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RESEARCH LETTER

Preiser's disease in a five-fingered hand

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ABSTRACT

Preiser's disease, also known as idiopathic avascular necrosis of the scaphoid, and five-fingered hand are rare hand conditions. In this report, we present a case of a 25 year old female patient who had avascular necrosis of the scaphoid and five-fingered hand.

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Five fingered hand; Grip strength; Midcarpal arthrodesis; Preiser disease

Introduction

Preiser's disease is characterized by idiopathic necrosis of the proximal pole or the whole scaphoid[1]. Related etiological factors are collagen vascular disease, steroid therapy, repetitive trauma, hypoplasia of the scaphoid, and idiopathic causes. Radial-sided wrist pain, sensitivity on scaphoid, and decreased wrist motion are among other significant findings.

Five-fingered hand is a rare and different entity from triphalangeal thumb and thumb in the plane of the hand[2,3]. The most radial finger is similar to the four ulnar fingers, with a long metacarpus having distally placed epiphysis like the others. Thenar muscles are absent and the most radial finger is in the same plane with the other fingers.

An association between congenital hypoplasia or absence of the scaphoid and retardation in the development of the thumb and thenar muscles was assumed to be related[4]. Here, we present a case with five-fingered hand and Preiser's disease, which may be an example supporting this association.

Case report

A 25 year old female patient, who was employed as a secretary, presented with a two-year history of pain in the left wrist. There had been no history of previous trauma at any point. The right hand was dominant and normal. Her left thumb consisted of three phalanges resembling the other four fingers, and the thenar muscles were atrophic (Figure 1). The thumb had a normal range of motion. However, she had tenderness in the anatomical snuffbox.

Radiographs of the left wrist and hand demonstrated a fragmented scaphoid and five-fingered hand. Marked deformation and avascular necrosis particularly in the proximal scaphoid were seen in the MRI (Figures 2 and 3). As the patient had wrist pain and decreased wrist mobility, a scaphoidectomy and capitohamate arthrodesis were performed through a dorsal midline incision (Figure 4). The capitate–hamate limited wrist arthrodesis was chosen because we wanted to protect carpal height. This type of arthrodesis is not an obstacle after conducting a proximal row carpectomy. Her left thumb did not need any surgical intervention. The histopathologic examination of the scaphoidectomy material was consistent with osteonecrosis.

The patient wore a short arm cast until complete fusion. In the second postoperative month, her left hand grip strength was 5 kg, visual analogue score (VAS) was 6, QuickDASH score was 45.5, wrist flexion was 20°, and wrist extension was 45°. In the fourteenth postoperative month, her left hand grip was 10 kg, VAS was 2, QuickDASH score was 36.4, and wrist flexion and extension were 40° and 50° respectively. Her QuickDASH was moderately high, but she reported satisfaction with the procedure.

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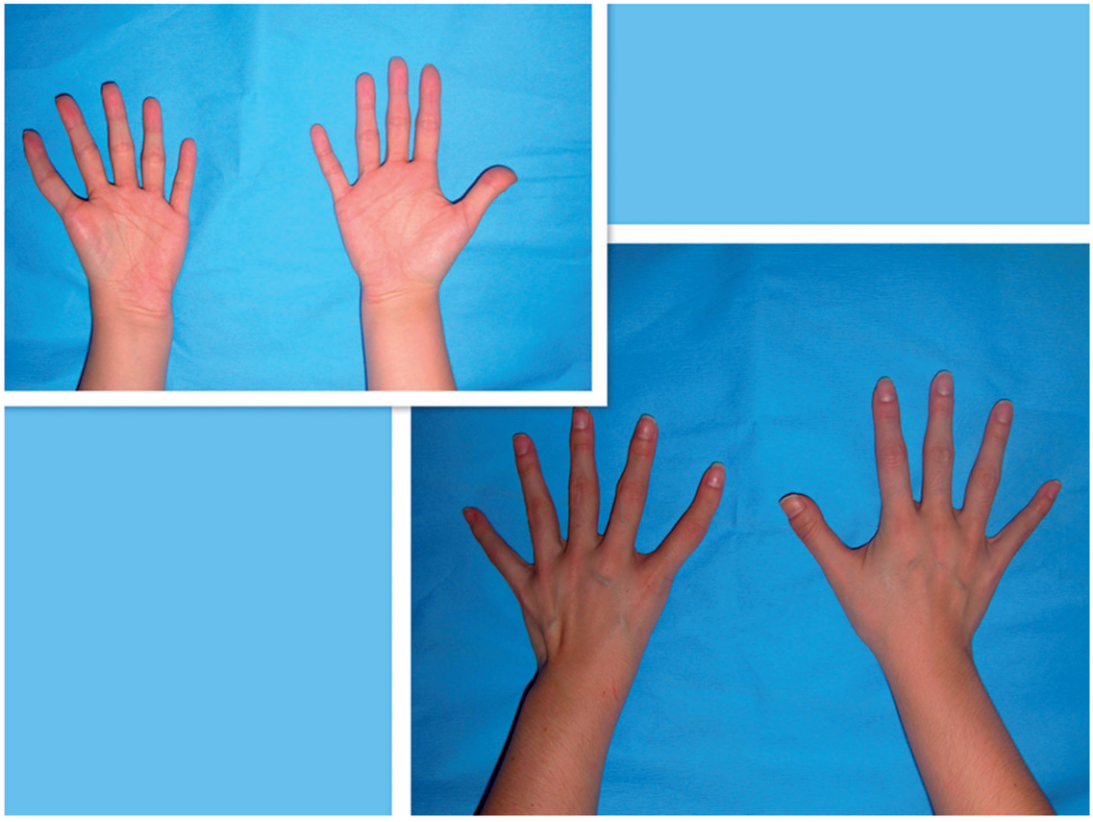

Figure 1. Five-fingered left hand and the normal right hand.

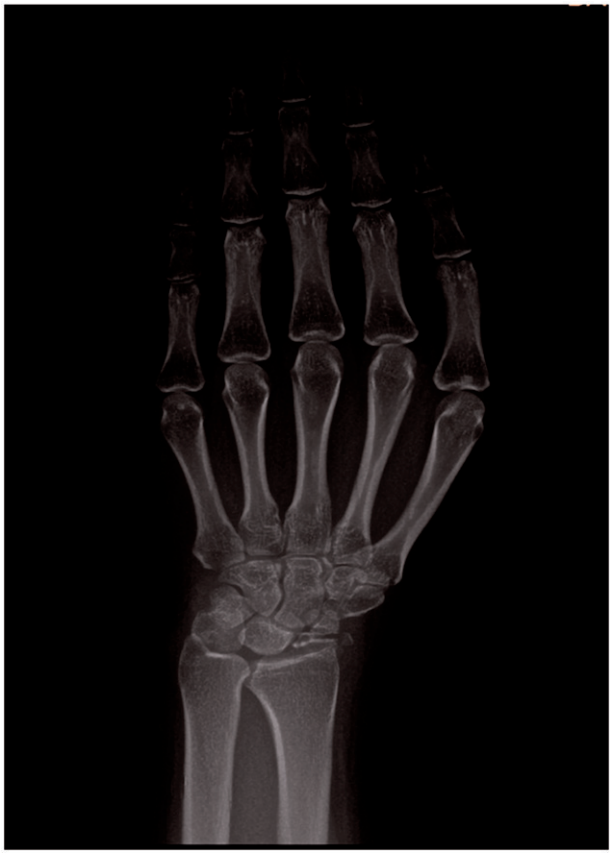

Figure 2. X-ray of the left hand with the thumb showing the finger characteristics and the fragmentation of scaphoid.

Discussion

Five-fingered hand is a rare condition, and only about 40 cases have been reported in the literature[2,3]. In a five-fingered hand, the scaphoid is generally not present, or it is hypoplastic[4]. In our patient, the scaphoid was present, but avascular.

The etiology of Preiser's disease is not fully understood; however, repetitive trauma, corticosteroids, chemotherapy, connective tissue disease, and alcohol excess are believed to be the causes. Late radiographic changes include cystic degeneration and fragmentation/fracture. Based on the reported cases, women and the dominant hand are affected by this condition more frequently, and the mean age is 42 years (9–76 years)[3].

Upton considered the five-fingered hand as a form of thumb hypoplasia[4,5]. This anomaly is generally associated with hypoplastic scaphoid[6].

Parkinson *et al.* reported two cases of scaphoid abnormalities in patients with congenital radial ray deficiencies and suggested an association between them[1]. In our case, a known etiologic cause could not be determined. However, because of the five-fingered hand, blood supply to the scaphoid might have been impaired and thus led to Preiser's disease. Scaphoidectomy and capitohamate arthrodesis were performed in order to allow for a proximal row carpectomy (PRC) in the future.

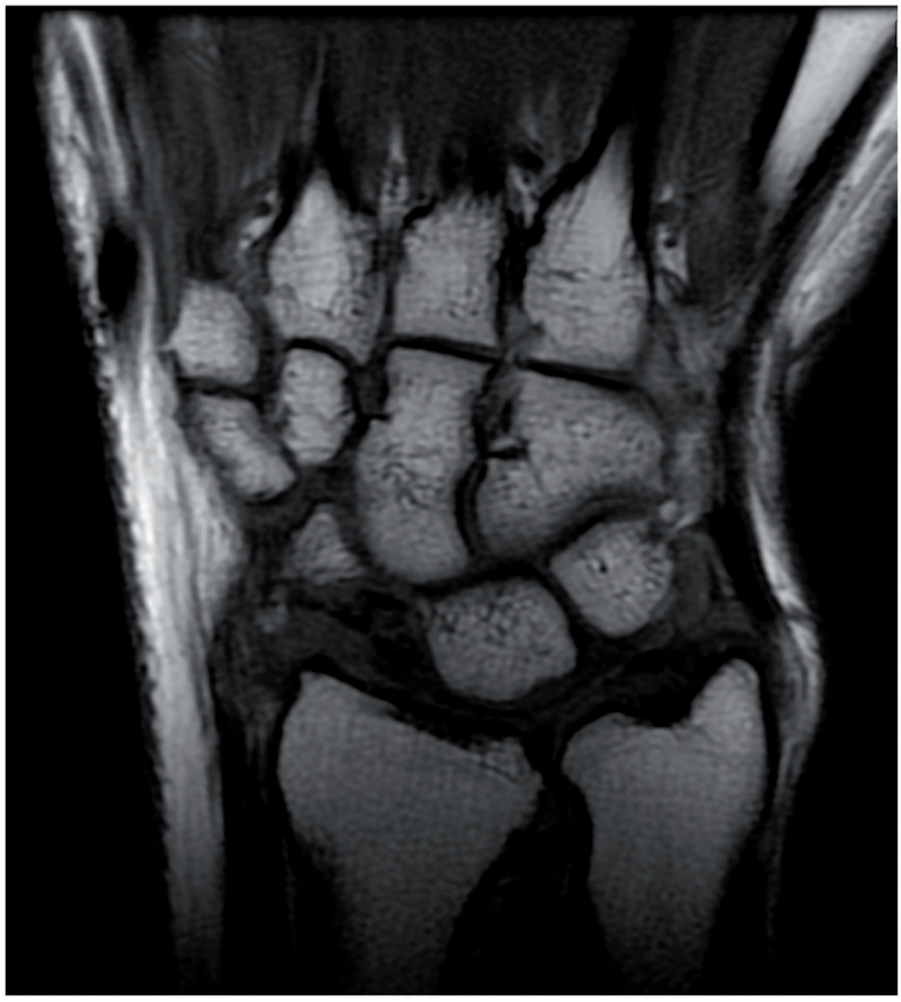

Figure 3. Coronal MRI image of the left hand.

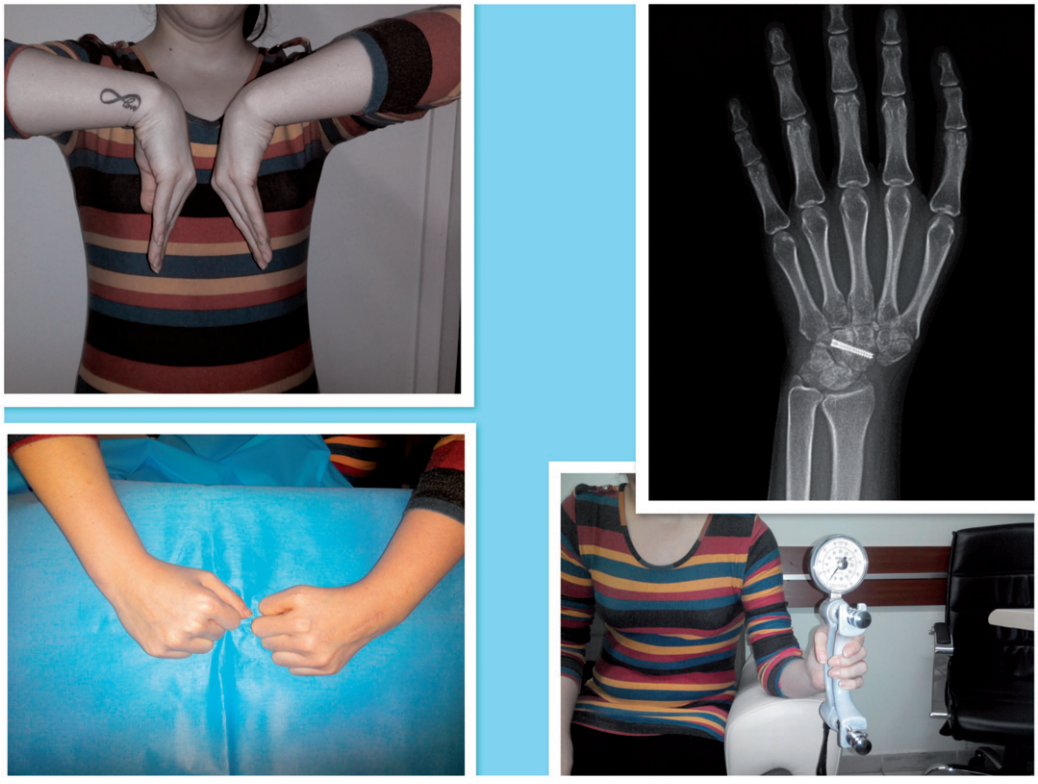

Figure 4. Postoperative image of the left hand.

Conclusion

The five-fingered hand is a rare hand condition. Problems of the scaphoid should be considered in the presence of pain around the radial styloid process. When accompanied by Preiser's disease, scaphoidectomy and capitohamate arthrodesis can be performed in order to allow for a chance of PRC in the future.

Transparency

Declaration of funding

This report was not funded.

Authors contributions: M.D. and A.S. carried out the study and drafted the manuscript. B.K., U.G., S.A., and K.O. participated in the design of the study, while U.G. and S.A. participated in carrying out the study and reviewing the references. All authors read and approved the final manuscript.

Declaration of financial/other relationships

M.D., A.S., B.K., U.G., S.A., and K.O. have disclosed that they have no significant relationships with or financial interests in any commercial companies related to this study or article.

CMRO peer reviewers on this manuscript have no relevant financial or other relationships to disclose.

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