A high incidence of extensor pollicis brevis insertion into the distal phalanx in rheumatoid arthritis patients who required the surgical reconstruction for thumb boutonnière deformity

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\textbf{ABSTRACT}

\textbf{Objectives:} The aim of the current study was to investigate the pattern of extensor pollicis brevis (EPB) insertion macroscopically and histologically using cadaveric thumbs, and to compare the incidence of different insertions with that of thumb boutonnière deformity in rheumatoid arthritis (RA) patients who required surgical reconstruction.

\textbf{Methods:} We examined 103 thumbs of 58 adult cadavers with no evidence of RA, and reviewed the surgical records of 28 thumbs of 23 RA patients who underwent surgical reconstruction for thumb boutonnière deformity. The incidence of different insertion patterns of the cadaveric thumbs and the RA thumbs were compared using the Fisher’s exact test.

\textbf{Results:} Macroscopic and histologic examination revealed that the insertion patterns of EPB could be divided into three groups: insertion into the base of the proximal phalanx (Type P1), integration of EPB into the dorsal fibrocartilage of the MCP joint (Type P2), and insertion into the distal phalanx (Type D). The incidence of Type D was significantly higher in RA patients with thumb boutonnière deformity (64\%) than that in the non-RA cadavers (29\%; \(P < .05\)).

\textbf{Conclusions:} EPB is inserted into the distal phalanx more frequently in RA patients who require surgery for thumb boutonnière deformity than non-RA cadavers, suggesting an additional possible mechanism of this deformity.

\textbf{Introduction}

Boutonnière deformity of the thumb, also known as the Nalebuff’s type 1, is the most common deformity seen in 50\% to 74\% of thumb deformities associated with rheumatoid arthritis (RA) \cite{1-4}, and is characterized by metacarpophalangeal (MCP) joint flexion and interphalangeal (IP) joint hyperextension. The pathophysiology of boutonnière deformity is understood to start with MCP joint synovitis, which causes extensor pollicis brevis (EPB) to stretch and causes the extensor pollicis longus (EPL) to be displaced ulnarward and palmarward. As a result, dorsal support of the MCP joint is lost, and yolar subluxation of the proximal phalanx develops. The displaced EPL further promotes MCP joint flexion and IP joint hyperextension \cite{5,6}.

Recent studies on the anatomy of the EPB suggest that there are several variations of EPB insertion \cite{7,8}. During clinical practice in surgery of the rheumatoid thumb, we often encountered cases where EPB did not insert into the base of the proximal phalanx, but this variation was not usually described in anatomy textbooks. To date, no study has focused on the relationship between the insertion pattern of EPB and the development of boutonnière deformity in RA patients. We investigated the pattern of EPB insertion and the incidence of different variations using cadaveric thumbs and compared this with that of RA patients with boutonnière deformity who required surgical reconstruction.

\textbf{Patients and methods}

The cadaveric study was conducted at our institute from April 2015 to May 2016. We observed 118 thumbs (59 right and 59 left) of 59 adult Japanese cadavers. We excluded two thumbs of one cadaver with history of RA, and 13 thumbs of 13 cadavers because they were dissected for the anatomy practice before our investigation. Finally, we investigated 103 thumbs (50 right and 53 left) of 58 cadavers (25 males and 33 females). The mean age of the cadavers was 83.4 years (range, 65–98 years). We used cadavers that had been fixed in 10\% neutral buffered formalin. After skin removal and careful dissection of the superficial fascia on the dorsum of each thumb, insertion levels of EPB were easily identified macroscopically, and recorded. Then, the cadaveric thumb samples were removed and cut vertically to confirm the insertion of EPB. After macroscopic observation, the samples for histologic study were fixed in 10\% formaldehyde.
overnight and decalcified in formic acid solution for 2–5 days. They were then embedded in paraffin, cut into 10-μm sections, stained with Masson’s trichrome and observed by light microscopy.

Next, we reviewed the surgical records of 28 thumbs (19 right and nine left) of 23 RA patients (22 females and one male) with thumb boutonnière deformity who underwent surgical reconstruction of the thumb between April 2014 and May 2018. All patients were Japanese, and the mean age of the patients was 61.6 (range, 27–80) years. The surgical procedures included silicone implant arthroplasty in 21 thumbs, and soft tissue reconstruction with synovectomy at the MCP joint in seven thumbs. All the records had descriptions of EPB insertion patterns and indicated whether or not EPB extended to the distal phalanx. We did not obtain pathological samples of the bone–tendon unit of EPB during boutonnière deformity reconstruction.

The incidence of the different insertion patterns of EPB that were identified macroscopically in the cadaveric thumbs and the RA thumbs were compared using Fisher’s exact test. A P-value < .05 was considered statistically significant. The study was approved by the institutional review board (1607-019), and all patients provided written informed consent.

Results

Macroscopically, the EPB insertion pattern was classified into two types: Type P: EPB ended at the level of the MCP joint; and Type D: EPB ended at the level of the IP joint (Figure 1). In Type D, EPB runs distally along EPL, like a conjoint tendon. Direct pulling of EPB resulted in extension of the MCP joint to a different extent in Type P, and in extension of both the MCP joint and the IP joint in Type D.

EPB was present in all the cadaveric thumbs and doubling was found in 4.9% (n = 5). Seventy-one percent (n = 73) of the thumbs were Type P thumbs and 29% (n = 30) were Type D. EPB inserted not only into the distal phalanx but into the base of the proximal phalanx in seven of the thumbs classified as Type D. Macroscopic variance of EPL were not found in the current study, and all the EPL inserted into the distal phalanx.

Histological examination revealed that the Type P thumbs consisted of 52% (n = 54) of Type P1 with EPB firmly attached to the periosteum at the dorsal base of the proximal phalanx of the thumbs, and 18% (n = 19) of Type P2 with EPB attached to the dorsal capsule–fibrocartilage complex in the thumbs (Table 1). In thumbs of Type D, EPB was firmly attached to the periosteum at the dorsal base of the distal phalanx in each case (Figure 2).

All the surgical records described whether EPB extended distally or not because inflammation of RA made it difficult to distinguish Type P1 from Type P2. In the RA patients with thumb boutonnière deformity, 36% (n = 10) of the thumbs were Type P, while 64% (n = 18) of the thumbs were Type D (Figure 3). The incidence of Type D was thus significantly higher (P < .05) in the thumbs of patients with RA and boutonnière deformity than in the cadaveric thumbs.

Histological analysis revealed that the dorsal fibrous complex of the MCP joint of the thumb was mainly composed of three layers: the superficial layer with transverse fibers that are interchangeably referred to as the extensor hood, the intermediate layer consisting of tendon, and the deep layer with the fibrous capsule integrated into the dorsal fibrocartilage in the vertical plane. Although a hood had variable thicknesses and sub-hood space, it lay upon the extensor in all thumbs and there was no insertion of EPB into a hood (Figure 4).

Table 1. Comparison of the insertion pattern of EPB between cadavers and RA patients with thumb boutonnière deformity.

<table>
<thead>
<tr>
<th>Type</th>
<th>Cadavers n = 103</th>
<th>RA patients with boutonnière deformity n = 28</th>
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<tbody>
<tr>
<td>Type P</td>
<td>73 (71 %)</td>
<td>10 (36 %)</td>
</tr>
<tr>
<td>Type P1</td>
<td>54 (52 %)</td>
<td>-</td>
</tr>
<tr>
<td>Type P2</td>
<td>19 (18 %)</td>
<td>-</td>
</tr>
<tr>
<td>Type D</td>
<td>30 (29 %)</td>
<td>18 (64 %)</td>
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</table>

Discussion

Most anatomy textbooks describe EPB as generally originating from the dorsal aspect of the radius and from the interosseous membrane, inserting distally into the base of the proximal phalanx of the thumb. Its action is described as an extension of the MCP joint [9]. However, several cadaveric studies have reported anatomical variations of EPB [8,10–16], and of these, some reports have described insertion of EPB into the extensor hood at the level of the MCP joint [8,15,16]. Shigematsu et al. [7] macroscopically classified EPB insertion into eight types after an anatomical study of cadavers. They reported that EPB was completely inserted into the extensor hoo in 29% (41 out of 144) of hands; completely inserted into the base of the proximal phalanx in 22% (32 out of 144) of hands; partially inserted into the
base of the proximal phalanx as well as into the extensor hood and extended to the base of the distal phalanx in 9% \((n = 13)\) of hands; and inserted into the extensor hood and the base of the distal phalanx in 9% \((n = 13)\) of hands. When we classified these results according to our classification criteria, 70% \((n = 101)\) of the thumbs were Type P thumbs and 30% \((n = 43)\) were Type D, those were very similar to our results. However, our histological findings indicated that the hood is a superficial transverse fiber, which runs over the dorsal surface of the MCP joint and that there were no instances of EPB insertion into the hood.

Several studies have described the anatomy of the dorsal fibrous complex of the MCP joint of the thumb [17–19]. Bade et al. [17], in a microscopic study, reported that the dorsal connective tissue of the thumb forms different layers of collagen lamella as a peritendinous system around EPL and EPB. Hunter-Smith et al. [18] described the dorsal triangular fibrocartilage of the MCP joint filling the dorsal space between the proximal phalanx and the metacarpal head to stabilize and match the congruity of the joint. Joshi et al. [19] described loose connective tissue connections between the dorsal triangular fibrocartilage and the extensor tendon. Our histological findings support these studies, and we found the dorsal fibrous complex of the MCP joint of the thumb was mainly composed of three layers: the extensor hood, the tendon, and the fibrous capsule integrated with the dorsal fibrocartilage in the vertical plane. Thus, in this study, we classified the insertion pattern of EPB into three Types; P1, P2, and D.

In RA patients with thumb boutonnière deformity, 36% \((n = 10)\) of the thumbs were Type P. In Type P2, EPB had a
fibrocartilage, causing thumb boutonnière detachment of EPB from the proximal phalanx with dorsal synovitis not only attenuates EPB but also can easily cause weaker attachment to the proximal phalanx than Type P1, deformity in Type P2 thumbs. EPB inserts into the dorsal capsule Schematic representation of the mechanism of development of boutonnière synovitis cannot easily cause detachment of EPB from the proximal phalanx. (b) Figure 5. (a) Schematic representation of the classical mechanism of the development of boutonnière deformity in Type P1 thumbs: Synovitis of the MCP joint causes detachment of EPB from the proximal phalanx (straight black arrow), and the displaced EPL promotes MCP joint flexion and IP joint hyperextension. However, the EPB inserts firmly into the proximal phalanx, so that synovitis cannot easily cause detachment of EPB from the proximal phalanx. (b) Schematic representation of the mechanism of development of boutonnière deformity in Type P2 thumbs. EPB inserts into the dorsal capsule–fibrocartilage complex. Synovitis not only attenuates the EPB but also easily detaches the dorsal capsule–fibrocartilage complex from the proximal phalanx (straight black arrow). The dislocated EPL leads to thumb boutonnière deformity. (c) Schematic representation of the mechanism of development of boutonnière deformity in Type D thumbs: EPB mainly inserts into the distal phalanx, which cannot extend the MCP joint independently but extends the MCP and IP joints collectively. The dislocated EPL leads to thumb boutonnière deformity.

Weaker attachment to the proximal phalanx than Type P1, with firm attachment of EPB to the proximal phalanx, so synovitis not only attenuates EPB but also can easily cause detachment of EPB from the proximal phalanx with dorsal fibrocartilage, causing thumb boutonnière deformity. Therefore, we speculated that in RA patients with thumb boutonnière deformity with Type P insertion, the incidence of Type P2 EPB might be higher than Type P1 (Figure 5). We assumed that it is congenitally determined whether the EPB is Type P or Type D, and the incidence of different insertion is not affected by inflammation of RA. However, it is difficult to distinguish Type P1 from Type P2 because the inflammation of RA may cause the disruption of the Type P1 insertion (Table 1). In the present study, the rate of Type D was higher in thumb boutonnière deformity compared with the normal population. If a patient with RA has Type D EPB, thumb boutonnière deformity might be easily developed as a result of synovitis and swelling at the MCP joint, because the Type D EPB insertion has no bony attachment around the MCP joint. This may explain the significantly higher incidence of Type D in hands with thumb boutonnière deformity among our RA patients who required surgery.

There are some limitations to this study. First, we did not take any samples from RA patients, because in most cases EPB was ruptured or had lost the insertion to the proximal phalanx. During surgical reconstruction of EPB insertion into the proximal phalanx, it is difficult to obtain samples of the bone–tendon unit. Second, the cadavers and RA patients used in our study were all Japanese. As Jabir et al. [9] observed, the anatomy and variations of EPB seem to show ethnic differences. Third, the small RA patient sample size reduced the power of the study. Finally, this study has a statistical weakness in comparing the result of RA patients with boutonnière deformity who required surgical reconstruction with that of cadavers. We have no data regarding the insertion pattern of RA patients with the deformity who did not require surgery or of RA patients without boutonnière deformity. Further studies on EPB variation in RA cases undergoing surgery or investigations of EPB in RA patients with boutonnière deformity by other imaging techniques will be needed to enhance our understanding of the mechanism of the deformity.

Conflict of interests
None

References