

Bamboo Spine Mimicking Bekhterev's Disease Caused by Long-term Acitretin Treatment

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Sir,

During the last 20 years several papers have been published regarding the skeletal toxicity of long-term retinoid therapy. The literature has been conflicting. Some centres report development of skeletal hyperostosis due to oral retinoids (1–6). Other investigators do not report radiographic changes during either short-term retinoid therapy (7) or prolonged treatment in adults (8). Osteoporosis caused by acitretin (9, 10) has also been discussed. We here report on 3 patients with lamellar ichthyosis who have been treated with acitretin for several years and developed spinal hyperostoses.

CASE REPORTS

Patient 1 is a 60-year-old woman with lamellar ichthyosis. She was given etretinate 0.75 mg/kg/day from 1979 and later switched to acitretin 0.5 mg/kg/day continuously, and she is still using this medication. In 2001 she complained of increased stiffness in her spine and lower back pain. X-ray examination of the spine showed extensive hyperostosis-like bamboo spine and initially the diagnosis of Bekhterev's disease (spondylitis ankylopoetica) was suspected. However, X-ray of her iliosacral joints was normal and she was HLA B27 negative. The diagnosis of Bekhterev's disease was therefore ruled out by her rheumatologist.

Patient 2 is a 69-year-old man with lamellar ichthyosis. He has used retinoids approximately 0.5 mg/kg/day continuously since 1978. During the last years he has complained of lower back pain and increased stiffness of his spine. X-ray examination showed extensive hyperostosis-like bamboo spine. His iliosacral joints were radiographically normal and he was HLA B27 negative.

Patient 3 is a 45-year-old woman with lamellar ichthyosis. Since 1978 she has used etretinate 0.7 mg/kg/day and, from 1992, acitretin 0.5 mg/kg/day continuously. She experienced increased stiffness of her spine since 2001 and X-ray examination showed hyperostosis-like bamboo spine (Fig. 1). She was HLA B27 negative and her iliosacral joints were normal.

DISCUSSION

Different dermatological centres monitor patients on long-term retinoid therapy differently regarding bone toxicity. Some centres do not make X-ray skeletal investigations, while others do. Interestingly several

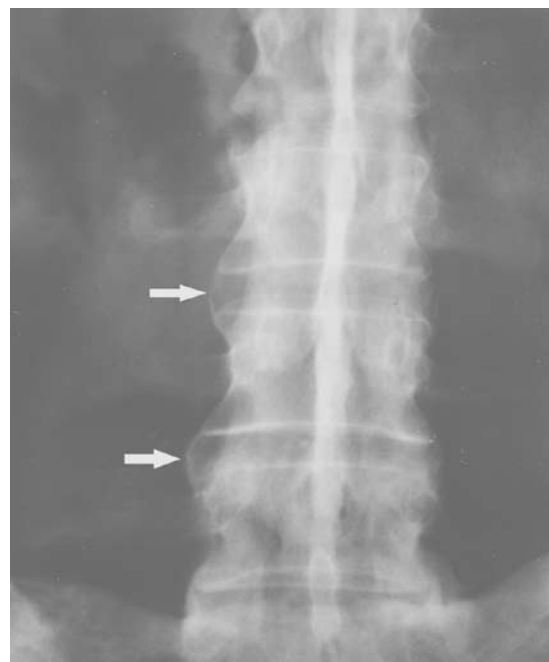


Fig. 1. Conventional X-ray image of lumbar spine in antero-posterior projection in patient 3. Syndesmophytes similar to those seen in Bekhterev's disease lateral to the intervertebral discs (arrows).

centres have not detected hyperostosis in their patients on long-term retinoid therapy. Whether this is explained by the lack of skeletal monitoring or genetic differences between groups of patients is not known. In our department we have, during the last 20 years, routinely performed X-ray surveys of the skeleton at baseline and every second year in patients on long-term retinoid treatment. We experienced two clearly abnormal calcifications (5) in the forearm of a patient with X-linked recessive ichthyosis and a laminar hyperostotic process in the forearm in a 66-year old woman with pustulosis palmoplantaris.

In our previous study (5) radiographs of the spine were of little practical value because skeletal abnormalities are frequently found in people over the age of 50 years. Kilcoyne (11) found X-ray abnormalities of the spine in 86% of the patients at the start of therapy in 236 patients treated with acitretin. In our department we therefore routinely performed X-rays only of the forearms, pelvis and knees and not of the spine of the patients on long-term retinoid.

In patients 1 and 3 we had baseline X-rays of their spines before etretinate was introduced in the late 70s, when the patients were between 35 and 45 years old,

and these radiographs just showed minor changes interpreted as normal. In patient 2 baseline X-rays were not performed. Our patients have used retinoids orally continuously for about 25 years. Their cumulative doses of retinoids are 300–500 g. Because of the excellent effect of acitretin in the treatment of their ichthyosis, our patients have preferred to continue with acitretin even after detecting the spinal hyperostosis over 5 years ago despite the stiffness of the spine is a major problem. We are convinced that their bamboo spines are caused by the drug. They are HLA B27 negative and their iliosacral joints are normal, and this excludes the diagnosis of Bekhterev's disease.

Based on our present knowledge, we propose a baseline skeletal survey of the forearms, pelvis, knees and spine before a planned long-term retinoid treatment in patients with congenital lamellar ichthyosis. Thereafter follow-up examinations should be performed if there is clinical suspicion of skeletal involvement. This should prompt further radiographs of the region in question.

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