the paucity in case numbers reported in the literature, and has ranged from topical pharmacological therapies to various types of excision (open, arthroscopic) and even irradiation and amputation.

Case History

A 36-year-old woman was initially referred by her General Practitioner for progressive swelling of the pulp of her right third toe over six months. She was otherwise healthy with no past medical or surgical history of note, and denied any history of trauma. The clinical diagnosis at the time was that of a giant cell tumour of the tendon sheath (GCTTS). Plain radiographs did not demonstrate any bony involvement (Figure 1) and an ultrasound scan performed was indeterminate.

The patient was then lost to follow-up before being re-referred three years later for persistent discomfort of the area when wearing shoes, and was otherwise, relatively asymptomatic. A repeat ultrasound scan was then performed for assessment of interval progression, and showed a modest increase in size of the lesion from 17 x 13 x 13 mm to 22 x 13 x 20 mm. The lesion was superficial and closely related to the dermis, located plantar to the extensor tendon of the third toe and centred distal to its insertion with heterogeneous internal reflectivity and a low level of internal perfusion (Figure 2). Overall, the imaging findings remained indeterminate with suspicions of an epidermoid cyst. Excision biopsy was recommended.

Following excision of the lesion under local anaesthetic, histopathological examination demonstrated sheets of foamy histiocytes with patchy collections of multinucleate giant cells, areas of fibrosis and wiry collagen deposition. There were central areas of necrosis including empty cleft-like spaces surrounded by multinucleate cells, suggestive of cholesterol clefts secondary to haemorrhage.

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*Correspondence:

Zhi Yang Ng, Department of Plastic Surgery, Queen Alexandra Hospital, Portsmouth University Hospitals NHS Trust, UK, Tel: 6589306137; E-mail: zhiyang.ng@gmail.com

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Figure 1: Plain radiographs of the right foot showing a soft tissue swelling in the third toe (yellow arrows) with no bony erosion.



Figure 2: Ultrasound image of the third toe showing a heterogeneously hypoechoic mass measuring 22 x 13 x 20 mm with low internal perfusion (yellow arrow).

Immunohistochemical staining showed the lesional cells to be positive for CD68, confirming a histiocytic origin. Therefore, the overall appearances established the diagnosis of GCTTS (Figure 3). Of note however, the excised lesion abutted the margins of resection and complete excision could not be guaranteed. At 10 months follow-up, the patient remained well with no suggestions of local recurrence.

Discussion

This rare case describes a pathology (GCTTS) occurring in an uncommon anatomical site – the third toe – with only one previous known report [3]. The aetiology of GCTTS remains unclear but most commonly, has been attributed to reactive hyperplasia associated with inflammation [4]. Fotiadis, et al. found that only 5% of patients had a definite history of trauma [5] and indeed, our patient did not report such preceding events. The histopathological findings in this case however, suggest otherwise although it may also represent secondary trauma from prolonged wearing of shoes. In terms of investigations for GCTTS, plain radiographs may show a soft tissue mass with or without erosion of the adjacent cortical bones but are often not diagnostic [6]. GCTTS may appear on ultrasonography as a solid homogeneous or heterogeneous hypoechoic mass [7] but is similarly non-diagnostic and can still be interpreted as other pathologies as in this case, an epidermoid cyst. Magnetic resonance imaging (MRI) is considered the optimal modality for pre-operative assessment and planning. GCTTS characteristically exhibits small, scattered foci of low signal on T1- and T2-weighted images due to the paramagnetic effect of haemosiderin in histiocytes and multinucleate giant cells,

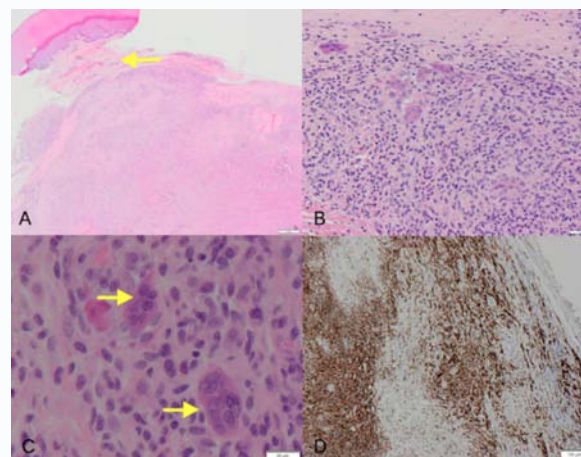


Figure 3: (A) This low-power image shows a lobulated, well-circumscribed tumour extending from the dermis of the overlying skin into the subcutaneous tissue (yellow arrow) (H&E). (B) The cellular areas contain plump spindle cells, polygonal cells and scattered multinucleate cells, which are set in a hyalinised stroma (H&E). (C) There are scattered characteristic multinucleate or 'ladybird' cells (yellow arrows). (D) There is widespread positive staining for CD68.

and the abundance of collagenous stroma [8]. MRI also delineates the extent of the tumour and its relation to surrounding structures, thus providing crucial information for preoperative planning. While ideal, MRI is not always available nor necessary and was not considered essential in our case. The patient was seen during the height of the COVID-19 pandemic in the United Kingdom, and was already clinically diagnosed as GCTTS.

Surgical excision remains the cornerstone of management of GCTTS with a reported recurrence rate of <15% [6]. While our follow-up period of 10 months was relatively short, the absence of recurrence was consistent with that reported in the literature in which there were only four cases that recurred out of 78 cases of localised TGTC of the foot and ankle region (no further breakdown by anatomical site reported) at an average of 52 months (range, 3 to 85) follow-up [2]. Our patient's presentation is consistent with the typical profile described by Barnett et al., who reported a female predominance with a palpable symptomatic mass [9]. Notably, Barnett et al. also distinguished between radiological recurrence, occurring in 14% of cases, and clinical recurrence requiring further intervention, observing that asymptomatic radiological recurrence does not always necessitate re-operation [9]. Recurrence rates vary considerably across series, likely reflecting differences in follow-up duration, case mix, and localised-diffuse overlap. Scheele et al. reported recurrence rates of 26.7% and 50.0% for localised and diffuse TGCT of the foot and ankle respectively, at a median time to recurrence of 7 months [10]. It is likely that in cases of recurrence, there may be an overlap between the localised and diffuse forms of TGTC, which may then translate to close or involved excision margins, as was the case here.

Conclusions

This case demonstrates the importance of maintaining a high index of clinical suspicion for GCTTS when faced with an enlarging and relatively asymptomatic mass in the toe. A history of trauma is not always obvious. Imaging oftentimes remain equivocal and surgical excision is the preferred treatment modality as it achieves both diagnostic and therapeutic aims concurrently. It is also important that

patients are made aware of the risk of recurrence, as the GCTTS may not inconceivably, present with an overlap of localised and diffuse forms of TGCT, leading to close or even involved resection margins.

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