**Intratendinous Ganglion in the Extensor Pollicis Longus: A Case Report and
Review of Literature**

Running title: Intratendinous ganglion in the EPL

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Abstract

Ganglion cysts are common benign lesions in the hand and wrist. However, intratendinous ganglion cysts are fairly rare lesions. We present the case of a 73-year-old woman with an intratendinous ganglion cyst occurring in the extensor pollicis longus (EPL) tendon of her right hand. The subcutaneous mass moved according to the movement of the EPL tendon of her right thumb. Magnetic resonance imaging showed a space-occupying lesion in the EPL tendon. Biochemical and hematological examinations ruled out diabetes mellitus, rheumatoid arthritis and other connective tissue diseases. She complained of a motion pain during thumb extension, and she had desired surgery. Intraoperatively, an intratendinous cyst was identified within the tendon substance of the EPL, in which a part of the cyst was herniated into a slit in the tendon substance, just distal to the extensor retinaculum without remarkable proliferative synovial tissue. The EPL tendon was opened longitudinally, and a cystic lesion was enucleated. The pathological findings showed that the cyst wall consisted of fibrous tissue with degeneration, lacking epithelial lining. Postoperative recovery was uneventful. Six months after surgery, the patient had no residual pain and cyst recurrence. The rarity of intratendinous ganglion cysts makes diagnosis and treatment challenging. Since intratendinous ganglion cysts and associated synovitis frequently weaken the structure of the affected tendons, prompt diagnosis and surgical excision are necessary.

Keyword: hand, extensor tendon, extensor pollicis longus, intratendinous ganglion

Introduction

Ganglion cysts are the most common benign soft-tissue tumors of the hand and wrist^{1,2}. The dorsal side of the wrist, which originates from the scapholunate ligament, and the volar side of the wrist, which originates from the scaphotrapezial and radiocarpal joint, are common sites of lesion occurrence. Furthermore, palmar digital ganglia or volar retinacular cysts arise from the flexor tendon sheath. Alternatively, intratendinous ganglion cysts are fairly uncommon lesions^{1,2}. Here, we present a case of an intratendinous ganglion cyst that occurred in the extensor pollicis longus (EPL) in the right hand and review the English literature, which reported these lesions of the hand and wrist.

Case Presentation

A 73-year-old woman was referred to our hospital with the complaint of right wrist pain and a dorsal mass that she noticed before 6 months. The subcutaneous mass moved according to the movement of the EPL tendon of her right thumb (Fig. 1A and 1B). A radiograph showed no abnormality around the hump, including osteoarthritic changes of the carpometacarpal joint. Magnetic resonance imaging (MRI) using T2-weighted fast spin-echo and T1-weighted sequences showed a space-occupying lesion measuring approximately $5 \times 5 \times 7$ mm in the EPL tendon (Fig. 1C and 1D). Biochemical and hematoimmunological examinations ruled out diabetes mellitus, rheumatoid arthritis and other connective tissue diseases. The patient complained of motion pain during thumb extension, and she had desired surgery. Informed consent was obtained from the

patient for surgery. Additionally, the patient gave a written permission allowing us to use her anonymized images in this publication.

Intraoperatively, an intratendinous cyst was identified within the EPL tendon substance, in which a part of the cyst was herniated into a slit of the tendon substance, just distal to the extensor retinaculum (Fig. 2A and 2B). The cyst had no connection to the wrist joint. Additionally, no remarkable synovitis was observed in the surrounding tissue (Fig. 2C). The EPL tendon was opened longitudinally, and a cystic lesion filled with a translucent gelatinous material was enucleated (Fig. 2D). Repair of the split EPL tendon was performed using 4–0 polypropylene non-absorbable sutures.

Cultures were performed to determine the presence of bacteria, acid-fast bacilli, and fungi, which were all negative. The pathological findings of the excised lesions showed no evidence of a specific bursitis such as rheumatoid arthritis. The cyst wall consisted of fibrous tissue with degeneration, lacking epithelial lining. These pathological findings were consistent with a ganglion cyst. Postoperative recovery was uneventful. Six months after surgery, the patient had no residual pain and cyst recurrence.

Discussion

An intratendinous ganglion cyst of the hand is a fairly uncommon lesion. Only 22 ganglion cysts occurring around the tendon of the hands and wrists have been published in 11 case reports in the English literature (Table 1)³⁻¹³. All these cysts have originated from 20 extensors and two flexors. Intratendinous ganglion cysts in extensor tendons are usually small and present an aesthetic concern. On the other hand, intratendinous

ganglion cysts in flexor tendons are difficult to recognize until they grow large enough to be noticed. Because of the difficulties to recognize intratendinous ganglion in the flexor tendon, this lesion in flexor tendon might be the reason why only a few cases have been reported previously.

Although the site of formation of intratendinous ganglion cysts may be similar to that of typical ganglion cysts, they are easy to distinguish by physical examination because they move according to the movement of the tendon. Thus, the differential diagnoses are granulation due to trauma, tenosynovitis of an inflammatory or infectious origin, and neoplasm³⁻⁶. A history of an injury around the lesion indicates a granulation due to trauma. Biochemical and pathological examinations can rule out inflammatory tenosynovitis due to diabetes mellitus, rheumatoid arthritis, gout, or other connective tissue diseases. Neoplasms, including giant cell tumor of the tendon sheath and angioleiomyomas, would be diagnosed using imaging modalities or pathological examinations.

Imaging modalities, including MRI and ultrasonography, are informative for the diagnosis of ganglion cysts; however, the diagnosis of intratendinous ganglion cysts is difficult because most lesions are small³⁻¹³. Radiographs provide meaningful information only if there is a clinical suspicion of an underlying bony abnormality, including osteoarthritis or bone spur. Thus, ultimately, the definitive diagnosis of an intratendinous ganglion cyst may not be possible until surgical exploration is performed⁵. In our case, biochemical and hematoimmunological examinations ruled out diabetes mellitus, rheumatoid arthritis and other connective tissue diseases, and surgical and pathological findings provided a definite diagnosis of the lesion.

Although the causes of intratendinous ganglion cysts are still unclear, most cases were reported to occur around the extensor or flexor retinaculum. In the present case, the intratendinous ganglion cyst was located just distal to the extensor retinaculum³⁻¹³. Thus, chronic mechanical stimulation to the tendon, including friction to the flexor or extensor retinaculum and irritation by a bone spur such as metacarpal bosses, may be the cause of intratendinous ganglion cysts^{5, 10, 13}. Alternatively, from their observation of intraoperative findings, Seidman and Margles have proposed the pathomechanism of these lesions, which caused synovial tissue invasion into the parenchymal tissue of the tendons⁶. However, other cases and our case did not show remarkable tenosynovitis, which indicates a different pathomechanism of ganglion cyst formation from the synovial invasion of parenchymal tissue of the tendons.

Treatment of intratendinous ganglion cysts of the hand remains controversial. Enucleating the tendon to excise the ganglion cyst followed by closure with a running suture would be adequate. Intratendinous ganglion cysts, however, have been reported to increase the risk of spontaneous tendon rupture³⁻¹³. This has been associated with degenerative tendinopathy and longitudinal tearing of the affected tendon^{8, 9}. Since intratendinous ganglion cysts weaken the tendon, the severely damaged tendon is difficult to preserve. Among the 22 tendons reported in the literature, five were resected. All the resected tendons, which includes a flexor and four extensor tendons, were covered with ganglia and severely damaged. If the function of the tendon is nonessential or can be compensated for by tendon transfer or grafting, the affected tendon can be resected^{6, 9, 12, 13}. However, reconstruction of the tendon is a more challenging task with worse functional outcomes than primary ganglion cyst excision and tendon repair. Furthermore, the risk of ganglion cyst recurrence contributes to the management of an

intratendinous lesion. As for the recurrence of intratendinous ganglion cysts, only one patient has been reported⁶. When the tendon is fully occupied and covered with ganglia, complete enucleation of the ganglia would be impossible, and the possibility of recurrence would be higher.

Although patients with a ganglion cyst are usually asymptomatic, prompt diagnosis and surgical enucleation can reduce the risk that the ganglion cyst will continue to grow or recur and lead to rupture of the affected tendon. The biggest issue is to detect intratendinous ganglion. Especially, intratendinous ganglion of flexor tendon is difficult to detect and some of the lesion in flexor might result in spontaneous closed flexor tendon rupture¹⁴ before being diagnosed as intratendinous ganglion in flexor tendon. In any case, prompt diagnosis and surgical excision of intratendinous ganglion cysts are important.

Conclusion

An intratendinous ganglion cyst of the hand is fairly uncommon, and definitive diagnosis is challenging because of the rarity of these lesions. Since intratendinous ganglion cysts and associated synovitis frequently weaken the structure of the affected tendons, prompt diagnosis and surgical excision are required.

158 **Disclosure**

159 The authors declare no conflicts of interest. The authors are responsible for the content
160 and writing of this article.

Figure Legends

Figure 1. Preoperative photographs (A, B) and magnetic resonance imaging (MRI) (C, D). A mass (black arrowhead) that moved according to the movement of the extensor pollicis longus (EPL) tendon was observed (A, B). MRI showed a space-occupying lesion (white) in the EPL on T2-weighted intensity and enhanced T1-weighted images.

Figure 2. Perioperative photographs of the extensor pollicis longus (EPL) tendon (A–D). An intratendinous ganglion cyst was identified within the EPL tendon substance, in which a part of the cyst (white arrow head) was herniated into a slit of the tendon substance (A, B). The cyst had no connection to the wrist joint. Consequently, no remarkable synovitis was observed in the surrounding tissue (C). The EPL tendon was opened longitudinally, and a cystic lesion filled with a translucent gelatinous material was found within the EPL tendon. The cystic lesion was enucleated (D).

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Fig.1

Fig.2

Table. 1 Previously reported intratendinous ganglion cysts of hand and wrist in the English literature (1)

No. of case report	Author	Year	No. of lesion	Age (years)/sex	Side	Occupation	History of injury or cause of the lesion	Symptom at the time of the occurrence	Duration of the wrist pain
1	Lucas GL ¹³	1979	1	13/F	Rt	None	None	Hump of the wrist	1 year
2	Young SC & Freiberg A ³	1985	1	51/M	Rt	Carpenter	Car accident	Hump of the wrist, dull pain	7 months
3	Rayan GM ⁴	1989	1	48/F	*	*	None	Painless swelling, mass of the wrist	2 years
4	Chen WS et al. ⁵	1992	1	42/F	Rt	Housewife	Carpometacarpal boss	Intermittent pain, hump of the wrist	10 years
			1	55/M	Lt	Farmer	Carpometacarpal boss	Dorsal mass of the wrist, soreness	3 years
5	Seidman GD & Margles SW ⁶	1993	2	56/F	Rt	*	Carpometacarpal osteophyte;1 1st carpometacarpal arthritis;1	Hump of the wrist, pain (mild tenderness;5)	10 years
			1	59/F	Lt	*			10–12 years
			1	47/F	Lt	*			>1 year
			1	42/F	Rt	*			Several years
			1	57/F	Rt	*			9 months
			1	56/M	Rt	*			9 months

			2	66/M	Lt	*			2–3 months
6	Ikeda K & Tomita K ⁷	2001	1	53/M	Lt	*	none	Hump of the wrist, inability to extended his left hand completely	*
7	Chew EM ⁸	2010	1	43/F	Lt	Waitress	None	Swelling, hump of the wrist	1 month
8	Satonaka H et al. ⁹	2014	1	45/F	Rt	*	None	Motion pain, painful triggering	3 years
9	Chia DS et al. ¹⁰	2015	1	73/F	Rt	*	None	Painless swelling	1 year
10	Lee HJ et al. ¹¹	2015	1	51/F	Lt	Housewife	None	Hump of the wrist, motion pain	*
			1	51/F	Rt	Housewife	None	Painful fusiform mass of the wrist, motion pain	1 month
11	Senda H et al. ¹²	2017	1	58/*	Rt	*	None	Mass of the wrist, motion pain	30 years
			1	56/*	Rt	*	None	Mass of the wrist, motion pain	*
12	Present case	2020	1	73/F	Rt	Housewife	None	Hump of the wrist, motion pain	8 weeks

F, Female; M, Male; No., number, Lt, Left; Rt, Right

The asterisk indicates no data.

Table. 2 Previously reported intratendinous ganglion of hand and wrist in the English literature (2)

No. of case report	Site of lesion formation	Tumor size	Adjacent lesion	Surgical procedure	Follow-up duration	Postoperative complication	Reconstruction surgery	Recurrence
1	EDBM	*	None	Resection	None	None	-	None
2	EPB	*	None	Enucleation	*	None	-	None
3	EPL	40 mm fusiform nodular	None	Enucleation	12 years	None	-	None
4	EDC (III)	10 × 3 × 3 mm	Carpometacarpal boss	Enucleation	6 months	None	-	None
	Bifurcation of EDC (II, III)	15 × 15 × 10 mm	Carpometacarpal boss	Enucleation	3 years	None	-	None
5	EDC (II), EDC (III)	*	Synovitis	EDC (II); resection, EDC (III); enucleation	12 years, 9 months	None	EDC (II); EIP side-to-side suture	None
	EDC (IV)	*	Synovitis	Enucleation	3 months	None	-	None
	EDC (III)	*	Synovitis	Enucleation	1 year, 10.5 months	None	-	Yes
	APL	*	None	Enucleation	2 years 7.5	None	-	None

					months			
	ECD (III)	*	Synovitis	Enucleation	8 years 4 months	None	-	None
	EDC (II)	*	Synovitis	Enucleation	9 years 9 months	None	-	None
	EDCx2	*	None	Enucleation	8 years 1.5 months	None	-	None
6	EDC (IV)	*	None	Enucleation	2.5 years	None	-	None
7	EPB	20 × 20 mm	EPL; synovial cyst	Enucleation	*	None	-	None
8	EPL	14 × 8 × 7 mm	Stenosing tenosynovitis	Resection	3 months	None	EIP tendon transfer	None
9	FDP (V)	15 × 10 mm	None	Enucleation	1 year	None	-	None
10	EDC (III)	12.6 × 5.5 × 13.2 mm	No synovitis	Enucleation	2 years	None	-	None
	EPL	22 × 2.9 × 6.2 mm	No synovitis	Enucleation	2 months	None	-	None
11	EDC (III)	10 × 20 × 30 mm	Synovitis	Resection of 2/3 tendon	6 months	Subcutaneous tendon rupture	Bridge graft	None
	FDS (IV)	7 × 7 × 20 mm	None	Blocked excision	6 months	None	-	None

				with tendon				
12	EPL	$5 \times 5 \times 7$ mm	Mild synovitis	Enucleation	6 months	None	-	None

APL, abductor pollicis longus; EDBM, extensor digitorum brevis manus; EDC, extensor digitorum communis; EIP, extensor indicis proprius; EPB, extensor pollicis brevis; EPL, extensor pollicis longus; No., number

The asterisk indicates no data.