

COEXISTENCE OF SHORT FIFTH METACARPALS IN A FEMALE PATIENT WITH ANKYLOSING SPONDYLITIS ASSOCIATED WITH COMPLEX REGIONAL PAIN SYNDROME TYPE-I

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Short digits, metatarsal and metacarpal bones may occur congenitally as an isolated case or as a manifestation of several genetic and acquired disorders resulting from a premature closure of the physeal plate¹. The fourth and fifth metacarpals tend to be the most affected bones in the hand². Symmetrical short fifth metacarpals have been described usually as case presentations in a few reports in the literature^{3,4}. We herein described a female patient with ankylosing spondylitis (AS) in association with complex regional pain syndrome (CRPS) type I in the right hand and congenitally bilateral short fifth metacarpals.

Case Report

A 59-year-old woman was admitted to our hospital because of low back pain for 20 years. Low back pain was aggravated by inactivity and associated with lumbar morning stiffness occasionally lasting over than 1 hour. She was also complaining of swelling with excessive pain to light touch on her right hand. She declared that she had bilateral short fifth finger since birth. On musculoskeletal examination, she had typical ankylosing spondylitis (AS) appearance with kyphosis and an excessive lumbar flexion posture. She had limited range of motion of the cervical and lumbar spine in all directions. Modified lumbar Schober test was 0,5 cm. Maximum expansion of chest was found to be 2 cm, measured at the level of the fourth intercostal space during maximum inspiration. Her right hand was very painful to palpation and the skin of her right hand was glossy, edematous, unwrinkled and reddish (Fi-

gure 1). Range of motion of right hand was limited. The results of routine laboratory assessments were normal, and HLA-B27 was positive. Radiological examination revealed syndesmophytes at several thoraco-lumbar vertebrae, unilateral grade III sacroilitis, and short fifth metacarpal bones with no local patchy osteoporosis (Figures 1, 2). Three phase bone scintigraphy was performed after i.v. injection of 20 mCi Tc-99m methylene diphosphonate [MDP]. On the flow and blood pool images radiopharmaceutical accumulation was increased on the right wrist and hand related to hyperemia. On delayed phase bone imaging diffusely increased Tc-99m MDP uptake was also observed on bony structures (Figure 3). Scintigraphic evaluation was compatible with CRPS type 1. AS and CRPS type-I



Figure 1. Patient's hands with the right hand appearing edematous (A) and bilateral short fifth metacarpals on the anteroposterior hand radiograph (B)

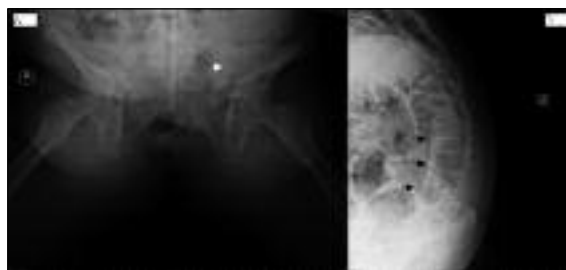


Figure 2. Anteroposterior lumbosacral radiograph showing grade III sacroilitis (A) and lateral thoraco-lumbar radiograph demonstrating syndesmophytes (B)

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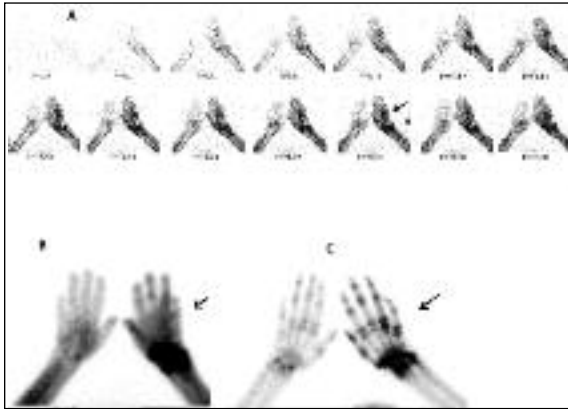


Figure 2. Three phase bone scintigraphy revealed increased Tc-99m MDP uptake (arrows) on the flow (A), blood pool (B) and delayed phase bone imaging (C) on the right hand and wrist

was diagnosed on the basis of clinical and radiological findings. Pregabalin 300 mg and salmon calcitonin 100 IU daily, in association with sulphasalazine 2g/ and a nonsteroidal anti-inflammatory drug therapy was initiated. Symptoms were resolved in one month.

CRPS type I is characterized by sensory pain and vasomotor disturbances, trophic changes and impaired motor function that frequently affects the extremities with hand, wrist, knee, ankle and foot being the commonest affected⁵. The occurrence of CRPS type I in combination with inflammatory disease has been reported rarely⁶. We herein report a CRPS type I associated with ankylosing spondylitis and coexisting with short fifth metacarpals in the same individual. We believe that inflammatory factors play a major role in the development of CRPS type I.

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References

1. Aarntzen EH, Barrera P. Short digit. What's up? *Arthritis Rheum* 2007; 57:1568-1571.
2. Armour CM, Bulmann DE, Hunter AG. Clinical and radiological assessment of a family with mild brachydactyly type A1: the usefulness of metacarpophalangeal profiles. *J Med Genet* 2000; 37: 292-296.
3. Patel V, Alban Davies H. Insulin resistance type A and short 5th metacarpals. *Diabetic Med* 2003; 20: 500-504.
4. Hyari M, Hamamy H, Barham M, Al-Hadidy A, Ajlouni K. Familial short fifth metacarpals and insulin resistance. *Skeletal Radiol* 2006; 35: 702-705.
5. Kumar R, Aggarwal A, Faridi MM. Complex regional pain syndrome type I and scurvy. *Indian Pediatrics* 2009; 46: 529-531.
6. Conca W, Laubenberger J, Krause T, Blumberg H, Peter HH. Post traumatic reflex sympathetic dystrophy antecedent inverse psoriasis and psoriatic arthritis. *J Rheumatol* 1995; 22: 783-785.