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IMAGES IN PAEDIATRICS

Cortical hyperostosis secondary to chronic treatment with prostaglandins☆

Hiperostosis cortical secundaria a tratamiento crónico con prostaglandinas

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We present the case of a female infant aged 7 months with a history of left ventricle and aortic arch hypoplasia arco who had undergone a bilateral pulmonary band. The anatomy of her heart precluded placement of an intraductal stent, which prompted initiation of treatment with prostaglandin E2 (0.02 μg/kg/min) from birth.

At 2 months and a half, follow-up radiographs started to show periosteal thickening of the long bones (Fig. 1) with gradual progression (Fig. 2). Hypervitaminosis A and D and congenital syphilis were ruled out as potential causes of the lesions. The patient underwent measurement of alkaline phosphatase at different timepoints, with levels peaking at 935 U/L at age 4 months (normal range for age, 85–235 U/L).

The patient was treated with prostaglandins through age 5 months, at which time she underwent heart transplantation. Six months later, hyperostosis persisted in the follow-up imaging tests (Fig. 3).

Cortical hyperostosis is the most frequent adverse event associated with chronic treatment with prostaglandin E2, and its incidence is associated with the dose of prostaglandin and the duration of treatment. Its diagnosis is based on clinical, radiographic and laboratory features, and it is essential to rule out other possible conditions, such as sepsis, osteomyelitis, cellulitis, syphilis, cancer, rickets, hypervitaminosis A and D and Caffey disease.[1–3]

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☆ Previous meeting: the study was presented as a poster titled ''Hiperostosis como efecto secundario del tratamiento crónico con prostagladinas en una lactante de 4 meses'' at the 36th National Congress of the Sociedad Española de Cuidados Intensivos Pediátricos; June 12–15, 2022; Seville, Spain.

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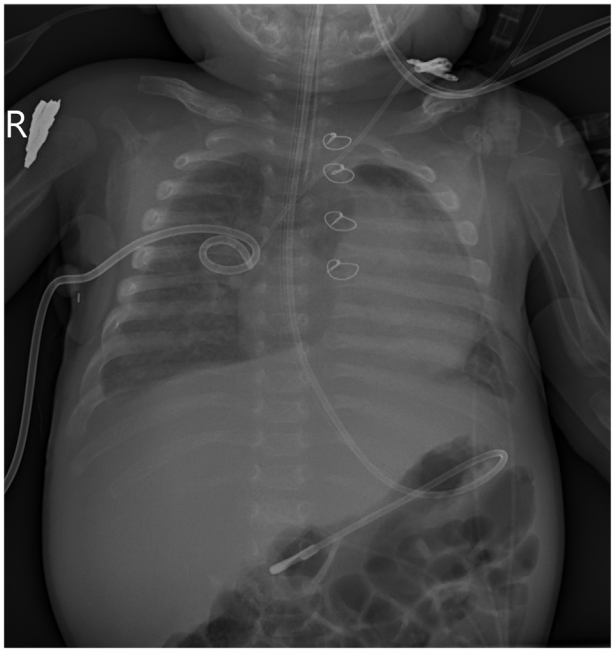

Figure 1 Plain radiograph at age 2.5 months. It is the first radiograph of the patient that showed cortical thickening, which was more evident in the diaphyses of both humeri (arrow), relative to the original appearance of the bones. Thickening was also apparent in other bones including ribs, the clavicles and the scapulae.

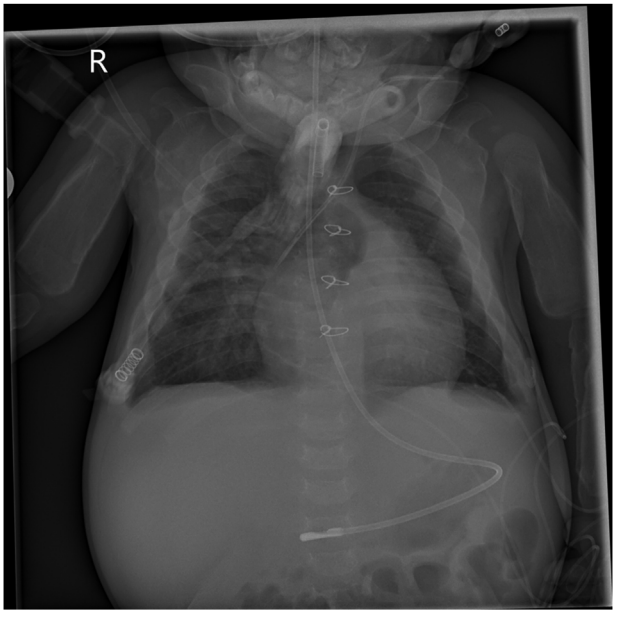

Figure 2 Plain radiograph at age 4 months evincing the progression of the bone abnormality, with hyperostosis and thickening of humeri, scapulae, clavicles and ribs.

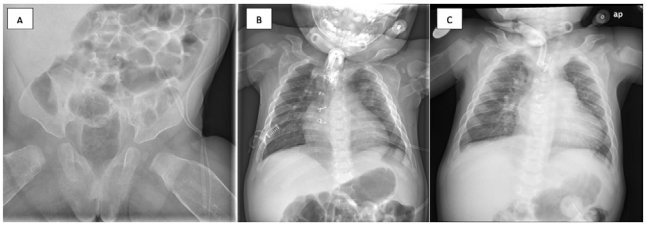

Figure 3 Plain radiographs taken after heart transplantation and at the end of treatment with prostaglandins. (A) At 3 months, (B) at 4 months, (C) at 6 months. They continue to show hyperostosis and bone thickening, without improvement or resolution after discontinuation of prostaglandin treatment. There seem to be no involvement of the pelvis or spine. The mandible was not affected either.

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