Primary Hypertrophic Osteoarthropathy with Gastric Hypertrophy

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Primary hypertrophic osteoarthropathy (HOA) is a rare congenital disease that is not well recognized. Gastric hypertrophy will be infrequently involved; only 3 case reports describe this scarce manifestation in the English-language literature[1,2,3].

A 19-year-old Chinese male presented with a 4-year history of thickened skin and joint pain and swelling. Marked sweating and intermittent low fever were also noted. He had been treated with methotrexate for 8 months and Chinese traditional medicine for 9 months, with no improvement. Examination revealed distinct grooves on his forehead and thickened scalp (Figure 1A). His wrists and hand joints were slightly swollen, and skin distal to wrists was darkened (Figure 1B). Symmetrical nonpitting edema of knees, ankles, and feet was observed (Figure 1C). Laboratory tests revealed elevated inflammatory markers, normal growth factor level, and normal immunological markers. Malignancy was excluded after examinations. Bone radiographs revealed thickened cortical plate and periosteal new bone formation in bilateral tibio-fibula, femur, radio-ulna and long bones of hands and feet (Figure 1D). Barium radiography showed hypertrophic mucosa of gastric body and fundus, with normal esophagus, small intestine, and ileocecal junction (Figure 2A). Gastroscopy also revealed gyrus-like hypertrophic and rough mucosa at gastric body and fundus (Figure 2B). Biopsy results showed mildly enlarged mucosa cells, with interstitial edema and inflammatory cell infiltration (Figure 2C). He was diagnosed as having primary HOA with gastric involvement, and treated with nonsteroidal antiinflammatory drugs and H_2-receptor antagonist.

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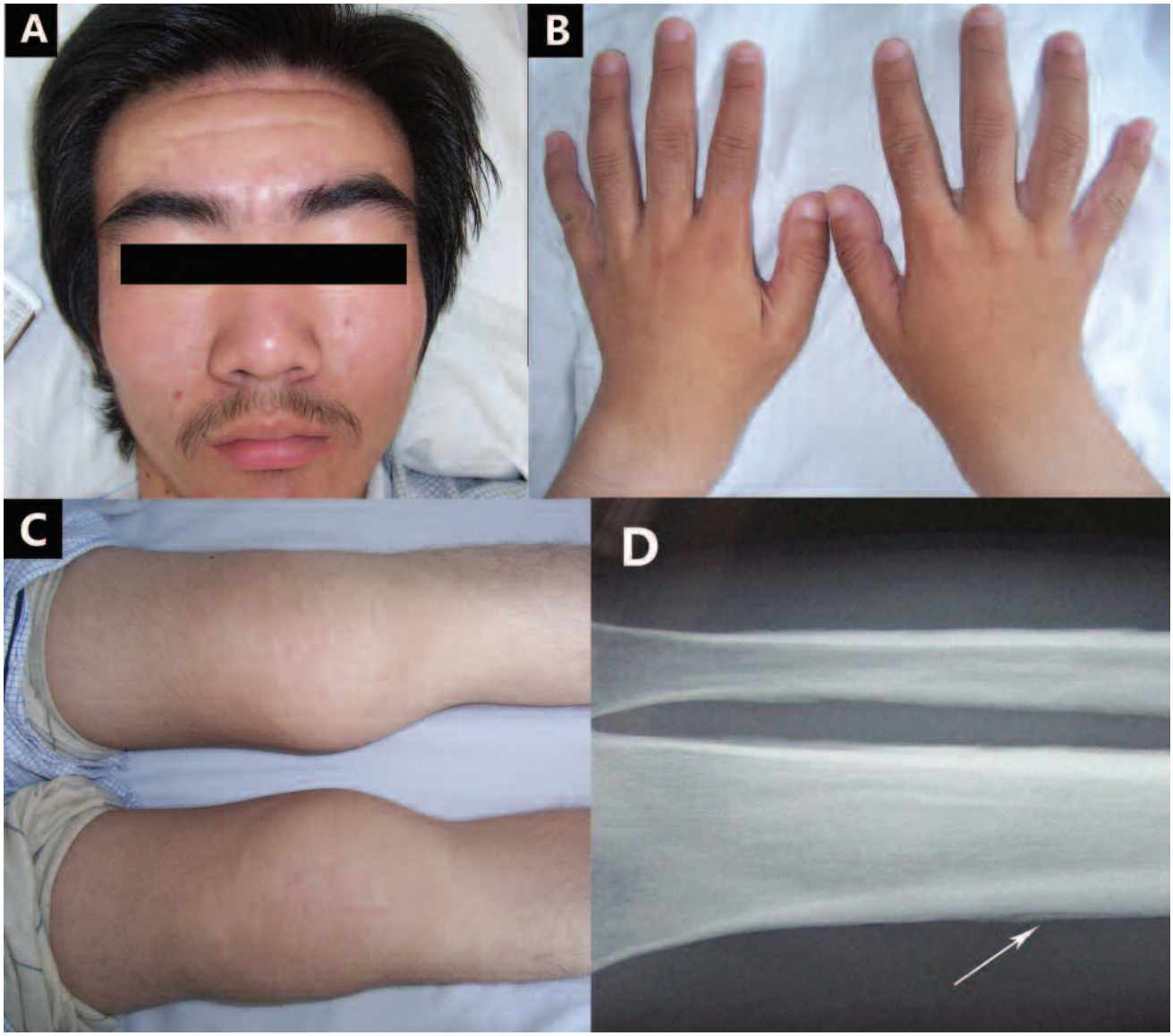

Figure 1. A-C. Dermal and bone manifestations of primary hypertrophic osteoarthropathy. D. Radiograph reveals thickened cortical plate and periosteal new bone formation in bilateral tibio-fibula (arrow).

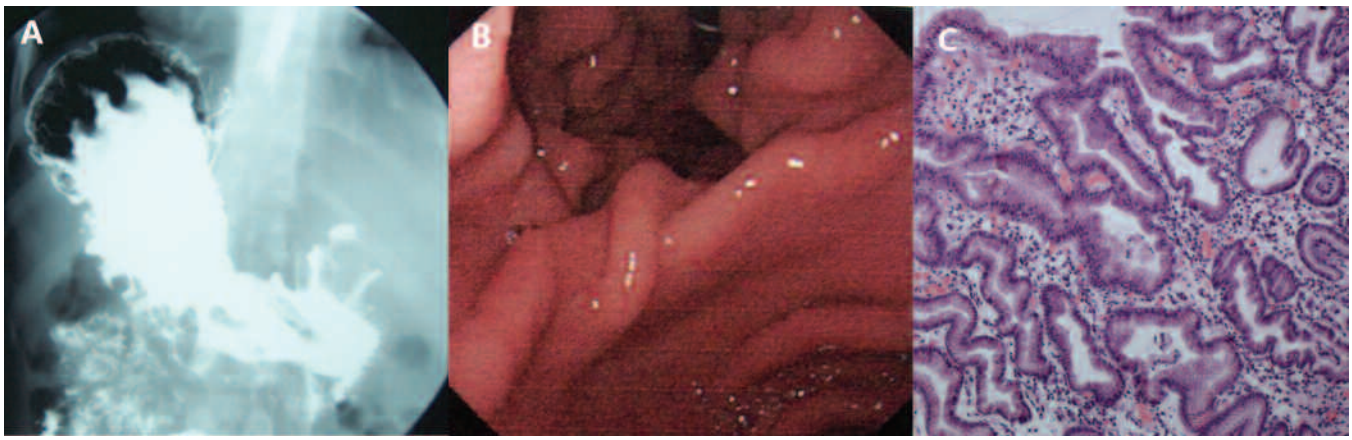

Figure 2. A. Barium radiography; and B. gastroscopy show gyrus-like mucus folds at gastric body and fundus. C. Gastric mucosa biopsy showed mildly enlarged mucosa, with interstitial edema and inflammatory cell infiltration.