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Case Report

Diffuse Idiopathic Skeletal Hyperosteosis with Parsonage–Turner Syndrome Appearance

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ABSTRACT

Diffuse idiopathic skeletal hyperosteosis is recognized by the appearance of at least 3 bony bridges in the anterolateral spine, and rarely it can be confused with Parsonage–Turner syndrome A 50-year-old male patient was admitted for left shoulder pain for approximately 2 years. There were weaknesses in deltoid, supraspinatus, biceps brachi, and wrist extension muscles. Hypoesthesia was found in the left C6 dermatom. Deep tendon reflexes were mildly hyperactive bilaterally and symmetrically. No pathological upper motor neuron signs were seen. On cervical magnetic resonance imaging T2-weighted images, hypointense osteophytes are observed in the posterior corners of 4 consecutive vertebral corpses, and disc spaces are preserved in the sagittal section. There were no findings in electromyography similar to Parsonage–Turner syndrome or cervical radiculopathy. Sacroiliac joint magnetic resonance imaging was found normal. According to the cervical computed tomography and dorsal radiography, we thought that the patient was diagnosed as diffuse idiopathic skeletal hyperosteosis syndrome. Even if Parsonage–Turner syndrome is considered in adult male patients presenting with shoulder pain, weakness, and atrophy of the shoulder girdle muscles, it would be valuable to consider diffuse idiopathic skeletal hyperosteosis disease involving the cervical vertebrae in the differential diagnosis.

Keywords: Diffuse idiopathic skeletal hyperosteosis, Parsonage–Turner syndrome, shoulder pain

INTRODUCTION

Diffuse idiopathic skeletal hyperosteosis (DISH) is recognized by the appearance of at least 3 bony bridges in the anterolateral spine opposite the aorta.[1] The shoulder, elbow, wrist, pelvis, hip, knee, and ankle joints may be ossified.[2] The pathogenesis of DISH is not yet known.[3] It is known that DISH is associated with coronary and aortic and respiratory diseases.[3] So the diagnosis of DISH is very important for the prevention of the complications of DISH.

Diffuse idiopathic skeletal hyperosteosis is generally asymptomatic and diagnosed only by radiological findings.[4,5] Some patients describe back pain and spinal stiffness.[3] Back pain is greater in the early stage and decreases in the mature stage of DISH.[3] Diffuse idiopathic skeletal hyperosteosis may also cause dysphagia, airway obstruction, and spinal fracture.[3] If the abundant bones locate anterior to the vertebral bodies in the cervical vertebrates, trachea and esophagus may be displaced.[6] The presence of dysphagia and airway obstruction should be considered in all patients with DISH.

Parsonage–Turner syndrome (PTS) is a brachial neuritis and characterized by acute onset of unilateral shoulder pain, followed by weakness in the proximal mid-arm or distal upper limb. This condition is associated with the involvement of each brachial plexus nerve (multiple mononeuropathies).[7,8] Infection, surgery, or less frequent vaccination may initiate the PTS.[8-10] Here, a case report of DISH resembling PTS is described.

CASE PRESENTATION

A 50-year-old man was admitted to the Physical Medicine and Rehabilitation outpatient clinic with complaints of left shoulder pain for about 2 years. Shoulder

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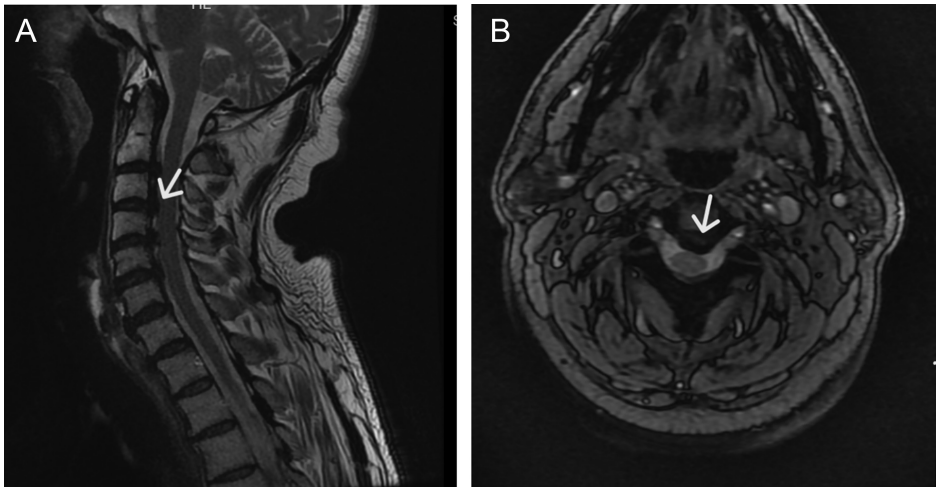

Figure 1. (A) On T2-weighted images, hypointense osteophytes are observed in the posterior corners of 4 consecutive vertebral corpses, and disc spaces are preserved in the sagittal section. (B) In addition, erasure of the anterior subarachnoidal space and contact with the spinal cord is observed in the axial section.

pain radiated to the forearm and was accompanied by numbness. The patient did not describe dysphagia and respiratory distress. Medical history was unremarkable except for smoking and a previous myocardial infarction. There was no family history of rheumatological disease. A detailed musculoskeletal examination was performed. Examination revealed atrophy of the left deltoid and supraspinatus muscles. There was no winging of the scapula. There was weakness in the deltoid, supraspinatus, biceps brachi, and wrist extension muscles (British Medical Research Council scale, muscle strength grade 4/5). Hypoesthesia was detected in the left C6 dermatome. Deep tendon reflexes were mildly hyperactive bilaterally and symmetrically. Pathologic upper motor neuron findings were not observed. A prediagnosis of PTS or brachial plexus lesion was considered, and investigations were planned. Posteroanterior chest radiography was normal. Non-contrast magnetic resonance imaging (MRI) of the cervical spine, left brachial plexus, and left shoulder was performed. Magnetic resonance imaging of the left brachial plexus was normal. Magnetic resonance imaging of the left shoulder joint showed partial tear of the supraspinatus tendon. Cervical MRI T2-weighted images showed hypointense osteophytes at the posterior corners of 4 consecutive vertebral corpses and preserved disc spaces on the sagittal section. In addition, the anterior subarachnoidal space is erased and in contact with the spinal cord in the axial section (Figure 1A and B). We performed the nerve conduction studies and needle electromyography (EMG) for investigating PTS or brachial plexus lesion. The patient's nerve conduction studies (ulnar and radial nerve motor and sensory nerve conduction studies, lateral antebrachial cutaneous studies, and medial antebrachial cutaneous sensory nerve conduction studies) were normal. Left median nerve sensory and motor nerve conduction studies showed that the patient had moderate carpal Tunnel syndrome. Needle EMG (deltoid, biceps brachi, triceps brachi, abductor pollicis brevis, abductor digiti minimi, and supraspinatus muscles) were also found normally. There were no definite electrophysiologic findings suggestive of left cervical radiculopathy or brachial plexus lesion. The patient was evaluated in the neurosurgery clinic with cervical computed tomography (CT) (Figure 2A and B) and dorsal radiography (Figure 3). In sagittal section of the CT, flowing ossification along the anterior and posterior aspects of 4 contiguous vertebrae was shown. Also, in the axial section, mild narrowing of the spinal canal was shown. In dorsal radiography, flowing ossification of the anterior longitudinal ligament involving the thoracic spine was shown. Sacroiliac joint MRI was found normal. Laboratory tests revealed normal C-reactive protein (3.19 mg/L) levels, negative Brucella tube agglutination (with Coombs) test, normal biochemistry test, and normal hemogram test. According to cervical CT and dorsal radiography, the patient was diagnosed as DISH syndrome.

MAIN POINTS

- Parsonage–Turner syndrome is a lesion of the brachial plexus associated with shoulder pain and weakness in the muscles around the shoulder.
- Diffuse idiopathic skeletal hyperosteosis may be confused with Parsonage–Turner syndrome (PTS), especially when it involves the cervical region.
- Diffuse idiopathic skeletal hyperosteosis disease should be considered in the differential diagnosis in patients with PTS.

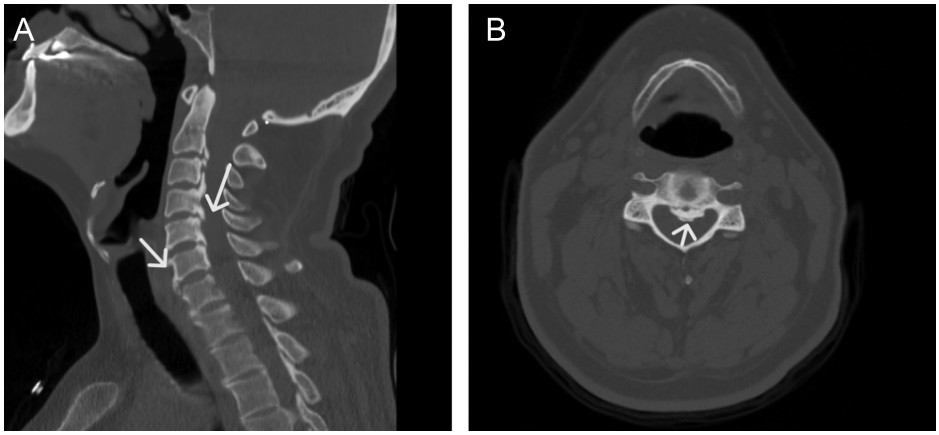

Figure 2. (A) In sagittal section of the Computed tomography, flowing ossification along the anterior and posterior aspects of 4 contiguous vertebrae was shown. (B) Also, in the axial section, mild narrowing of spinal canal is shown.

DISCUSSION

We report a case of DISH involving especially the upper cervical vertebrae (C3-6). According to the history and physical examination findings, the clinical presentation of the patient was similar to that of the Parsonage–Turner syndrome. Similar to our case report, PTS is characterized by sudden onset of shoulder pain and upper-extremity muscle weakness. This syndrome affects the scapular arch and may involve motor and sensory fibers together.[11] Rotator cuff tears, adhesive capsulitis, amyotrophic lateral sclerosis, and cervical spondylopathy are considered as differential diagnosis.[1]

In PTS, acute denervation (fibrillation and positive sharp waves) and axonal degeneration findings are found on EMG.[11] In addition, intramuscular edema (T2-weighted signal increase), muscle atrophy, and fat infiltration can be demonstrated on MRI.[12,13] Our patient did not have intramuscular edema, muscle atrophy, and fatty infiltration on shoulder MRI. In our case report, we thought that the diagnosis was not PTS because brachial plexus MRI and EMG were normal. In particular, a partial tear in the left supraspinatus tendon may have caused the patient to experience severe shoulder pain in addition to neuropathic pain. If there had been no tendon rupture in the supraspinatus, we might not have seen severe shoulder pain in this patient with DISH. The prognosis in PTS is good, and the pain resolves spontaneously in approximately 80%-90% of cases.[11] The lack of spontaneous recovery and the persistence of pain for 2 years in our patient lead us away from the diagnosis of PTS.

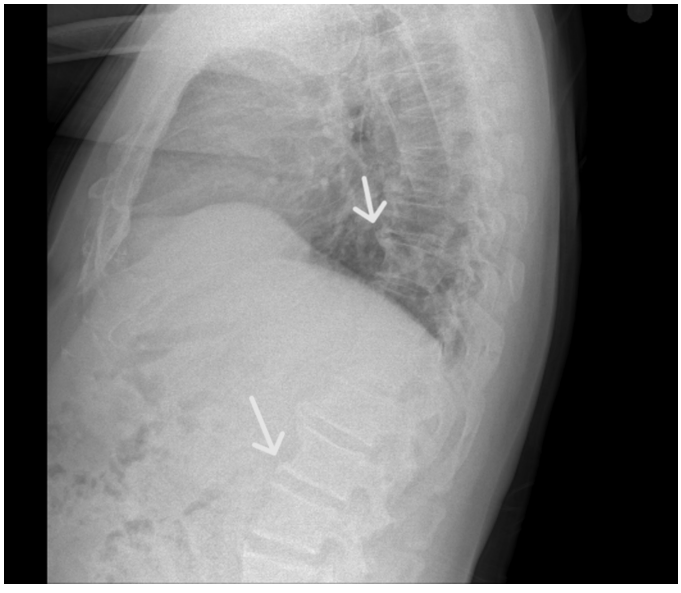

Figure 3. Flowing ossification of the anterior longitudinal ligament in the thoracic spine.

Diffuse idiopathic skeletal hyperosteosis is characterized by the presence of at least 3 bony bridges at the anterolateral spine opposite to the aorta.[1] In 1976, the radiographic criteria of DISH were described as follows:[14] (1) the presence of "flowing" ossification along the anterolateral aspects of at least 4 contiguous vertebral bodies; (2) the absence of extensive radiographic changes of degenerative disc disease (vacuum phenomenon and marginal sclerosis of the vertebral body); (3) the absence of apophyseal joint bony ankylosis and sacroiliac joint erosion, sclerosis, or bony fusion.[14] In recent years, early DISH diagnostic criteria have been tried to be established. Early DISH was described as follows: (1) the presence of a spinal segment with a complete bone bridge with an adjacent segment of at least a near-complete bone bridge and another adjacent segment with at least the presence of newly formed bone and (2) at least 3 adjacent segments as showing a near-complete bone bridge.[15] We diagnosed our case report as a DISH disease according to 1976 radiographic criteria. In DISH, patients may complain of difficulty in breathing and swallowing, especially when the cervical vertebrae are involved.[3]

Our patient did not have a difficulty in breathing and swallowing.

A significant proportion of DISH patients have back pain. However, our patient had no back pain and neck pain despite involvement of the thoracic and cervical vertebrae. Similar to our patient, there are some studies showing that DISH patients have less low back and back pain compared to the normal population.[16]

Diffuse idiopathic skeletal hyperosteosis is associated with the metabolic syndrome, coronary and aortic diseases, and respiratory problems.[3] So the clinicians should not oversimplify DISH and should keep it in mind in the differential diagnosis in patients.

In conclusion, we described a patient with DISH who had similar findings to Parsonage–Turner syndrome. Even if PTS is considered in adult male patients presenting with shoulder pain, weakness, and atrophy of the shoulder girdle muscles, it would be valuable to consider DISH disease involving the cervical vertebrae in the differential diagnosis.

Informed Consent: Written informed consent was obtained from patient who participated in this study.

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Declaration of Interests: The authors have no conflicts of interest to declare.

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