Discussion

Pigmented villonodular synovitis (PVNS), a benign, predominantly monolateral, proliferative process of the synovial membrane (Rydholm 1998), was first described by Jaffe in 1941 (Jaffe 1958). Although it has been reported in most joints (Pantazopoulos et al. 1975, Dorwart et al. 1984), few cases have been found in the wrist. The tenosynovial form, with tendon-sheath involvement, is seen oftener in the wrist, but primary intra-articular involvement is rare, as shown by the few cases reported in the literature (Moynagh 1968, Schajowicz and Blumenfeld 1968, Patel and Zinberg 1984, Duriez et al 1986, Valer et al 1997).

In our cases, the diagnosis was considerably delayed. This is often the case with PVNS (Rollo and Wapner 1993), since pain at first is usually mild and radiographs and laboratory tests are usually normal.

Early diagnosis and treatment of PVNS to minimize joint destruction may be of value. Attention should also paid to the lack of relationship between the clinical symptoms and bone lesions, since the patient with the worst bone lesions recovered better than the one with less severe bone lesions.

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Unusual course of the extensor pollicis longus tendon associated with tenosynovitis, presenting as de Quervain disease—a case report

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Submitted 99-11-27. Accepted 00-02-15

A 50-year-old, right-handed woman had had severe right radial wrist pain for 4 months, without trauma. She had never been diagnosed as having inflammatory arthropathy or connective tissue disorders. An orthopedic surgeon diagnosed de Quervain tenosynovitis. Non-steroidal anti-inflammatory drugs and corticosteroid injections were ineffective. After 1 month she underwent surgery, without improvement.

1 month later, she visited our hospital complaining of pain over the dorsum of her right radial wrist. She was unable to extend actively either the interphalangeal (IP) joint or the metacarpophalangeal (MP) joint of her right thumb.

Physical examination revealed localized tenderness and soft tissue swelling radial to Lister's tubercle. She had severe pain on passive extension and flexion of the thumb, and moderate pain at

Figure 1. The duplicated tendon of the EPL (closed white arrow) passed radial to Lister's tubercle (open white arrow) just adjacent to the APL tendon (small black arrow).

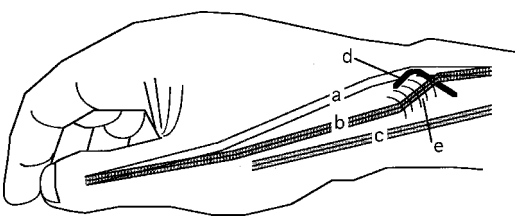

Figure 2. Ilustration showing the intraoperative findings.
a. the normal course of the EPL
b. the course of the duplicated EPL tendon seen in our present case
c. the course of the duplicated APL tendon
d. Lister's tubercle
e. the thickened fibrous sheath containing the EPL

rest. The Finkelstein test was positive. Her grip-strength was 30 kg on the left, while no grip-strength was recorded on the right side. Her erythrocyte sedimentation rate and C reactive protein level were normal. Rheumatoid factor was negative.

During surgery, the first extensor compartment was found to have been completely released. There were 2 tendons in the scar tissue, which were found to be a double tendon for the abductor pollicis longus (APL). We found no extensor pollicis brevis (EPB) tendon around the radial styloid process. Just ulnar to the APL lay a thickened fibrous sheath. We released this, which contained 2 tendons (Figure 1). To more precisely identify the tendons, we made an incision over the dorsum of the MP joint of the thumb. There was only a double tendon passing into the IP joint, which was identical with the double tendon running ulnar to the APL. There was no tendon inserting at the MP joint. Therefore, we concluded that the EPB was absent and those we had observed ulnar to the APL were duplicated EPL tendons.

The EPL tendons were found to pass radial to Lister's tubercle (Figure 2). We also observed the synovial tissues around the tendon, and the point of stenosis. Distal to this point, the tendon was slightly thickened. Its surface was rough and the normal gloss had been lost. We removed the tendon sheath and confirmed that the patient could extend her thumb actively during surgery.

After surgery, her radial wrist pain improved substantially. 6 months later, the grip-strength of her right hand measured 26 kg (nearly equal to that of the left hand). In her left hand, the EPL tendon was palpated, passing ulnar to Lister's tubercle in a normal course. She can now extend her right thumb, but not hyperextend the IP joint. This weakness of extension may be due to the radial passage of the EPL, or the tendon bowstringing after surgery.

Postoperative CT scanning revealed that the groove for the EPL on the distal radius of the involved side was shallow and ill-defined, whereas on the uninvolved side, the structure of the groove was normal. This was clearly demonstrated on 3D reconstruction images.

Discussion

Tenosynovitis of the first extensor compartment, de Quervain disease, is a common condition, and anatomical variations of the first extensor compartment have been well studied (Giles 1960, Jackson et al. 1986, Minamikawa et al. 1991, Tountas and Bergman 1993, Bahm et al. 1995). The reported incidence of absent EPB tendons, as was seen in our case, varies from 0 to 6%. There are, however, only a few reports on the anatomical variations of the third extensor compartment. Yoshida (1990) dissected 832 upper limbs and reported that the EPL was consistently found in all limbs and that its tendon was single in 802 and duplicated in 30 cases. The course of the EPL has been reported to be constant (Kaplan 1981, Valentin 1981, Yoshida 1990), and there are no previously reported instances of an EPL tendon passing

radial to Lister's tubercle. It is possible that 1 or both of the 2 tendons which we assumed to be a double EPL tendon could be an EPB tendon displaced ulnarly by the previous surgical procedure. However, there are 3 reasons we believed them to be duplicated EPL tendons. First, as described before, we explored the dorsum of the MP joint of the thumb, and found no tendon inserting at the MP joint. There was only a double tendon extending into the IP joint, which was identical with the 2 tendons passing ulnar to the APL. Secondly, the sheath in which the 2 tendons lay was left completely intact, with no evidence of a previous surgical procedure. Finally, as shown in Figure 1, the duplicated tendon passed on the fibrous groove, from a more ulnar direction than the usual course of an EPB. This was similar to that of the EPL, though we did not dissect the muscle belly proximally.

Furthermore, as the tendon passed through the third compartment, it suffered from severe tenosynovitis. In the absence of connective tissue disorders, stenosing tenosynovitis of the third compartment is also a rare condition, and only 3 cases have been reported. Mogensen and Mattsson (1980) reported 2 cases in whom the musculotendinous junction of the EPL extended into the compartment. McMahon and Posner (1994) described a case of trigger thumb caused by stenosing tenosynovitis of the EPL. Our case is unique in that the tendon passed radial to Lister's tubercle and the clinical presentation was similar to that of de Quervain disease.

For treatment of tenosynovitis of the EPL, both Mogensen and Mattsson (1980) and McMahon and Posner (1994) recommended surgical release of the compartment only if closed treatment fails. However, Froimson (1993) pointed out the risk of a spontaneous rupture of the EPL tendon due to mechanical attrition between the tendon and Lister's tubercle, and suggested that not only the third extensor compartment should be released urgently, but the tendon should also be transposed radially to Lister's tubercle. Our case did not require transposition of the tendon because it originally passed radial to Lister's tubercle.

The authors thank Fumie Satoh for help in the preparation of this manuscript.

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