Tenosynovial Giant Cell Tumors in the Carpal Joint

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Abstract
This paper examines three cases of localized-type and diffuse-type tenosynovial giant cell tumor (TGCT) in the carpal joint. Usually, the site of the localized-type TGCT is the finger and the site of the diffuse-type TGCT is the knee joint. TGCT is rarely found in the carpal joint. In each case, there was only swelling in the carpal joint, but no pain or disturbance in wrist motion. X-rays and Computed Tomographic (CT) scans showed bony impressions in the carpal joint. Cases 1 and 2 had both intra- and extra-articular lesions. The intra-articular lesions were a diffuse-type TGCT and the extra-articular lesions were localized-type TGCT. Case 3 only had an intra-articular lesion, which was a localized-type TGCT. The tumors were completely excised under a surgical microscope. There were no recurrences at the follow-up examinations which took place one, four and five years later, because the tumors were completely excised. Although a localized-type TGCT is prominent out of the joint, a diffuse-type TGCT was carefully excised from the joint. We noticed that diffused-type TGCT often exists in a hand and finger joint lesion, and if these lesions are not excised completely, recurrence might occur.

Keywords: Giant Cell Tumor; Tendon Sheath; Tenosynovial; Carpal Joint

Introduction
Giant cell tumors (GCT) originating from tenosynovia are common soft tissue tumors in the hand and knee area. GCT of the tendon sheath in the hand is usually classified as either nodular, such as nodular tenosynovitis in the knee joint, or diffuse, such as pigmented villonodular synovitis (PVNS) in the knee joint [1]. However, World Health Organization (WHO) changed the classification of GCT originating from tenosynovia either localized-type or diffuse-type tenosynovial GCT (TGCT) in 2013 [2] (Table 1). Usually, the site of the localized-type TGCT is the finger and the site of the diffuse-type TGCT is the knee joint. TGCT is rarely found in the carpal joint. In this report, we discuss three cases of TGCT in the carpal joint, that present both localized-type and diffuse-type TGCT (Table 2).

Case Report
Case 1: 20 year-old female
The patient noticed swelling in her left wrist five years ago. She felt no pain, but the swelling became worse (Figure 1A). A physical examination detected no neurological defect, or disturbance in wrist motion. A Magnetic Resonance Imaging (MRI) found intra- and extra-articular lesions in the radio-scaphoid joint. Both the intra- and extra-articular tumors were excised (Figure 1B). The tumor was excised completely under a surgical microscope (Figure 1C). There was no recurrence for five years after surgery.

Case 2: 28 year-old male
The patient noticed swelling in his left dorsal wrist two years ago. He felt no pain, but the swelling became worse and the radial and ulnar sides were also swollen. He was referred to our hospital, because a biopsy preformed at his previous clinic indicated TGCT. A physical examination detected no neurological defect, or wrist...
motion disturbance. An MRI found radial and ulnar side, as well as intra- and extra-articular lesions in the carpal joint (Figure 2A). Both the intra- and extra-articular tumors were excised (Figure 2B). Since the bone was impressed but the tumors were not invasive, it was excised completely under a surgical microscope (Figure 2C). The extra-articular lesion was the brown-yellowish color which is characteristic of localized-type TGCT. Giant cells were also seen in the area of the localized-type TGCT. However, the intra-articular lesion was in dark-reddish color, which characterizes diffused-type TGCT (Figure 2C, arrows). The intra-articular pathological findings showed a synovial-like lesion which is characteristic of diffuse-type TGCT. It had a villous appearance with many cleft-like spaces (Figure 2D). Pigmented macrophages (siderophages) were often observed, but few osteoclastic giant cells were detected. There was no recurrence for four years after surgery.

Case 3: 36 year-old female

Although the patient injured her left hand in a fall, an X-ray showed a bony lesion in the right hand by chance. The woman was referred to our hospital in order to diagnose the lesion. A physical examination found no neurological defect, or right wrist motion disturbance. The Computed Tomographic (CT) scan, however, detected a bony impression at the base of the 3rd, 4th and 5th metacarpal bones, the hamate and the capitate (Figure 3A). An MRI showed a small tumorous lesion with the same intensity of

Figure 1A: A tumor is excised completely. (The upper and right part is from the outside of the wrist joint, which is a characteristic of localized-type TGCT. The synovial-like lesion existed from the joint is a diffuse-type TGCT (arrow)).

Figure 1B: Intraoperative findings shows diffuse-type TGCT lesion concomitant to the wrist joint (an arrow). There is a large localized-type TGCT lesion beneath the retractor.

Figure 2A: The MRI shows a tumor at the radial and ulnar sides of the carpal joint. (Both intra- and extra-joint lesions are seen. The intra-articular lesion displays osseous impression of the triquetrum, the hamate, and the scaphoid).

Figure 2B: Intraoperative findings shows complete excision of the tumor from the triquetrum-hamate joint. (The tumor is excised with a joint capsule. Carpometacarpal joint cartilage (an arrow) and a cave of the hamate bone (an arrow head) are seen after complete excision).

Figure 2C: The tumor is excised in two pieces. (The upper specimen is from the radial side and the lower is from the ulnar side. The nodular lesion looks yellowish brown color, which is a characteristic of localized-type TGCT. The synovial-like lesion existed from the joint looks like a dark reddish tongue, and is a diffuse-type TGCT (arrows)).
muscle and synovia. The tumor was well encapsulated and excised completely under a surgical microscope (Figure 3B). Pathologically, the tumor was grayish white, nodular and slightly lobulated. In the surface of the tumor, the stroma was fibrous with foci of hyalinization and there was a sparse distribution of tumor cells (Figure 3C). There was no recurrence for one year after surgery.

**Discussion**

GCT originating from tenosynovia used to be divided into two main subtypes according to the site (intra- or extra-articular) and growth pattern (localized or diffuse). The two types differed in their clinical features and biological behaviors, but appeared to share a common pathogenesis. This family of lesion included localized GCT of the tendon sheath/tenosynovial GCT, a more diffuse and destructive variant called diffuse-type GCT/PVNS. However, GCT originating from tenosynovia was renamed as TGCT by WHO and classified as either localized-type or diffuse-type in 2013 [2] (Table 1). Localized-type TGCT, according to WHO [2], is composed of synovial-like mononuclear cells, accompanied by a variable number of multinucleate osteoclast-like cells, foam cells, siderophages and inflammatory cells, which used to be a GCT of the tendon sheath. Diffuse-type TGCT is composed of synovial-like mononuclear cells, admixed with multinucleate GCT, foam cells, siderophages and inflammatory cells. This type of GCT used to be classified as PVNS in the knee joint (Table 1). Usually the site of the diffuse-type TGCT is the knee joint. Since there is no definitive factor to establish the type of TGCT, definition of localized- or diffuse-type TGCT should be determined, not only by the patho-histology but also by the location and growth pattern. GCT of the tendon sheath is classified as localized-type TGCT according to WHO. However, GCT of the tendon sheath of finger sometimes has both intra- and extra-articular lesions [1]. Although extra-articular lesions display the qualities of localized-type TGCT, intra-articular lesions exhibit the characteristics of diffuse-type TGCT. Hence, one tumor sometimes includes two subtypes of TGCT at the same time (Table 1). We should recognize that there are not only localized TGCT but also diffuse type TGCT in the hand and finger lesion.

At our hospital, we treated three cases of carpal joint TGCT (7.1%) out of 42 TGCT in the hand. There was no report of carpal lesion out of 27 cases and 46 cases in the hand [3,4]. Although there was a report of wrist TGCT, it was not in the carpal joint, but in the tenosynovia at the wrist level [5]. Hence, the carpal joint is recognized as a rare location for the origin of TGCT.

The average age of our three patients with carpal lesion TGCT was 28 years old, which is younger than the 43 years old and 36 years old of prior reports of TGCT in the hand [1,3]. While there were no symptoms of carpal lesion TGCT, two cases had painless-swelling at the carpal joint. There was no pain, inflammation, intra-articular hemorrhage, or disturbance in the range of motion of the carpal joint.

**Figure 2D**: Pathological findings of the diffuse-type TGCT. (A villous proliferation with a synovial-like lesion in the joint characterizes diffuse-type TGCT. In this area, there was an admixture of synovial-like mononuclear cells, and foam cells, including numerous inflammatory cells).

**Figure 3A**: 3D-CT shows osseous compression at base of the 3rd, 4th, and 5th metacarpal bones and hamate and capitate.

**Figure 3B**: Intraoperative findings shows whitish localized tumor in the 3rd and 4th carpometacarpal joint.

**Figure 3C**: Pathological findings of localized-type TGCT. (Tumor appearance is nodular and well-circumscribed, and partially covered by a fibrous capsule. In fibrous stroma, aggregated mononuclear cells are detected).
wrist. Although diffuse-type TGCT at the knee joint usually includes inflammation and intra-articular hemorrhaging, the carpal joint TGCT displays no symptoms.

Since the TGCT in the hand has not been reported malignant change, we must follow the patients focused in only recurrence. The recurrence rates of TGCT in the hand were reported as 27%, 8.3%, and 13% [6-8]. Our three cases of TGCT showed bony impression around the tumors, which is a risk factor for recurrence [6]. Involvement of the joint capsule is also a high risk factor [8]. The risk of recurrence does not seem to be correlates with any histological parameter other than positive excision margins. Since intra-articular diffuse-type TGCT has a high risk of recurrence, the tumor should be completely excised with meticulous procedure. Out of 18 cases, one recurrence was reported in our institute [1]. The reason for the recurrence was that a surgical-microscope was not used for diffuse-type TGCT [1]. Hence, in order to avoid the recurrence, a surgical-microscope should be used to excise diffuse-type TGCT. However, complete synovectomy is not necessary, diffuse type tumorous lesion should be excised completely. WHO classification showed that only localized TGCT appeared in a hand and finger lesion, and if these lesions are not excised completely, recurrence might occur.

References