Porčić et al. Scr Med. 2025 Jan-Feb;56(1):203-7. DOI:10.5937/scriptamed56-53460

CASE REPORT

Paediatric Post-Traumatic Tenosynovial Chondromatosis of the Foot: A Case Report

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Abstract

Tenosynovial chondromatosis (TC) is a rare benign condition characterised by the formation of cartilaginous nodules from tendon sheaths or articular membranes. It primarily affects the hand and wrist structures. The condition predominantly affects individuals in their thirties to fifties. The post-traumatic TC is not reported in a paediatric patient, to the best of our knowledge. The purpose of this case was to describe a five-year-old girl's case of TC, which had a highly unusual localisation in the foot following a post-traumatic event. In order to handle this challenging lesion, the clinical presentation, imaging modalities, histopathology and surgical treatment is presented. Performing surgery with wide margins is the gold standard.

Key words: Chondromatosis, synovial; Chondromatosis, tenosynovial; Synovial membrane; Paediatric; Foot; Tendons; Flexor tendon.

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Citation: Porčić Š, Jokić A, Todorovac Hodžić I. Paediatric post-traumatic tenosynovial chondromatosis of the foot: a case report. Scr Med. 2025 Jan-Feb;56(1):203-7.

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Received: 15 September 2024
Revision received: 3 November 2024
Accepted: 3 November 2024

Introduction

Synovial chondromatosis (SC) is a rare, progressive benign condition of the synovial joints characterised by metaplastic transformation resulting in the formation of cartilaginous nodules within the synovium.[1, 2] This condition has a marked predilection for the knee, hip and elbow joints. Men in the age range of 30 to 50 years are most frequently affected. Instances of multinodular extra-articular cartilaginous proliferation involving the tenosynovial membranes are rarely reported in the medical literature.[3-5] When SC occurs extraarticularly within the synovial lining of the tendon sheath, it is termed tenosynovial chondromatosis (TC). In contrast to SC, which primarily affects the joint cavities, TC typically manifests in the fingers and feet. TC of the foot is extremely rare, particularly in the paediatric population.[6, 7]

Case history

Female, five-year-old Caucasian patient presented with a two months history of pain, swelling and enlarging mass on the second toe of her left foot, following non-displaced second proximal phalanx fracture of the foot (Figure 1).

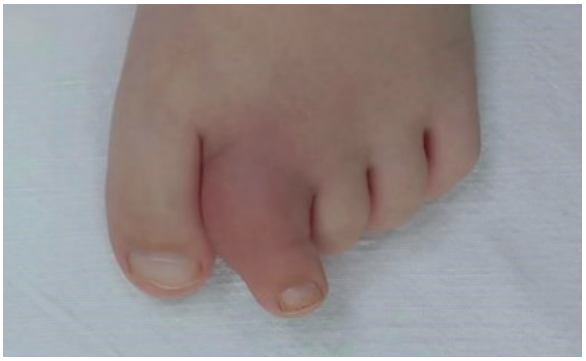

Figure 1: Clinical presentation: skin redness, swelling and enlarging mass on the second toe

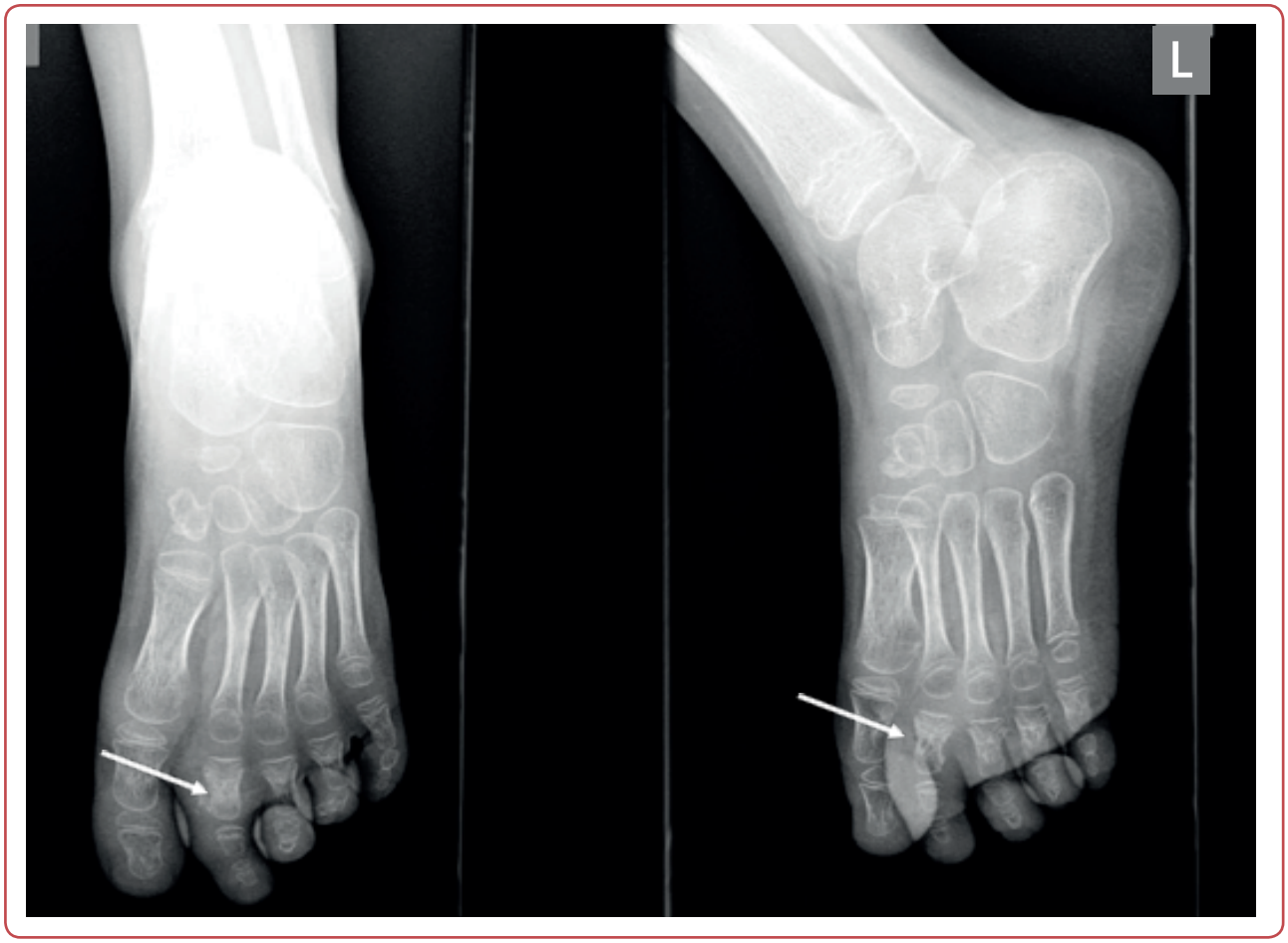

Figure 2: Radiographs of the left foot: partially calcified soft tissue mass at the level of the second proximal interphalangeal joint (white arrows)

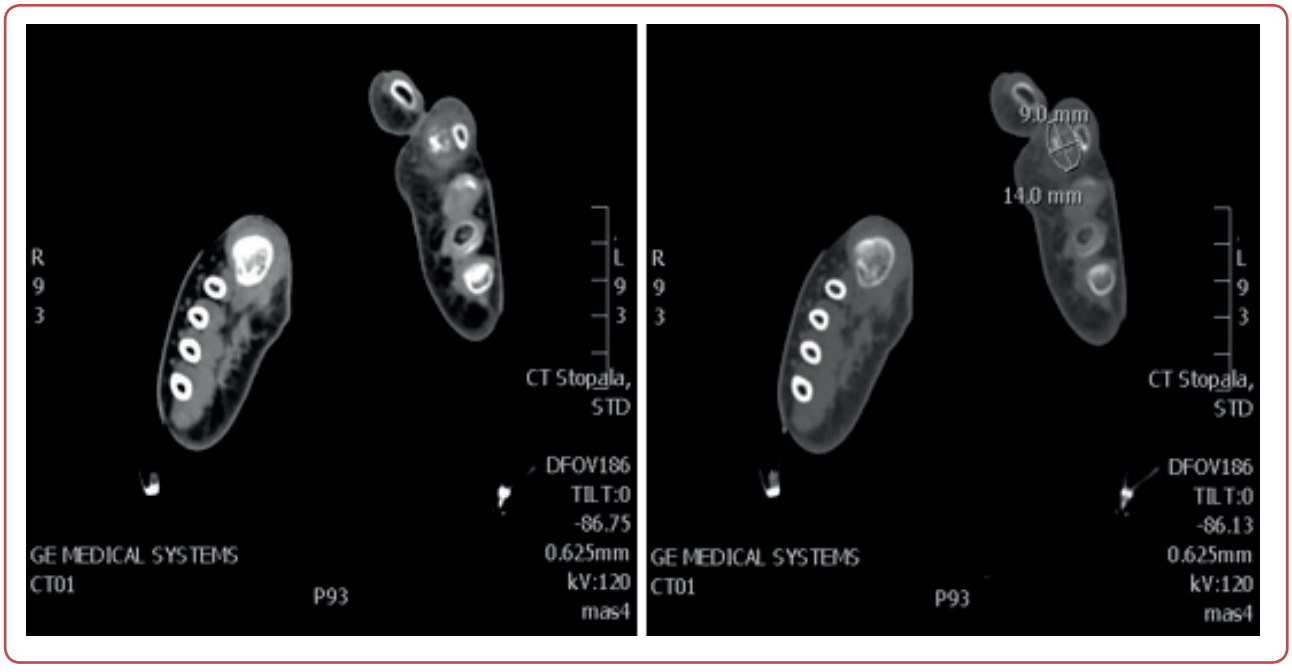

Figure 3: Computed tomography (CT) scan of the feet showing partially calcified soft tissue mass size 9.0 mm x 14.0 mm

At evaluation, clinical assessment revealed skin redness and swelling of her left second toe with tenderness on palpation and restriction on the range of motion (ROM) at active and passive flexion. The left foot exhibited no signs of neurological or vascular compromise. Laboratory tests, blood and urine were with no pathological findings. Radiographs and computed tomography (CT) scans showed a partially calcified soft tissue mass at the level of the second proximal interphalangeal (PIP) joint of the left foot (Figure 2 and 3). The mass was imaged with ultrasound, which revealed a solid mass with heterogeneous echotexture at the level of her left second toe.

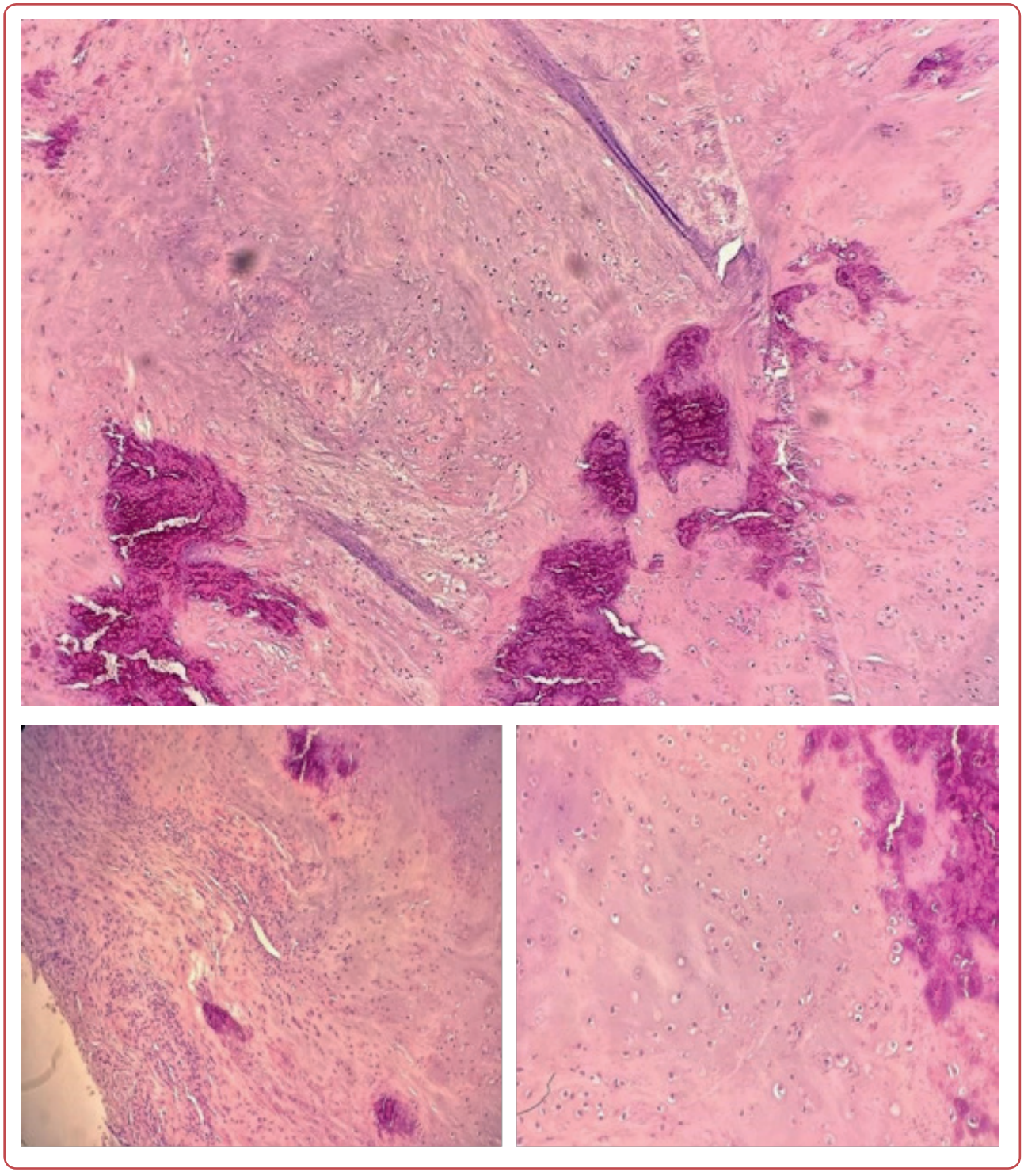

Figure 4: Haematoxylin and eosin (H&E) stain: histopathological examination of the excised mass, from low to high magnification, reveals well-differentiated chondrocytes located within individual lacunae with diffuse calcium deposits

Upon diagnostic confirmation, patient was scheduled for a surgical extirpation. At surgery, patient underwent general anaesthesia. A 60-degree plantar Z-shaped incision was performed and the tumour was visualised at the level of proximal phalanx of the second toe. During the procedure care was taken to preserve plantar digital arteries and nerves. *In toto* tumour resection with synovectomy of *flexor digitorum longus* and *brevis* tendon sheath was performed. Patient tolerated procedure without major complications. Histological examination revealed synovial chondromatosis, with no evidence of malignant transformation (Figure 4). At one-week follow up, the patient was asymptomatic and demonstrated a full ROM. At the six-month postoperative point during the preschool health screening, the patient showed no symptoms.

Discussion

Synoviocites are cells lining the articular cavity, tendons or bursae. They are responsible for secretion of larger biomolecules and also mediate the filtration of plasma from the underlying capillaries. Some histological findings suggest that synoviocytes are only anchored in the loose connective tissue, without the support of basal membrane, allowing more transudate to escape into the cavity. This is especially important during the inflammation reaction, as more fluid is able to accumulate in the articular cavity, causing the compression of the surrounding tissue. Some findings suggest that synoviocytes are reactive to this kind of stimulation. Namely, it was indicated in the paper by Momberger et al that stretching of the rabbit synoviocytes, induced change of gene expression, that resulted in increased secretion of hyaluronan.[8] Another important aspect underlying the histological arrangement of the synovial membrane implies the insufficiency of mechano-regulatory component epithelial cells have, opposed to epithelium in other parts of the body. Antiproliferative effect in epithelium having basal membrane is acquired by interaction of transmembrane proteins such as integrins and the proteoglycans. Not having this kind of control mechanism, could explain the tendency for chondromatous and malignant alteration, as a response to trauma, proinflammatory cytokines, mechanical stimulation and consequent change of gene expression. It is believed that chondroid metaplasia is caused by bone morphogenetic protein (BMP). Ozyurek et al proposed a neoplastic origin for primary TC associated with abnormalities on chromosome 6. These cytogenetic abnormalities do not appear in secondary synovial chondromatosis.[9, 10]

SC is classified under two main types: primary and secondary synovial chondromatosis. The exact cause of the changes in the primary form of chondromatosis, which is manifested by the appearance of ectopic cartilage tissue in the synovial sheath, cannot be determined. Unlike the primary form, the secondary form is always associated with pre-existing pathology of the affected joint such as trauma, osteoarthrosis, infections. To the best of our knowledge, there are no reported cases of post-traumatic SC in paediatric patients. At this unusual age and location, there is a greater chance of a missed diagnosis.

Although the precise pathophysiology of SC remains uncertain, it has been hypothesised that synoviocytes initially experience neoplastic cartilage tissue proliferation along with synovial and cartilage nodule hyperplasia. The discrepancies in TC aetiology may be caused by two separate kinds. Secondary TC is associated with a history of osteoarthritis, osteochondral fractures, or neuroarthropathy, whereas primary TC has an unclear aetiology.[11]

Malignant change is extremely rare, but it is documented usually in the primary form. Surgeons are advised to undertake a meticulous synovectomy with extensive margins and excise any loose bodies.[12]

Conclusion

A rare case of post-traumatic TC in paediatric patient is presented. The early diagnosis and surgical excision of this rapidly growing tumour are important for preventing complications such as malignant transformation, blood vessel occlusion and tendon destruction. Besides TC, several other things should be considered in the differential diagnosis such as ganglion cyst, giant cell tumour of tendon sheath (GCTTS), osteochondroma, as well as synovial sarcoma. Regardless of its clinical presentation, malignant change is atypical and it is usually documented in the primary form of SC. Surgeons are advised to undertake a meticulous synovectomy with extensive margins and excise any loose bodies.

Ethics

Our institution does not require ethics approval for reporting individual cases or case series. A written informed consent for anonymised patient information to be published in this article was obtained from the patient legal guardian.

Acknowledgement

We extend our sincere appreciation to the Department of Pathology at Cantonal Hospital Bihać for their valuable contribution to the development of this manuscript.

Conflicts of interest

The authors declare that there is no conflict of interest.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Data access

The data that support the findings of this study are available from the corresponding author upon reasonable individual request.

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