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Case Report

# Intraarticular Involvement of Extraarticular Giant Cell Tumor of the Tendon Sheath in the Thumb of a Pediatric Patient: A Case Report

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**Abstract**
Giant cell tumors of the tendon sheath (GCTTS) are locally invasive and have a high likelihood of recurrence in adults, but it is rare in children. Its potential to be invasive or recur has not been characterized. We present a 9-year-old patient with a dumbbell-shaped GCTTS that had small intraarticular and large extraarticular components with a narrow connection between them. This case suggests that GCTTS can be locally invasive in children. This case report could help hand surgeons who treat GCTTS in children.

**Keywords**
giant cell tumor of the tendon sheath, hand, joint, pediatric

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## Background

Giant cell tumor of the tendon sheath (GCTTS) is a benign soft tissue tumor arising from the synovium, bursae, or tendon sheath. Its reported incidence ranges from 1.8 to 50 per million[1,2)]. It is classified into intra- and extraarticular types and is subclassified into localized and diffuse types[1,3)]. Localized GCTTS is approximately three times more common than the diffuse type[1)]. The hand and large joints are commonly affected in female adult patients aged 20-50 years. Regarding the hand, occurrence near the distal interphalangeal joint of the index and middle fingers is the most common[4)]. Although GCTTS is locally invasive and has high likelihood of recurrence in adult patients, it has not been well characterized in pediatric patients. This study describes a pediatric patient with a dumbbell-shaped GCTTS in the thumb consisting of intra- and extraarticular lesions.

## Case Presentation

A 9-year-old boy presented with a tender nodule on the radiopalmar aspect of the interphalangeal joint of the left thumb. He first noticed a painless nodule 3 months prior at the site of the tumor, which gradually became tender. There was no history of trauma, deformity, or pain in the thumb. Doppler ultrasound examination revealed a round 12 × 9-mm heterogeneous hypoechoic lesion with poor vascularity. The lesion was diagnosed as a ganglion cyst due to its continuity with the interphalangeal joint space (**Figure 1**). Computed tomography showed a subcutaneous tumor with heterogeneous internal intensity. There were no morphological abnormalities in adjacent phalanges. Radiologists diagnosed it as a ganglion cyst.

Surgery was performed under general anesthesia. Under tourniquet control, a Y-shaped skin incision was made on the radiopalmar aspect of the thumb. A hard, round, yellowish mass was subcutaneously exposed. The tumor was resected with the radial neurovascular bundle preserved. Further inspection around the interphalangeal joint revealed a small residual mass on the radial side of the A2 pulley (**Figure 2(a)**). The mass had a narrow pedicle on the bottom, which arose from the interphalangeal joint, indicating that intraarticular lesion might have not been completely resected. By dissecting the pedicle circumferentially, we extirpated the mass en bloc with a smaller intraarticular lesion with a dumbbell-like morphology (**Figure 2(b)**). Histopathological findings of a mixture of multinucleated giant cells and histiocytic cells associated with giant cells lead to the

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diagnosis of GCTTS (**Figure 3**). The incision healed without any complications. There were no functional disabilities or signs of recurrence at 1 year after surgery.

## Discussion

GCTTS in children is very rare. Its exact incidence in children is unclear since the age range for pediatric cases varies in the literature. Ushijima et al.[4] reported that 9 (4.3%) of 207 patients with GCTTS were aged 9 years or younger. Shi et al.[5] reported that 16 (4.4%) of 135 patients with GCTTS were aged 16 years or younger. Based on these reports and the overall incidence of 1.8-50 per million[1,2], the incidence of pediatric GCTTS is estimated to be 0.1-2 per million in the population. Since little is known about the etiology, recurrence, or other features of GCTTS specific to children, accumulating knowledge on GCTTS in children from case reports is of scientific and clinical importance.

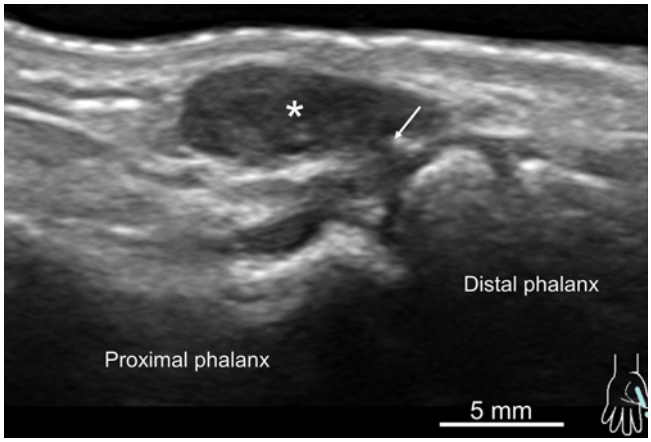


**Figure 1.** Ultrasound image showing a hypoechoic subcutaneous mass with poor vascularity (asterisk). Note the presence of a narrow continuity into the interphalangeal joint space (arrow).

The etiology of GCTTS remains controversial. Jaffe et al. argued that GCTTS is a reactive or regenerative hyperplasia associated with inflammatory responses[6]; this theory has been widely accepted, but there is insufficient evidence. Kerfant et al.[7] described a unique case in which a patient had multiple GCTTS on the volar and dorsal sides of three digits in the same hand. Based on the separate occurrence of volar and dorsal lesions and the patient's history of repetitive trauma to the hand, they suggested that microtrauma could be a cause of GCTTS. In contrast, Abdul-Karim et al. suggested that GCTTS is a neoplastic lesion[8]. When they investigated the DNA content and proliferative index of localized GCTTS, diffuse GCTTS, and pigmented villonodular synovitis, they found that diffuse GCTTS is associated with a significantly higher proliferative index, although histopathological findings were similar for those three conditions. There are no reports on the etiology of GCTTS in children. The patient in this case had played baseball for several years, which may be associated with the cause of GCTTS.

In adults, GCTTS recurrence rates range from 9% to 44%[9,10]. Radiographic evidence of bony erosion, presence of degenerative joint disease, tumor involvement in joints and adjacent soft tissues, and high cellularity and mitotic activity on histological examination are risk factors for recurrent GCTTS. Reilly et al.[9] evaluated the radiological characteristics of 70 patients with GCTTS who were surgically treated

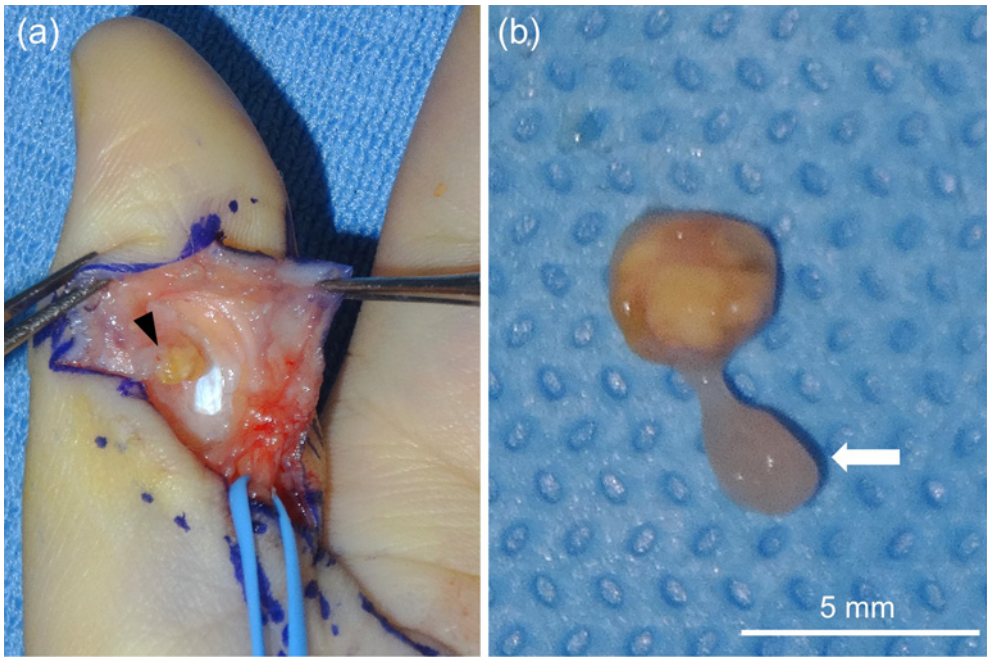


**Figure 2.** (a) Intraoperative photograph showing a small yellowish mass (arrowhead) adjacent to the flexor pollicis longus tendon at the level of the interphalangeal joint. (b) The extirpated mass had a dumbbell-like morphology with a smaller intraarticular component (arrow).

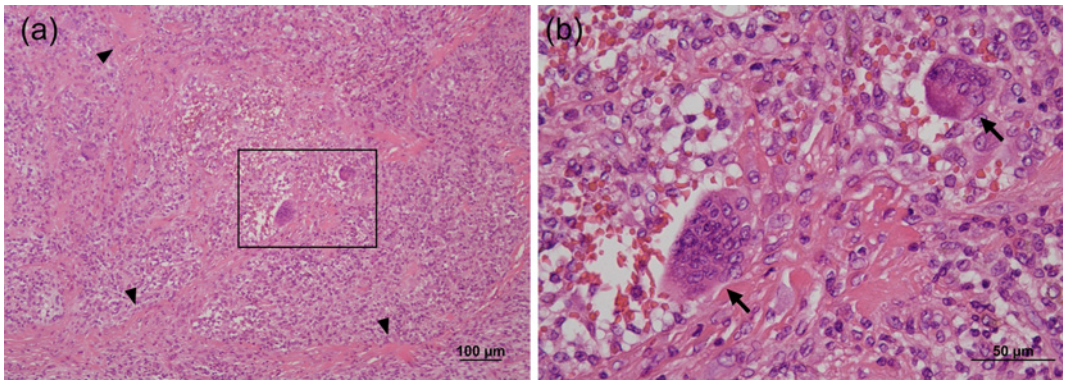


**Figure 3.** Histopathological examination with hematoxylin-eosin staining. (a) Low power view (×100 magnification) and (b) close-up view of the rectangular region in (a). Histiocytoid mononuclear cells that lack cytologic atypia and multinucleated giant cells (arrows) were distributed in the hyalinized stroma (arrowheads).

and followed. Bony erosion was more frequently observed in patients with recurrence [5 (26%) of 19 patients with recurrence vs. 3 (6%) of 51 patients without recurrence]. Moreover, degenerative joint disease was observed in patients with recurrence [8 (42%) of 19 patients with recurrence vs. 9 (18%) of 51 patients without recurrence].

Few cases of recurrent GCTTS have been reported in children. Civan et al.[11] treated nine patients with GCTTS between the ages of 10 and 17 years, one of whom experienced recurrence. Gholve et al.[12] reported that there are no recurrences among 29 patients in a pediatric case series. The absence of degenerative joint disease in children might be associated with the low incidence of recurrent GCTTS. Intraarticular or near-joint occurrence is another risk factor for recurrent GCTTS[9]. Moreover, Mane et al.[13] treated intraarticular nodular tenosynovitis of the carpometacarpal joint in a 15-year-old male patient with endoscopic resection. Our patient had a narrow connection between the intra- and extraarticular parts of the lesion, which had a dumbbell-like shape. The larger extraarticular part suggests that the original lesion might have been extraarticular, with subsequent intraarticular extension.

Preoperative diagnosis is crucial to avoid residual joint lesions, especially for lesions in para- or intraarticular regions. Careful palpation is used to confirm stiffness and adhesions to surrounding tissues. Plain radiography and computed tomography can be used to detect bone erosions and degenerative changes in adjacent joints. Ultrasonography provides information on size, solid or cystic nature, vascularity, location relative to surrounding tissues, and presence of satellite lesions[9,14]. Magnetic resonance imaging can show characteristic features specific to GCTTS, such as a mass with heterogenous low-intensity signals on T1- and T2-weighted images due to the presence of hemosiderin[15]. The differential diagnosis of GCTTS in the hands includes ganglion cyst, lipoma, fibroma, and foreign body granuloma[16]. Ganglion cyst is the most common. GCTTS is sometimes misdiagnosed as ganglion cyst due to clinical similarities in stiffness, immobility, and near-joint occurrence[17]. Ganesh et al.[18] reported a case of clinically and radiologically suspected recurrent ganglion cyst with osteomyelitis that was finally revealed to be GCTTS. Monaghan et al.[19] reported that GCTTS was diagnosed provisionally only in 3 of 71 patients; the remaining patients were misdiagnosed as having a ganglion cyst or an epidermal cyst. In this study, we used ultrasound because it is noninvasive and there was no need for sedation, which is suitable for use in pediatric patients. Intra- and extraarticular lesions and their connections were successfully visualized with a high-frequency transducer. However, the tumor was misdiagnosed as a ganglion cyst. This fact suggests that magnetic resonance imaging should also be considered if a solid mass is identified near a joint. If intraarticular involvement is preoperatively suspected, careful intraoperative inspection of the area around the joint should be conducted.

## Conclusion

We treated a pediatric patient with a dumbbell-shaped GCTTS that had both small intraarticular and large extraarticular components with a narrow connection. This manifestation suggests that pediatric GCTTS can involve joints and be locally invasive. We believe that this case report could help hand surgeons who treat GCTTS in children.

**Author Contributions:** A.M., S.S., and T.T. treated the patient and contributed to the writing of the manuscript.
**Conflicts of Interest:** There are no conflicts of interest.
**Ethical Approval:** This study does not require ethical approval because this is a case report.
**Consent to Participate:** Written informed consent was obtained from the patient included in this study.
**Disclaimer:** Author Susumu Saito is one of the Editorial Committee members of Journal of Plastic and Reconstructive Surgery. This author was not involved in the peer-review or decision-making process for this paper.

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