



## XL COMU Awards 2021 – Case Reports

### **Association between diffuse idiopathic skeletal hyperostosis and long-term use of isotretinoin: case report**

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**ABSTRACT:** Diffuse Idiopathic Skeletal Hyperostosis (DISH) is a systemic condition that affects mainly the anterolateral spine, with abundant bone growth and formation of bridges between the vertebral bodies, but ossification in peripheral entheses may also occur. Its prevalence increases with age and is higher in males. Diagnosis at ages under 40 years are extremely rare. Hence, this report aims to describe a rare case of DISH in a young woman probably due to the long-term use of isotretinoin and discuss its presentation, bone involvement and differential diagnosis. A 37-year-old female patient was admitted to the Rheumatology service with progressive limitation of neck rotation and back flexion during the past 3 years, associated with great loss of function and impairment in daily activities. She had a previous diagnosis of congenital ichthyosiform erythroderma, managed with low doses of isotretinoin for 12 years. The current weekly isotretinoin dosage was 180mg. Prior image exams showed axial spondylopathy with diffuse peripheric enthesopathy since she was 33 years old and sacroiliitis with asymmetric syndesmophytes in the lumbar spine that resembled a psoriatic arthritis. Other diseases were denied. Family history included a case of rheumatoid arthritis but no history of psoriasis. Physical exam included erythema and diffuse skin peeling in the upper and lower limbs. Her hands were deformed with ulnar deviation but had no signs of arthritis. Cervical rotation was stiff and ranged 30° to the left and 35° to the right. Schober test distanced 1cm and Patrick's Faber test was negative. Laboratory tests showed no alteration except for a serum 25-OH vitamin D of 7.25ng/mL (reference value: >30ng/mL). Biochemical and bone turnover markers were normal, including serum total calcium, magnesium, phosphorus, parathyroid hormone and alkaline phosphatase. Inflammatory markers C-reactive protein and erythrocyte sedimentation rate were within normal ranges. Human leukocyte antigen B27 was negative. Conventional radiographies were performed and showed increased bone texture with calcification of anterior and lateral ligaments of the cervical, thoracic and lumbar spine. Bone mineral density, evaluated by dual-energy X-ray absorptiometry (DXA), showed a Z-score of +3.2 SD at lumbar spine. Trabecular bone score by DXA was normal and vertebral fracture assessment showed no fracture and confirmed the formation of bridges between vertebral bodies. Clinical presentation was not compatible with ankylosing spondylitis nor psoriatic arthritis as there was no typical inflammatory pain, no elevated inflammatory markers, no typical skin lesions and no family history of psoriasis. However, the hyperostosis affecting the spine and the diffuse enthesopathy were very exuberant for DISH, but the female sex, young age and bilateral spine involvement were uncommon finds. As this patient was on long-term isotretinoin therapy, DISH development is more likely an adverse effect of retinoids on bone metabolism. Thus, clinicians and dermatologists should be always alert to the adverse effects of prolonged use of isotretinoin and, in cases of movement restriction, possible bone involvement must be further evaluated, as DISH may be a differential diagnosis. Since there is no specific treatment, early diagnosis can be crucial to avoid extensive bone damage and irreversible morbidity.

**Keywords:** Diffuse idiopathic skeletal hyperostosis; Isotretinoin; Congenital ichthyosiform erythroderma; Case reports.