Dysphagia Related to Esophagus Compression by Anterior Cervical Ossification in a Patient with Ankylosing Spondylitis

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A 37-year-old elderly white man presented with dysphagia for 3 months. He had been followed for the previous 12 years for HLA-B27–positive ankylosing spondylitis (AS) without extraarticular manifestations, and treated with tumor necrosis factor-α (TNF-α) inhibitors for the last 6 years (infliximab, then adalimumab).

At presentation, he reported progressive difficulty with swallowing solid foods; but he had no pain, cough, or weight loss. The patient was obese (body mass index 30.1 kg/m^2), but without diabetes or metabolic syndrome. He had no skin disease and had never received retinoids. Circulating vitamin A levels were normal. Physical examination showed dorsal kyphosis, a limited range of cervical motion (occiput wall distance 14 cm), and the Bath Ankylosing Spondylitis Disease Activity Index was 5.7. Flexible laryngoscopy showed bulging of the posterior pharynx wall. Exuberant anterior cervical spine ossification was observed on radiographs at C4–C5 (Figure 1). This was confirmed by computed tomography showing prominent osseous formation at the anterior corners of C4–C5, compressing the esophagus (Figure 2). A cervical spine radiograph performed 8 years earlier that was available showed more limited spinal ossification that developed vertically and was diagnosed at the time as syndesmophyte (Figure 3). Dysphagia progressively improved with swallowing rehabilitation therapy.

Dysphagia related to cervical spine disease is observed in diffuse idiopathic skeletal hyperostosis (DISH), but has rarely been associated with cervical spine involvement in AS[1,2]. This patient had established AS with bilateral sacroiliitis and syndesmophyte formation at the cervical spine, but did not satisfy the DISH criteria. The anterior cervical ossification progressed from a preexisting syndesmophyte, despite TNF-α inhibitors.

Figure 1. Lateral cervical spine radiograph in 2014 at the time of dysphagia showing exuberant ossification developed from the anterior corners of C4–C5, bony bridging at C2–C3, and ossification at the anterior corner of C6.

REFERENCES

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Figure 2. Cervical spine computed tomography of the patient in 2014 showing prominent ossification that compressed the esophagus.

Figure 3. Lateral cervical spine radiograph in 2006 showing syndesmophyte at C4–C5.