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CASE REPORT

Camptodactyly resulting from anatomical variation of lumbrical muscles: imaging findings

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Abstract
We report three cases of camptodactyly in adolescent patients, presenting with a passive flexion deformity of the fifth finger. Ultrasound findings include aberrant lumbrical insertion and decreased lumbrical size, confirmed with magnetic resonance imaging, and aberrant dynamics. Surgery confirmed these findings in one patient. To the best of our knowledge, these imaging findings have not been reported previously.

Keywords Camptodactyly · Lumbricals · Ultrasound

Introduction

Camptodactyly is derived from a Greek term meaning "bent finger." It is a nontraumatic congenital flexion deformity, occurring in the proximal interphalangeal (PIP) joint in one or several fingers. Most commonly, the fifth finger is affected. The metacarpophalangeal and distal interphalangeal (DIP) joints are usually not affected. The deformity may be flexible or fixed and can be isolated or associated with a number of syndromes. Camptodactyly is usually gradually progressive if not treated [1].

The differential diagnosis includes locked trigger finger, Boutonnière deformity, Dupuytren's disease, and clinodactyly. Clinodactyly describes a nontraumatic flexion deformity with angulation in the radioulnar plane.

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Unlike plain radiographic findings, resulting from long-standing flexion deformity of the PIP joint, imaging findings on ultrasound and MRI have never been reported. The purpose of this article is to present the ultrasound and MRI findings in camptodactyly in a series of three sporadic cases.

Case series

Three adolescent patients presented with an isolated flexion deformity of the fifth finger. There was no previous trauma. Physical examination revealed an inability to actively extend the PIP joint of the little finger (Fig. 1).

Plain radiography confirmed the flexed position of the fifth PIP (Fig. 2).

Ultrasound showed three similar findings in each patient. First, an aberrant insertion of the lumbrical of the fifth finger was seen. In individuals without camptodactyly, the fourth lumbrical muscle belly follows a curved course, deep and distal to the metacarpal heads (Fig. 3a). Its distal tendinous extension partly blends with the radial side of the extensor expansion of the fifth finger. In our series, a superficial distal course of the muscle belly was seen, inserting on the radial side of the flexor digitorum superficialis (FDS) of the affected fifth finger, at the level of the fifth metacarpophalangeal joint (Fig. 3b).

Second, a decreased lumbrical muscle size was noted on the affected side (Fig. 4a, b).

Finally, ultrasound showed altered dynamics at the involved finger. Passive motion of the superficial flexor tendon

Fig. 1 A 13-year-old girl with camptodactyly: clinical picture

(passive flexion at the PIP with maintained extension of the DIP) caused motion of the attached lumbrical muscle, whereas passive motion of the deep flexor tendon (passive flexion at the DIP) did not affect motion of the lumbrical muscle (Supplementary Fig. 1). Passive motion of the superficial flexor tendon did not cause coordinated lumbrical muscle motion in the non-affected fingers (Supplementary Fig. 2).

Magnetic resonance imaging was performed on 3-Tesla MRI scanner (Discovery; General Electric Medical Systems, Milwaukee, WI, USA) with dedicated coils and protocols in three planes, including axial and coronal fat-suppressed (FS) T1-weighted images (WIs), coronal FS T2-WI, and axial, coronal, and sagittal proton-density-weightedfast spin echo (FSE) fat-suppressed MRI. The slice thickness was 2 mm, except for the sagittal proton-density-weighted FSE fat-

Fig. 2 The flexed position of the fifth proximal interphalangeal (PIP) joint is illustrated on plain radiography in a 16-year-old boy with camptodactyly

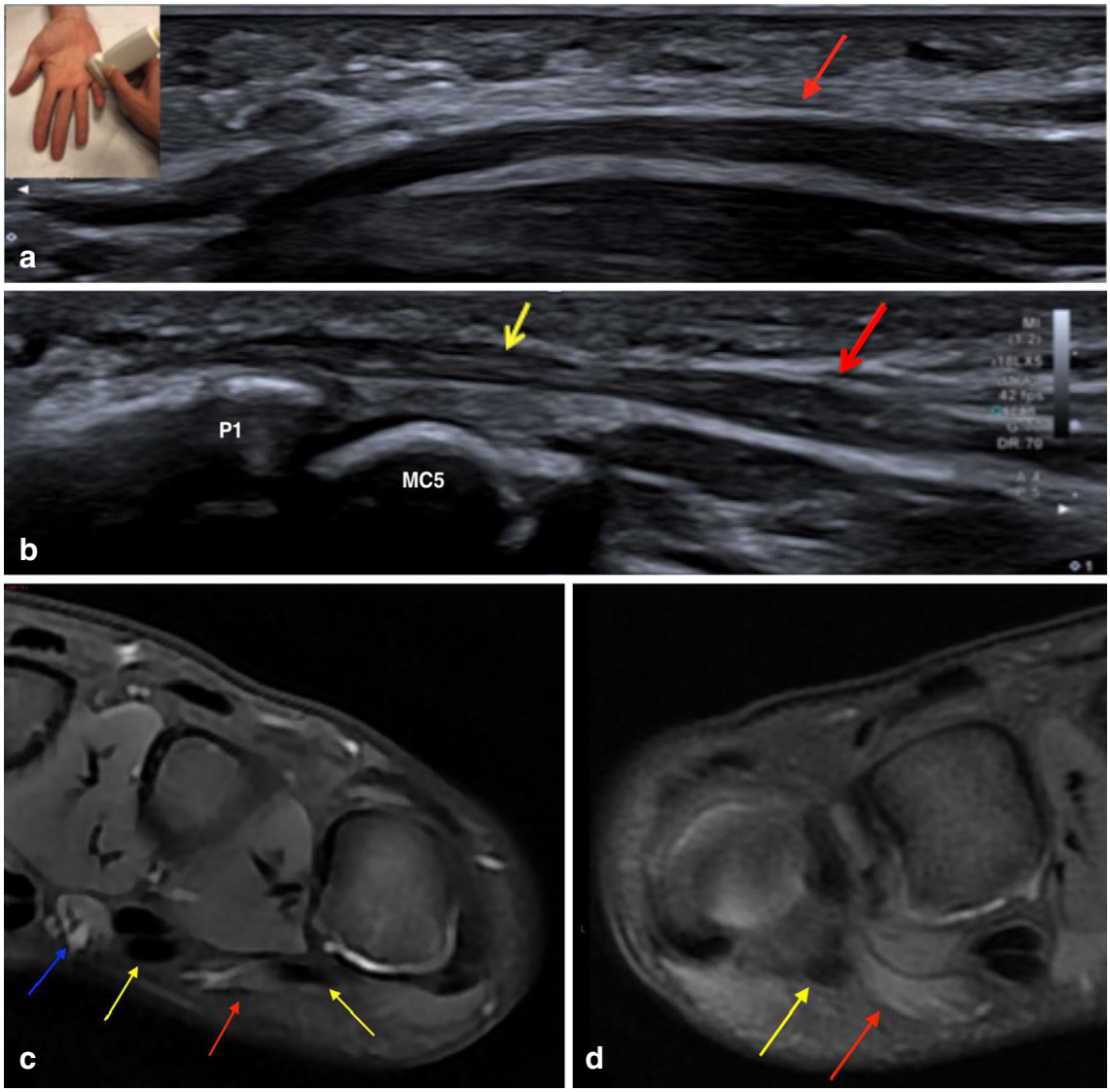

Fig. 3 **a**, **b** Aberrant lumbrical insertion: a 13-year-old girl with camptodactyly, oblique longitudinal ultrasound of the fifth finger. **a** On the unaffected side, the fourth lumbrical muscle (*red arrow*) distally curves dorsally where its distal tendinous extension blends with the radial extensor apparatus at the level of the proximal phalanx (not seen). **b** On the affected side a straight and superficial course of the fourth lumbrical muscle is seen, and the muscle inserts on the superficial flexor tendon (*yellow arrow*). *MC5* fifth metacarpal, *P1* proximal phalanx of the fifth finger. **c** Axial proton-density-weighted fast spin echo (FSE) fat-suppressed MRI of the affected hand at the level of the fifth metacarpal head shows a superficial position of the fourth lumbrical (*red arrow*), closely related to and partially covering the superficial flexor tendon (*yellow arrow*). The fourth lumbrical has a large contact surface with the adjacent flexor tendon. The third lumbrical (*blue arrow*) is more deeply positioned. **d** Axial proton-density-weighted FSE fat-suppressed MRI of the unaffected side at the same level as **c** shows the fourth lumbrical muscle (*red arrow*) coursing deep between the fourth and fifth metacarpals toward the extensor expansion (not seen)

saturated MRI, which were 1 mm thick. MRI confirmed the aberrant course, insertion (Fig. 3c, d), and size loss (Fig. 4c, d) of the fourth lumbrical muscle in all patients.

One patient was treated surgically. Surgery confirmed a hypotrophic lumbrical muscle inserting on the superficial flexor tendon of the fifth finger. Release of the lumbrical muscle and the checkrein ligaments was performed. Full extension was achieved and the joint was locked in extension with the use of a Kirschner wire through the PIP joint. Despite the uneventful immediate postoperative result, the long-term cosmetic result was poor. Two patients were treated conservatively with serial casting, which resulted in an unchanged and a slightly improved cosmetic result respectively.

Discussion

Camptodactyly is a congenital flexion deformity, occurring either as a sporadic anomaly, inherited as an autosomal dominant trait with incomplete penetrance or as part of an often ill-defined malformation spectrum such as orofaciodigital syndrome, trisomy 13–15, and Jacob–Downey syndrome [2].

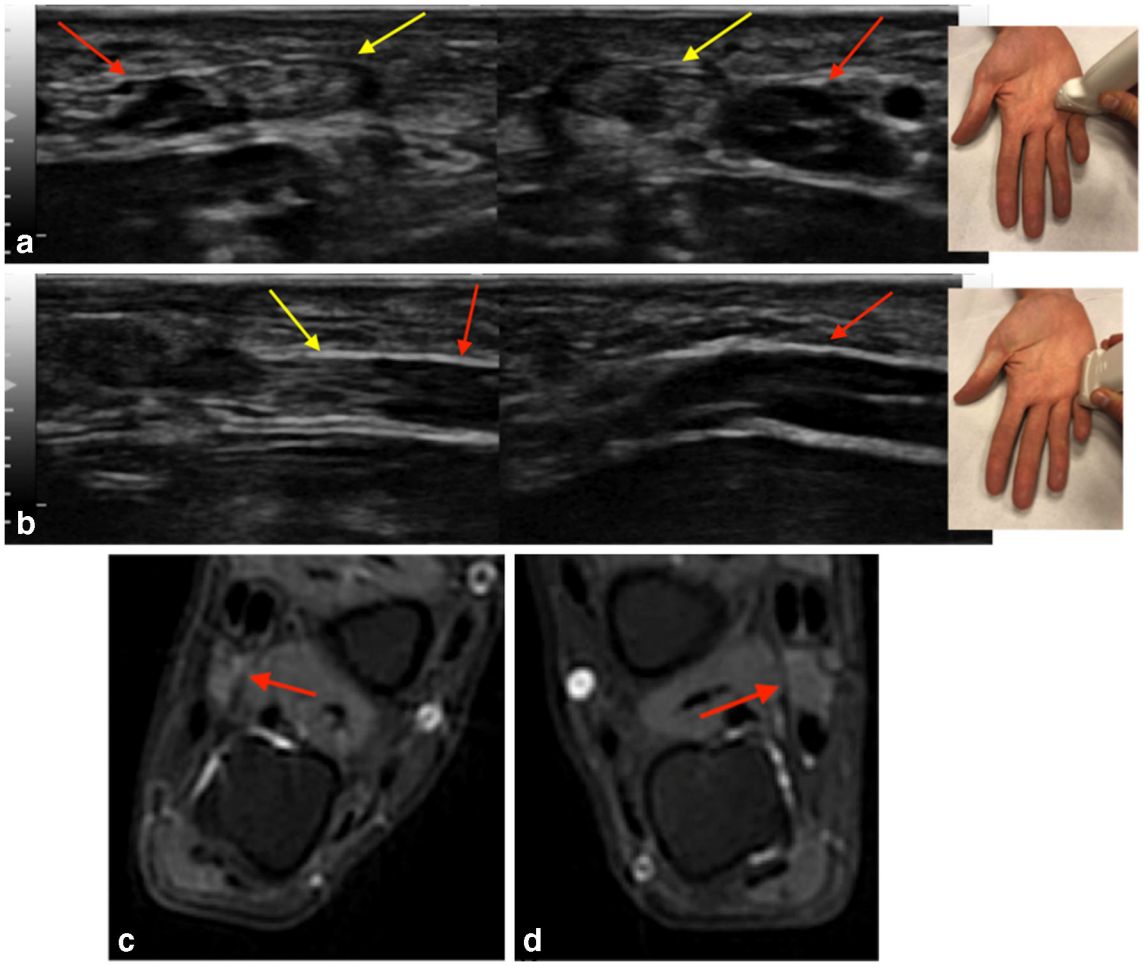

Fig. 4 Decreased size of the fourth lumbrical muscle. A 16-year-old boy with camptodactyly. **a** Axial and **b** longitudinal ultrasound shows an important decrease in muscle size of the fourth lumbrical (*red arrows*) on the affected side (images on the left). Note the adjacent flexor tendon (*yellow arrows*). On the affected side, the superficial flexor tendon lies close to and has a large contact surface with the fourth lumbrical. Axial proton-density-weighted fat-suppressed MRI shows the small size of the fourth lumbrical muscle (*red arrow*) on **c** the affected side compared with **d** the non-affected side

It usually involves the PIP joint of the fifth finger, but other digits may be affected as well, with a decreasing frequency toward the radial side of the hand. The disorder is usually gradually progressive. Distal interphalangeal joint or metacarpophalangeal joint involvement suggests a post-traumatic cause rather than true camptodactyly [1]. The prevalence of camptodactyly is less than 1% [2]. It may be unilateral (33%) or bilateral (66%) [1]. It may be diagnosed at birth or during (pre)adolescence and can be divided into a reducible or flexible and a fixed or irreducible type. Depending on the age at onset, it can also be classified into three subtypes (infantile, adolescent, and neonatal/syndromic) [3].

Camptodactyly is very rarely associated with significant functional deficits. It is painless and does not cause motor or sensory deficits [1].

The precise pathogenesis of camptodactyly is still a matter of debate and has been attributed to many factors [1]. These include contractures of the skin or fascia, or abnormalities of the volar plate or collateral ligaments. Anomalies in the collateral and other restraining ligaments have been described as a cause of camptodactyly. The flexor digitorum superficialis can be contracted, underdeveloped, or devoid of normal function.

The lumbrical muscle may be of aberrant origin, from the transverse carpal ligament and/or ring flexor tendon. Aberrant insertion on the superficial flexor tendon, metacarpophalangeal joint, ring finger extensor apparatus, or in the lumbrical canal or absence of lumbrical muscle have also been described in the surgical literature [1, 2].

The lumbrical muscle is thought to be the principal cause of camptodactyly. In a series of 74 consecutive operations for camptodactyly, McFarlane et al. found that the fourth lumbrical muscle to the small finger had an anomalous insertion in all cases [4], although other authors have suggested that other factors might cause the deformity [5].

According to anatomical textbooks, the lumbrical muscles usually arise in the palm from the deep flexor tendons and insert on the radial sides of the extensor expansion of the second to the fifth fingers, distal to the metacarpophalangeal joint level (Fig. 5) [6, 7].

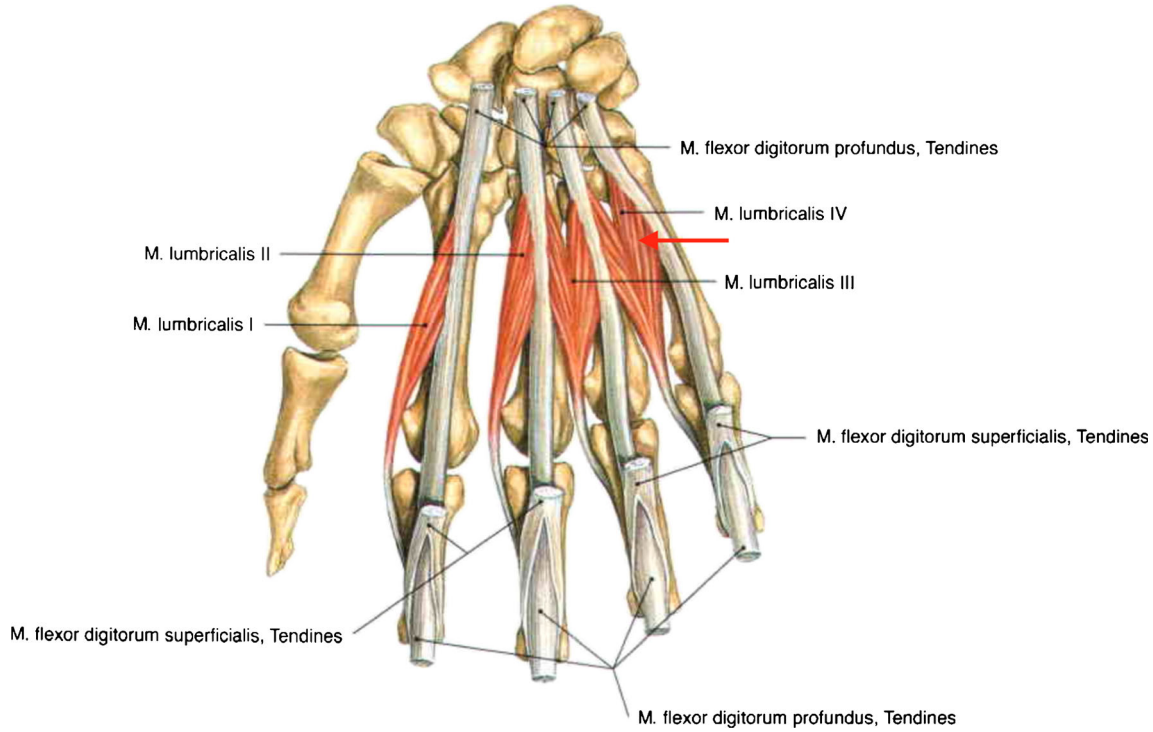

Fig. 5 Normal lumbrical anatomy. Note the bipennate fourth lumbrical, inserting on the extensor expansion of the fifth finger (*red arrow*). Image from Sobotta Atlas of Human Anatomy, 14th edn [6]. Reprinted with permission (Supplementary Fig. 3)

Most authors believe that the lumbricals act as a deflexor of the PIP joint, contributing to extension of the PIP joint. Their role in metacarpophalangeal flexion is rather limited [8].

Until present, only radiographic features of long-standing cases of camptodactyly have been described, showing changes of the PIP joint secondary to prolonged flexion deformity. No soft-tissue imaging findings have been described. In our small series, ultrasound findings in camptodactyly were threefold: aberrant lumbrical insertion, size loss, and aberrant dynamics.

In our opinion, ultrasound and plain radiographs are the imaging modalities of choice to evaluate patients with camptodactyly. Plain films are used to evaluate the degree of flexion. Ultrasound and MRI may directly visualize the aberrant lumbrical insertion and decreased size. The advantage of ultrasound is the ability for dynamic evaluation.

Camptodactyly is difficult to treat, and inconsistent results have been reported [1]. Conservative treatment is favored in most cases and is best for PIP contracture less than 30°. Surgical treatment is preferred in progressive/severe deformity or in the case of failure of conservative treatment. However, as demonstrated in our three cases, the outcome of both conservative and surgical treatment remains relatively poor. This is in line with the literature on the topic. In a case series of 57 patients (79 fingers), Siegert et al. compared the outcome of surgery and conservative treatment. Of the surgically treated patients who were available for follow-up 6 years after treatment, 66% of results were poor, 16% were fair, 18% were good, and there were no excellent results. In the conservative group, there were 15% poor, 20% fair, 65% good, and no excellent results. [9]

Although this report of abnormal ultrasound and MRI findings in camptodactyly in a small series may not have a clear direct impact on the choice of the treatment option, the ultimate prognosis and outcome of treatment, we believe that further documentation of abnormal imaging findings will be helpful in unraveling the complex anatomy and pathogenesis of camptodactyly. For definite conclusions, further studies in a larger series of patients are mandatory.

Compliance with ethical standards

Conflicts of interest The authors declare that they have no conflicts of interest.

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