

Diffuse Idiopathic Skeletal Hyperostosis in a Young Male

Ashish Sharma¹ , Ashok Kumar² , Anunay Agarwal³ 

A 32-year-old man was being treated elsewhere with sulfasalazine for axial spondyloarthritis for the past 6 months. The response to treatment was suboptimal. Moreover, he developed oligozoospermia leading to infertility. After 8 months of symptom onset, he presented to our rheumatology clinic. He had progressive back pain along with stiffness, which did not change with activity or rest. The examination was notable for high body mass index (38 kg/m²), kyphosis, and reduced spinal mobility. Laboratory results showed normal erythrocyte sedimentation rate and C-reactive protein; human leukocyte antigen B27 was negative. Computed tomography of the spine revealed ossification of the anterior longitudinal ligament (OALL) in the thoracic spine (white arrows in Figures 1, 3, and 4; asterisk in Figure 2) and calcification of the supraspinous ligament (black arrows in Figure 1). However, sacroiliac joints and intervertebral discs were normal on computed tomogram and magnetic resonance imaging. The patient was diagnosed with diffuse idiopathic skeletal hyperostosis (DISH), which was treated with physiotherapy, vitamin D supplementation, and weight reduction; sulfasalazine was discontinued. Ten months later, the back pain had reduced, spinal mobility had marginally improved, and semen sperm count became normal.

Diffuse idiopathic skeletal hyperostosis, also known as Forestier disease, causes ossification of ligaments and entheses, predominantly involving the thoracic spine. Ossification of the anterior longitudinal ligament is a common radiological finding in DISH. It gives rise to calcification in front of the vertebral bodies. When multiple contiguous vertebrae are involved, the appearance is likened to flowing calcification.¹ Ossification of the anterior longitudinal ligament may be mistaken for bridging syndesmophytes which are seen in axial spondyloarthritis, as had happened in our patient. However, syndesmophytes result from ossification of annular fibrosus of intervertebral discs and, therefore, the area in front of the vertebral bodies is unaffected.² Moreover, OALL is more bulky as compared to syndesmophytes. Diffuse idiopathic skeletal hyperostosis is closely associated with obesity and occurrence before the fifth decade of life is unusual. Intervertebral discs and sacroiliac joints are characteristically spared, differentiating them from spinal osteoarthritis and spondyloarthritis, respectively.

ORCID iDs of the authors:

A.S. 0000-0002-4810-8921;
A.K. 0000-0002-3875-9683;
A.A. 0000-0002-2310-4748.

Cite this article as: Sharma A, Kumar A, Agarwal A. Diffuse idiopathic skeletal hyperostosis in a young male. *Eur J Rheumatol.* 2023;10(2):84-85.

- ¹ Department of Rheumatology, Rheumatology Clinic, New Delhi, India
² Department of Rheumatology, Fortis Ft. Lt. Rajan Dhall Hospital, New Delhi, India
³ Department of Medicine, Friarage Hospital, UK

Corresponding author:
Ashish Sharma
E-mail: ash.bluneney@gmail.com

Received: June 16, 2022
Accepted: May 04, 2023
Publication Date: July 13, 2023

Copyright@Author(s) - Available online at
www.eurjrheumatol.org.

Content of this journal is licensed under a Creative Commons Attribution-NonCommercial 4.0 International License.



Figure 1. Computed tomography of the spine, sagittal section, showing ossification of anterior longitudinal ligament (white arrow) and calcification of supraspinous ligament (black arrows).



Figure 2. Three-dimensional reconstruction of the computed tomogram of the spine, coronal section, showing ossification of anterior longitudinal ligament (asterisk).



Figure 3. Three-dimensional reconstruction of the computed tomogram of the spine, sagittal section, showing ossification of anterior longitudinal ligament (arrow).

Sulfasalazine is commonly used in the treatment of peripheral forms of spondyloarthritis. Some studies have shown its efficacy in axial spondyloarthritis.³ Oligozoospermia is an uncommon adverse effect of sulfasalazine, which is usually reversible after cessation of



Figure 4. Computed tomography of the spine, axial section at fourth thoracic vertebra, showing ossification of anterior longitudinal ligament anterior to the vertebral body (white arrow).

the drug.⁴ There is no role of disease-modifying anti-rheumatic drugs (DMARDs) in the treatment of DISH. Ossification of the anterior longitudinal ligament can also occur in spondyloarthritis. However, in the absence of sacroiliitis, HLA B27, and normal acute phase reactants, the diagnosis of spondyloarthritis is unlikely. This case highlights the importance of differentiating DISH from spondyloarthritis to prevent unnecessary treatment with DMARDs, without any benefit.

Informed Consent: Informed consent was obtained from the patient described in the manuscript.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – A.S.; Design – A.S., A.K.; Supervision – A.K.; Resources – A.S., A.K., A.A.; Materials – A.S., A.K., A.A.; Data collection and/or processing – A.S., A.K., A.A.; Analysis and/or interpretation – A.S., A.K., A.A.; Literature search – A.S., A.A.; Writing – A.S., A.K., A.A.; Critical review – A.K.

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

Funding: The authors declared that this study has received no financial support.

References

1. Belanger TA, Rowe DE. Diffuse idiopathic skeletal hyperostosis: musculoskeletal manifestations. *J Am Acad Orthop Surg.* 2001;9(4):258-267. [\[CrossRef\]](#)
2. Sharma A, Arya V, Kasetty A, eds. *A Rheumatologist's Guide to Interpreting Imaging in Spondyloarthritis.* New Delhi: Jaypee; 2020.
3. Chen J, Liu C. Is sulphasalazine effective in ankylosing spondylitis? A systematic review of randomized controlled trials. *J Rheumatol.* 2006;33(4):722-731.
4. Toovey S, Hudson E, Hendry WF, Levi AJ. Sulfasalazine and male infertility: reversibility and possible mechanism. *Gut.* 1981;22(6):445-451. [\[CrossRef\]](#)