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CASE REPORTS

DUPUYTREN'S DISEASE IN AN ADOLESCENT – CASE REPORT

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Dupuytren's disease is typically observed in adults, in children and adolescents being rarely diagnosed. Only several cases were reported in children under the age of 10 years, including two infants under one year. There were no such reports in Polish literature. We presented a case of Dupuytren's disease in a 17 year-old adolescent, in whom the first signs of the disease were observed in early childhood.
Key words: Dupuytren's disease, children, digital contracture

Dupuytren's disease is a proliferative disorder of the palmar fascia consisting in the creation of myofibroblasts from fibroblasts, their excessive proliferation, the formation of nodules and fascicles within the palmar fascia, as well as the future development of digital contractures, most often of the fourth and little fingers. Dupuytren's myofibroblasts produce a particular type III collagen, characteristic of the disease. Factors favoring the development of the disease include alcohol abuse, smoking, anti-epileptic drugs, diabetes mellitus, and liver cirrhosis, although not all have been confirmed (1). Dupuytren's contracture incidence varies geographically and racially with the highest incidence in northern Europe and the Caucasian population, as compared to Asians and Africans (1, 2). A family history has also been observed, which might be evidence of its genetic basis.

Dupuytren's disease is typically observed in the adult population, while in children and adolescents it is rarely diagnosed. Only several cases were reported in children under the age of 10 years, including two infants under one year (3-8). Thus far, there were no such reports in Polish literature, considering adolescents. The study presented a case of Dupuytren's disease in a 17 year-old adolescent, in whom the first signs of the disease were observed in early childhood.

CASE REPORT

A seventeen year-old patient was referred to the department, due to contracture of the little finger of the right (dominant) hand. Based on data obtained from the mother and patient the thickening of the fifth finger was observed at the age of 6 years. During childhood and early adolescence the protuberance remained unchanged, when at the age of 14 years it began to grow leading towards little finger contracture. Both the patient and mother denied injury as a possible cause of the contracture.

On admission, the physical examination revealed a well-palpable band of cohesive tissue on the proximal and middle phalanx of the little finger (right hand), located on the side of the arm. The fascicle adhered closely to the skin. The fifth finger was flexed at an angle of 90^{o} in the proximal interphalangeal joint, while the distal interphalangeal and metacarpophalangeal joints were not subject to contracture (fig. 1). The ultrasound examination showed the presence of a fibrous fascicle within the proxi-

Fig. 1. Fifth finger contracture in a patient on admission for surgery

mal phalanx of the fifth finger, located under the subcutaneous tissue and skin. The image was typical of Dupuytren's disease.

The patient was qualified for surgery, which was performed under spinal anesthesia (brachial plexus block) and ischemia. Intraoperatively, a fibrous fascicle was observed leading to the contracture of the fifth finger, being affixed to the distal fibrous sheath of the flexor tendons, and proximally protruding from the palmar fascia (fig. 2). The fascicle was excised, which allowed digital extension. The postoperative course was uneventful, the wound healed by first intention. The histopathological examination was typical of Dupuytren's disease. Follow-up performed one year after surgery showed a slight contracture, which was associated with scar reconstruction, and not disease recurrence (fig. 3).

DISCUSSION

Dupuytren's disease is moderately common amongst adult Europeans, while in children and adolescents it is rarely diagnosed. Considering the above-mentioned patient the first manifestation of the disease was observed at the age of 6 years in the form of a perceptible protrusion of the fifth finger, remaining unchanged for the next 8 years. At the age of fourteen the lesion began to grow leading to digital contracture, typical of Dupuytren's disease. One may assume that hormonal changes associated with puberty led to the progression of the previously dormant disease.

Differential diagnosis of fifth finger contracture includes a malformation termed camptodactyly, although its clinical manifestation is slightly different (9). In case of camptodactyly the little finger is subject to contracture in the proximal interphalangeal joint, while there is no palpable fascicle in the proximal phalanx. The contracture is usually diagnosed during childhood, although may be present since birth. Additionally, camptodactyly usually occurs on both hands. Our patient was consulted at the age of 14 years and diagnosed with discreet camptodactyly, requiring no treatment.

Literature data presented only isolated cases of Dupuytren's disease in children, as well as series numbering several patients. The largest study group comprised 8 patients, and the youngest patients were aged between 6 and 10 months (7, 8). Mikkelsen et al., in their study of the natural history of Dupuytren's disease, considering 900 cases, observed 3 (0.3%) patients, who declared that initial symptoms occurred at the age under 10 years, and 12 patients that noticed first symptoms at the age of 10-20 years (10). Other Authors analysed a group of 900 patients with soft tissue nodules of the hand demonstrating histological lesions typical of Dupuytren's disease in two male patients, aged 8 and 11 years. Treatment of Dupuytren's disease in children is the same as in adults, and consists in the excision of the

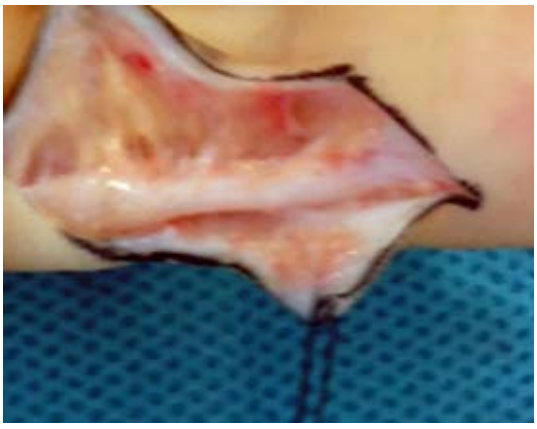

Fig. 2. Intraoperative image of the fibrous fascicle located on the proximal phalanx and metacarpus

Fig. 3. Operated finger upon examination one year after surgery

changed fragment of the palmar fascia. In adults, recurrence is often observed (up to 50% considering a 10-year perspective), similarly, in case of children after surgery (6).

The presented study case is the first in Polish literature describing the above-mentioned pathology, and thus, we considered it justified to present to the readers.

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