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Tenosynovial giant cell tumor in the elbow of a child with the sole symptom of extension disturbance

Akio Sakamoto, MD, PhD*, Takashi Noguchi, MD, PhD, Shuichi Matsuda, MD, PhD

Department of Orthopaedic Surgery, Graduate School of Medicine, Kyoto University, Kyoto, Japan

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Tenosynovial giant cell tumor (TGCT) is characterized by variable proportions of synovial-like mononuclear cells, multinucleated giant cells, and foamy histiocytes. TGCTs are classified based on growth pattern, whether they are a localized nodule ("nodular-type") or a diffuse villous hyperplasia ("diffuse-type") of the lesion. The localized-type TSGCT tends to occur in small joints. The diffuse-type TGCT (D-TGCT) is often referred to as pigmented villonodular synovitis, and predominantly affects large joints, such as the knees. D-TGCTs tend to be locally aggressive and to recur,[7] in contrast to the localized-type.[5] TSGCT arising in the elbow is rare regardless of type.[1]

D-TGCT can have an osseous extension. In the case of D-TGCT in the spine, the bone lesion is typically characterized by osteolytic and expansive findings.[2] Therefore, differential diagnosis of D-TGCT with osseous domination includes benign or borderline aggressive bone tumors. TSGCT arising in the elbow is rare.[1] In this illustrative case, the sole symptom was extension disturbance, initially recognized as a bone tumor from images because of the osseous extension.

Case presentation

A 13-year-old boy noticed an extension disturbance without pain in the elbow. He engaged in no sports activities besides school gymnastics class. Plain radiographs showed no obvious findings. The range of motion was restricted to -15° extension, while full extension of the other elbow was possible. No tenderness over the joint was observed and there was no swelling. Computed tomography (CT) showed an expansive osteolytic lesion in the cubital fossa (Fig. 1). Magnetic resonance imaging (MRI) revealed a lesion with low- to intermediate-signal intensity on T1-weighted images, and high-signal intensity regions on T2-weighted images (Fig. 2). The preoperative diagnosis was a benign bone tumor, such as a chondroblastoma, considering the patient's age. Extension difficultly was thought to be due to extraosseous expansion of the lesion. Retrospectively, a small synovia-like nodule at the tip of the olecranon was recognized with low- to intermediate-signal intensity on T1- and T2-weighted images.

During surgery under general anesthesia, the extension disturbance was -10°. Under arthroscopy and fluoroscopy, intraoperative frozen histological analysis of a sample taken from the osseous lesion showed a proliferation of spindle-shaped cells, but a conclusive diagnosis could not be made due to the small size of the sample. On careful arthroscopic examination, the synovial-like nodule looked brownish, continuing to the osseous lesion (Fig. 3, *A*). Additional intraoperative frozen histological analysis sampling from the synovial-like nodule showed a proliferation of fibrohistiocytic cells without atypia, the same as for the osseous lesion. Based on these observations, TSGCT was diagnosed during surgery. Upon diagnosis, complete resection and curettage was performed in an open surgery with longitudinal incision on the posterolateral elbow. The joint space was revealed by splitting the triceps muscle on the radial side. After resection of the lesion, full extension of the elbow joint was regained. The resected specimen revealed proliferating fibrohistiocytic cells, embedded in a fibrous stroma (Fig. 3 *B* and *C*), confirming the diagnosis of TSGCT. No recurrence or symptoms of extension disturbance were recognized one year after surgery.

This case report has been approved by the institutional review board (R2499), and the patient was informed of the report.

Discussion

The elbow is a rare site for TSGCT with only a few cases reported. A previous review of TSGCT in the elbow indicated that it

This study has been approved by Kyoto University Institutional Review Board (R2499).

* Corresponding author: Akio Sakamoto, MD, PhD Department of Orthopaedic Surgery, Graduate School of Medicine, Kyoto University, Shogoin, Kawahara-cho 54, Sakyo-ku, Kyoto, Japan.

E-mail address: akiosaka@kuhp.kyoto-u.ac.jp (A. Sakamoto).

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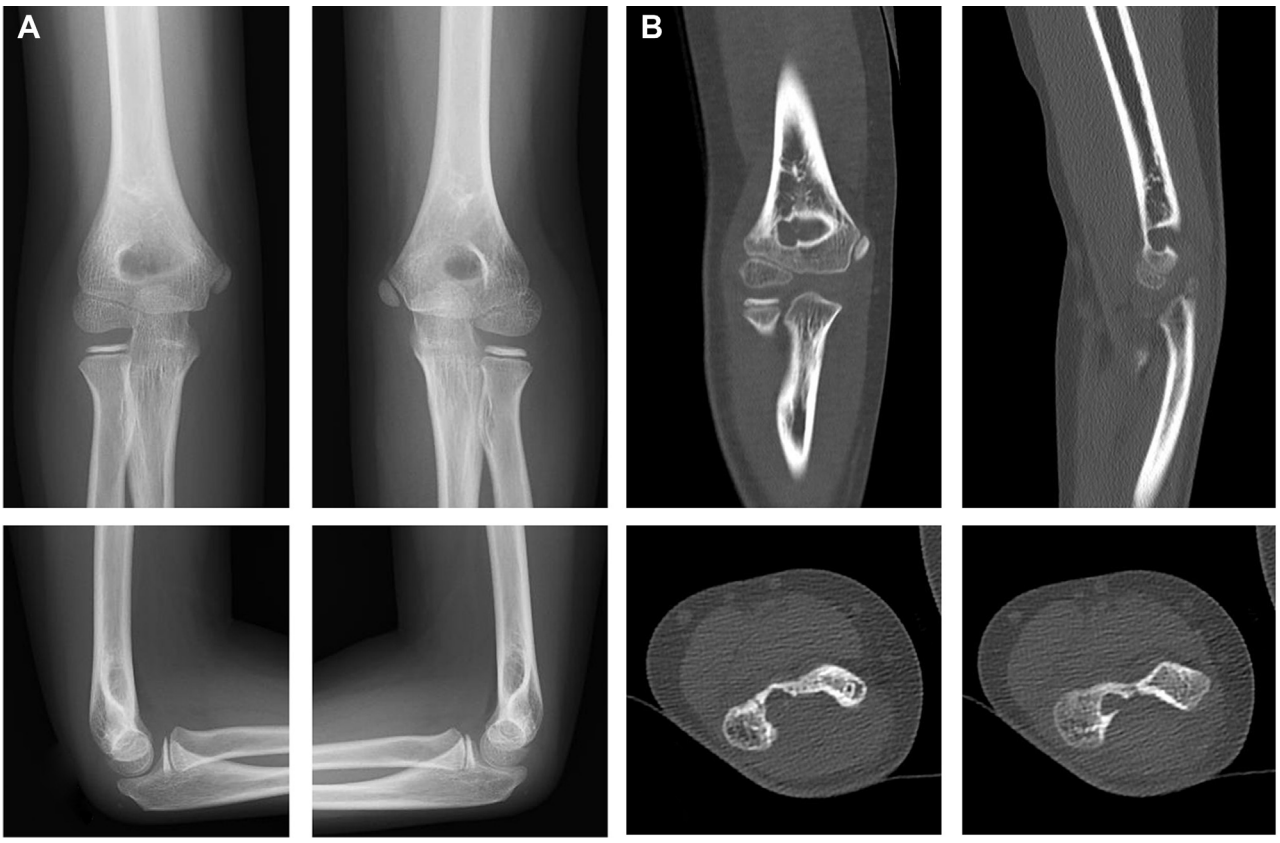

Figure 1 A 13-year-old boy with tenosynovial giant cell tumor in the elbow. (**A**) Plain radiographs show subtle changes. (**B**) The CT shows an expansive osteolytic lesion in the cubital fossa.

predominantly occurs in females between the ages of 6 and 82 years, with an average age of 40.3 years.[1] Clearly, TSGCT in the elbow can occur at any age, as in the current case of a 13-year-old boy. Reported common complaints of TSGCT in the elbow are pain and swelling. Here, a disturbance of elbow extension was the only symptom. A limited range of motion is reported in one-third of cases with elbow TSGCT. However, a limited range of motion as a sole symptom has not been observed among the review series of 27 cases.[1]

The average duration of symptoms is reported to be 33.2 months.[1] The current patient was symptomatic for only 3 months, but osteolytic findings were apparent in the initial image assessment and long-term existence of the lesion was considered. The osseous lesion seemed to be asymptomatic but a small synovia-like nodule at the tip of the olecranon created the extension disturbance. The appearance of a limitation to extension will be different according to the tumor location in the elbow.

Bone involvement in TSGCT is characterized by expansile osteolysis. In the current case, the osteolytic finding was demonstrated by CT and mimicked a bone tumor. TSGCTs are reported to show low- to intermediate-signal intensity on both T1- and T2-weighted images on MRI.[4,5] Low-signal intensity, particularly on T2-weighted images, is reported to be due to the presence of hemosiderin.[4] In this case, a synovia-like nodule caused the low- to intermediate-signal intensity on both T1- and T2-weighted images. The osseous lesion had high-signal intensity on the T2-weighted image, distinct from that of the synovia-like nodule.

A diagnosis of chondroblastoma was made based on imaging. Chondroblastoma occurs in the epiphysis or apophysis in children. In a previous report of chondroblastoma, almost all cases had a geographic margin with well-defined borders. Most lesions had high signal intensity on T2-weighted images and iso- to hypo-signal intensity on T1-weighted images. A chondroblastoma demonstrates peri-lesional marrow edema.[3] The difference from the typical chondroblastoma in our case was that there was no bone edema, and the osteolytic lesion was seen in the metaphysis. The anatomical difference between epiphysis and metaphysis is especially important. However, a chondroblastoma occurring in the femoral metaphysis has been reported in a 15-year-old girl.[4]

Curettage with intralesion resection is an acceptable treatment for TSGCT. The local recurrence rate of TSGCT in the elbow has been reported to be 17.4%.[1] A review found that a diffuse-type TSGCT in any joint has a tendency to present with more concerning symptoms than the localized form.[6] The majority of TSGCT in the elbow are reported to be the diffuse type.[6] In the current case, it was difficult to determine whether the lesion was a diffuse type or localized type. It appeared to be located in the cubital fossa, but the obvious osseous extension looked diffuse.

In the current case, because the lesion was small, intraoperative pathological diagnosis was performed. After the diagnosis of TSGCT, resection via open surgery was performed. The osteolytic lesion was located on the radial side of the distal humerus. A longitudinal incision at the cubital fossa on the radial side minimizes the risk to radial or ulnar nerve. Furthermore, split of the triceps minimizes damage to this muscle.

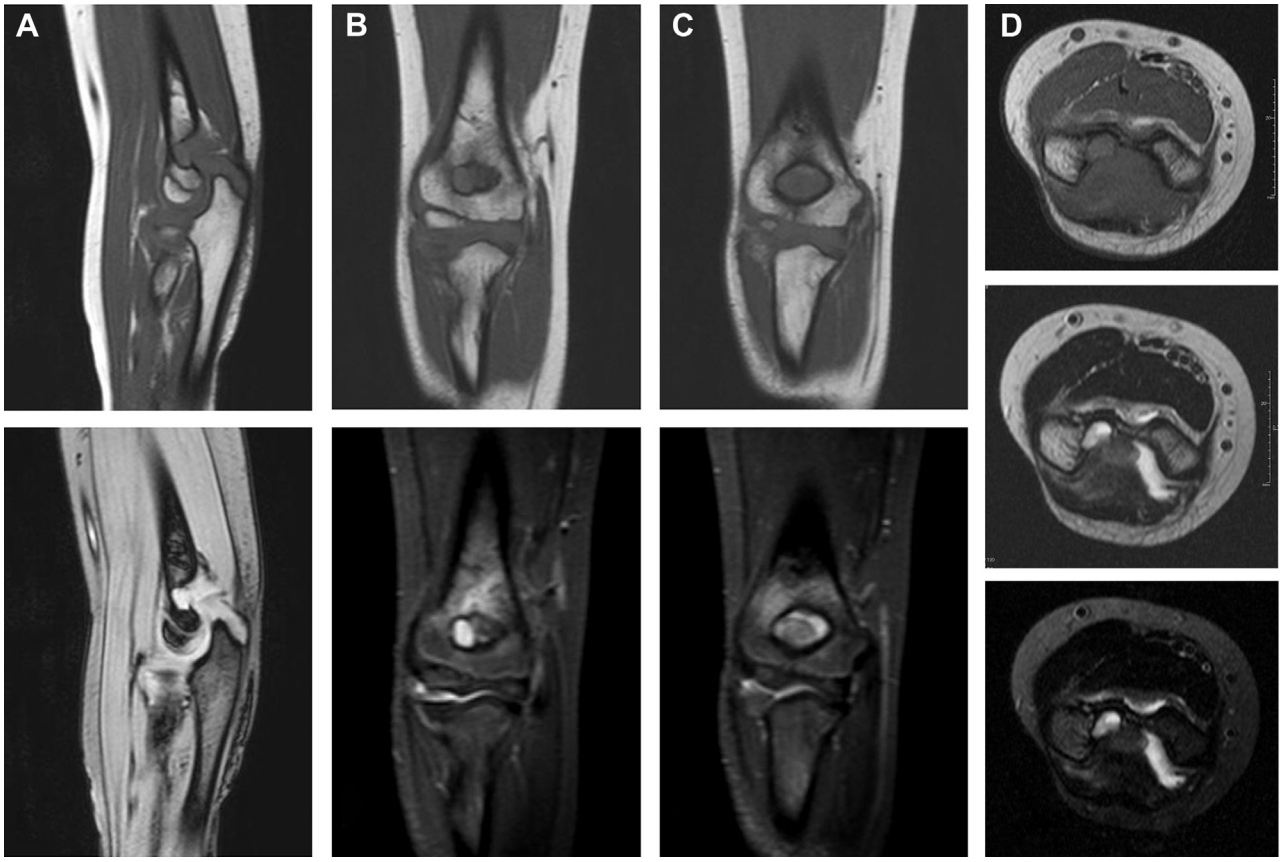

Figure 2 Tenosynovial giant cell tumor in the elbow of the 13-year-old boy. MRI shows the lesion with low- to intermediate-signal intensity on the T1-weighted image (**A**-top) and high-signal intensity on the T2-star (T2*) weighted image (**A**-bottom). Coronal views focusing on osseous (**B**) and extraosseous (**C**) lesions show low- to intermediate-signal intensity on the T1-weighted image (**B**-top, **C**-top) but high-signal intensity for the osseous (**B**-bottom) and low- to intermediate-signal intensity for the extraosseous (**C**-bottom) lesion on T2-weighted fat-suppression images. Axial views show the lesion located at the tip of the olecranon process in extension (**D**) (top: T1-weighted image; middle: T2-weighted image; bottom: T2-weighted fat-suppression image).

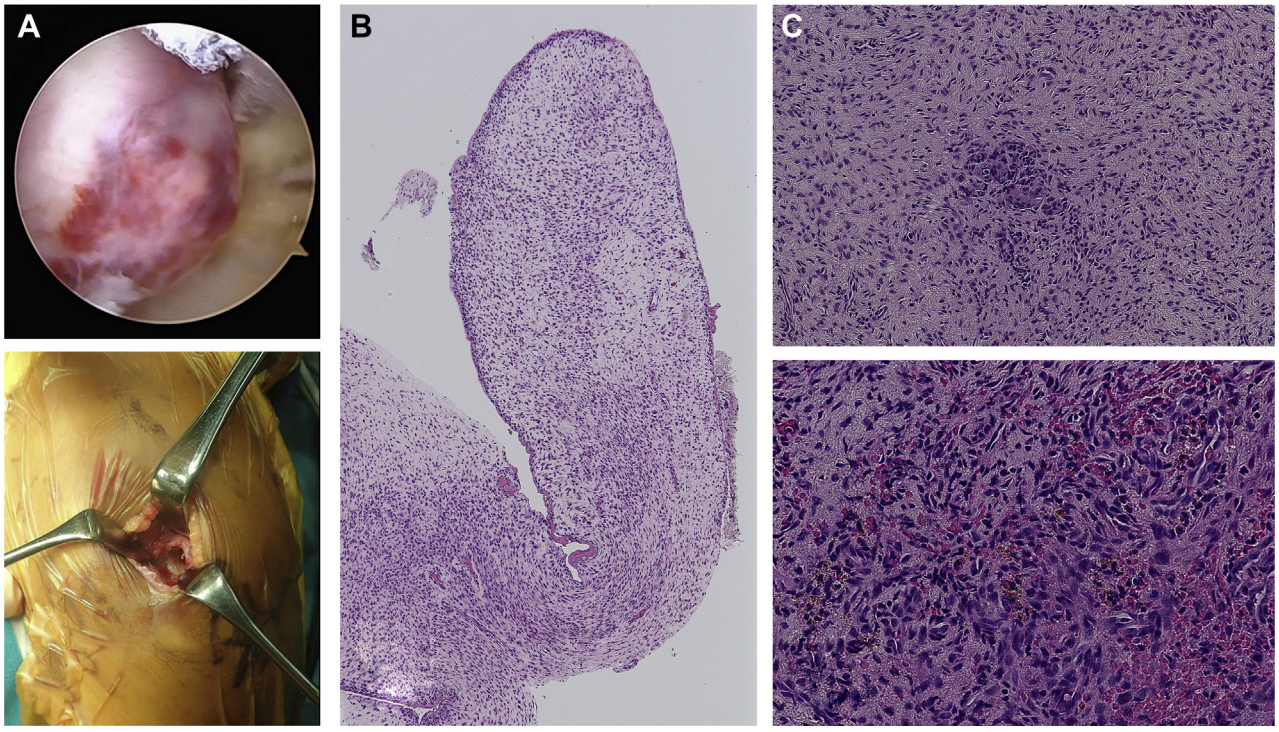

Figure 3 Arthroscopy shows the extraosseous lesion as a brown nodule (**A**-top). After resection, the bone cavity is obvious (**A**-bottom). Low magnification view shows a villous structure (**B**). The lesion is composed of proliferating mononuclear fibrohistiocytic cells in various amounts within fibrous stroma (**C**). Hemosiderin deposition can be observed (**C**-bottom).

Conclusion

A case of TSGCT in the elbow with the sole symptom of extension disturbance is reported. For diagnosis of TSGCT, CT is useful for the osteolytic identification, which can otherwise mimic an aggressive bone tumor. Different signal intensities on MRI between osseous and extraosseous lesions made the diagnosis of TSGCT difficult. The current case with atypical clinical and image findings is instructive for the correct diagnosis of TSGCT in the elbow.

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