

Occipital Enthesophyte in Axial Spondyloarthritis: Report of two cases

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Abstract

Axial spondyloarthritis (axSpA) is a chronic inflammatory condition primarily affecting the sacroiliac joints and spine, with enthesitis as its hallmark feature. While enthesitis commonly involves lower limb sites, cranial localizations, such as the external occipital protuberance, are exceptionally rare. We report two cases of young male patients with long-standing axSpA presenting with occipital enthesophytes. A 34-year-old male with juvenile-onset axSpA exhibited bilateral sacroiliitis, coxitis, and cervical stiffness. A 33-year-old male with chronic polyarthritis since adolescence showed advanced axial and peripheral structural damage. Cervical radiographs in both revealed an enthesophyte at the external occipital protuberance. Laboratory findings indicated elevated inflammatory markers, with HLA-B27 positivity in one patient. Treatment with TNF inhibitors (etanercept and golimumab) resulted in clinical improvement. These cases highlight the importance of comprehensive imaging to identify rare enthesal sites in axSpA, enabling tailored therapeutic strategies.

Keywords: Axial Spondyloarthritis; Occipital Enthesophyte; Rare Localization; Radiography; Cervical Spine

1. Introduction

Axial spondyloarthritis (axSpA) is a chronic inflammatory rheumatic disease primarily affecting the sacroiliac joints and spine. Enthesitis, inflammation at the site of tendon or ligament attachment to bone, is a hallmark of the disease and may lead to new bone formation such as syndesmophytes or enthesophytes. Enthesophytes are most frequently described at peripheral sites including the Achilles tendon, plantar fascia, and patellar tendon [1,2]. Axial enthesophytes occur predominantly at vertebral endplates and spinal ligaments. Involvement of the external occipital protuberance, corresponding to the insertion of the ligamentum nuchae and neck extensor muscles, is exceptionally rare and scarcely documented in the literature [3,4]. We describe two cases of occipital enthesophyte in patients with long-standing axSpA, illustrating the clinical presentation, imaging features, and therapeutic response. This report describes two cases of axSpA with occipital enthesophytes, emphasizing the need to recognize atypical enthesal localizations to enhance diagnosis and management.

2. Case Reports

2.1. Case 1

A 34-year-old male, ex-smoker (7 pack-years, quit 2 years prior), with a history of hepatic hydatid cyst surgery in 2010, presented with inflammatory cervico-lumbar pain, hip pain, and buttock pain since age 11. Examination revealed restricted hip mobility and incomplete cervical spine syndrome. Laboratory tests showed elevated CRP (25 mg/L) and

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ESR (52 mm/h). Pelvic radiography confirmed bilateral stage IV sacroiliitis and coxitis. Cervical spine radiography demonstrated loss of cervical lordosis, C7 vertebral squaring, and an enthesophyte at the external occipital protuberance (Figure 1). Juvenile-onset axSpA with bilateral coxitis was diagnosed. Etanercept (50 mg/week) was initiated after NSAID failure, resulting in clinical improvement.

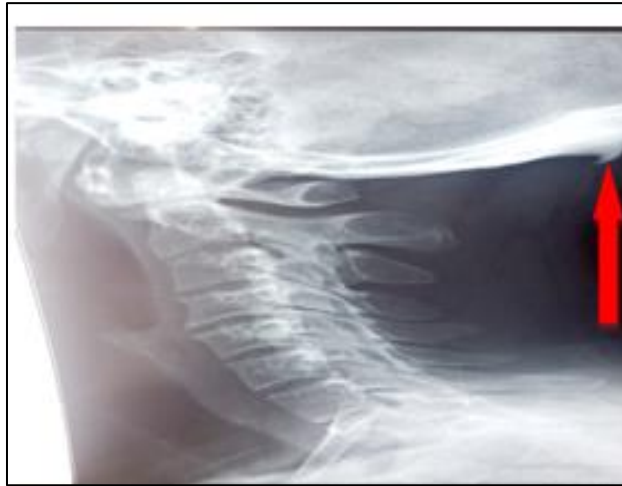


Figure 1 Lateral cervical spine radiograph showing an enthesophyte at the external occipital protuberance

2.2. Case 2

A 33-year-old male, followed for chronic polyarthritis since age 16, presented with inflammatory back pain, buttock pain, and heel pain. Examination revealed diffuse spinal stiffness, sacroiliac tenderness, and arthritis of the hips, knees, ankles, and proximal interphalangeal joints. Imaging showed bilateral stage IV sacroiliitis, hip joint narrowing, and enthesopathies at the ischial tuberosities, pubic symphysis, and greater trochanters. Cervical spine radiography revealed an occipital enthesophyte (Figure 2). Laboratory tests indicated CRP at 12.8 mg/L and HLA-B27 positivity. Golimumab was initiated for advanced structural damage and persistent inflammation, with a favorable response.



Figure 2 Lateral cervical spine radiograph demonstrating an enthesophyte at the external occipital protuberance

3. Discussion

Enthesophytes develop at tendon or ligament insertion sites as a consequence of chronic enthesitis followed by abnormal bone repair. In axSpA, this process is driven by persistent inflammation at the bone–enthesis interface, leading to fibrocartilage metaplasia, calcification, and ossification.

Peripheral entheses are more often affected than axial or cranial sites. Occipital localization is rare, possibly due to the lower mechanical and inflammatory burden compared to weight-bearing entheses [1,2]. The external occipital protuberance serves as the insertion point for the ligamentum nuchae and multiple cervical muscles (trapezius, splenius capitis, occipitalis), which may be subjected to combined inflammatory and mechanical stress.

The literature contains very few reports of occipital enthesophyte in axSpA [3,4]. In most cases, it occurs in advanced disease stages or in patients with insufficient control of inflammation. In our patients, both had more than 10 years of disease evolution and severe structural changes, suggesting that occipital involvement may be a late manifestation. Detection requires targeted imaging, as standard spinal radiographs may not include the skull base. Occipital pain, tenderness, or swelling in axSpA patients should prompt lateral cervical or skull-base imaging.

Both of our patients responded symptomatically to anti-TNF therapy, consistent with the known efficacy of biologics on enthesial inflammation [5,6]. However, established ossified lesions did not regress, underscoring the importance of early diagnosis and aggressive inflammation control to prevent irreversible damage. This report emphasizes the need to consider rare enthesial sites in axSpA evaluation, both for accurate disease assessment and for tailored imaging strategies.

4. Conclusion

Occipital enthesophytes are a rare manifestation of axial spondyloarthritis, typically observed in advanced stages of the disease. Their detection, often incidental, highlights the importance of comprehensive imaging including the upper cervical spine and skull base to fully assess disease extent

Compliance with ethical standards

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Disclosure of conflict of interest

The author declares no conflict of interest.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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