

## **Hypertrophic Osteoarthropathy in Childhood Malignancy**

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**Key Words.** Bones, abnormalities · Children, malignancy · Thorax, neoplasms

**Abstract.** A case of hypertrophic osteoarthropathy associated with carcinoma of the nasopharynx in a child is presented. Peculiar findings were the minor changes on the chest film, and the absence of periosteal reactions in the radii.

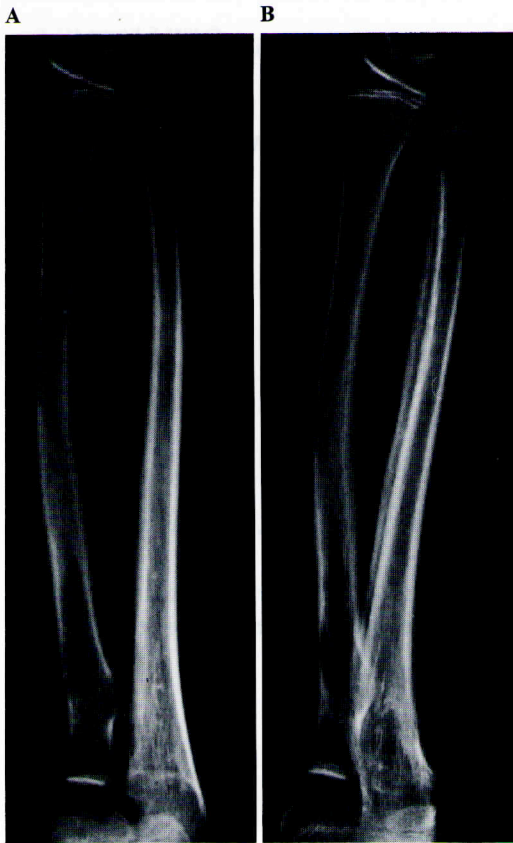
### **Introduction**

Hypertrophic osteoarthropathy (Marie-Bamberger's disease) is well known in adults, but is rarely encountered in children [1–9]. Clinical features may include clubbing of the fingers and toes, arthritis, and a sometimes very painful ossifying periostitis of the tubular bones. Apart from a heredofamiliar form, most cases in children are connected with chronic suppurative lung processes [3–6, 8], congenital cyanotic heart disease [3, 6, 11], or biliary atresia/cirrhosis [3, 8, 10]. The association with malignant conditions, relatively common in adults, is very rare in children [1, 7].

This report describes the occurrence of aching periostitis of the long tubular bones in a patient with 'cured' carcinoma of the nasopharynx.

### **Case Report**

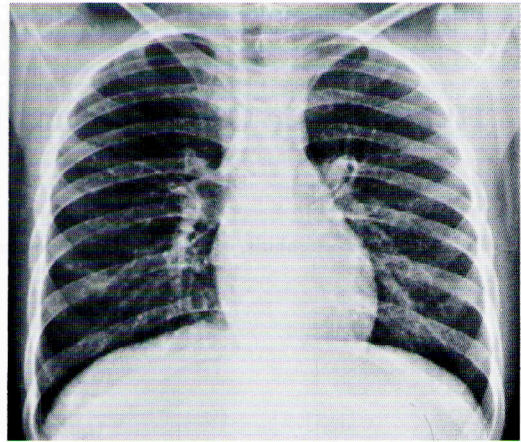
A.D., a 13-year-old boy, was admitted with a 2-month history of complaints of throat, nose, and ears. A diagnosis of undifferentiated epithelial cell carcinoma with regional lymph node metastases had been made elsewhere, and was confirmed on revision of the pathologic specimen. Treatment consisted of 14 weeks of chemotherapy (high doses of MTX and cyclophosphamide) and 7 weeks of radiation therapy (30 Gy). At the end of the treatment, no tumour could be found. Three weeks later the patient developed severe pains in his arms and legs. Roentgenographic examination (fig. 1A) revealed generalized symmetrical periosteal reactions along most long tubular bones. The chest film (fig. 2) showed some small abnormal shadows and CT confirmed the presence of five lung metastases, 3 on the right side and 2 on the left side. Three months later a metastasis in the tenth thoracic vertebra was detected (not represented). The lung metastases remained unchanged. The bone pains continued, and the periosteal reactions grew thicker (fig. 1B). Interferon was administered but till now no improvement has been observed.



**Fig. 1.** **A** Right forearm at start of HOA complaints. Periosteal reaction on radial side of ulna. **B** Three months later. Increase of periosteal changes. Note absence of changes in radius.

## Discussion

Hypertrophic osteoarthropathy (HOA) is a curious condition. Despite many hypotheses, among which long-standing passive hyperaemia, the aetiology remains unclear [3–5, 7, 10, 12]. The skeletal changes of HOA include periosteal reactions along the tubular bones, usually bilaterally symmetrical, especially the distal thirds of the tibiae, fibulae, radii and ulnae, less frequently the femora,



**Fig. 2.** Chest film at start of HOA complaints. Two or three small lung metastases are discernable.

humeri, metacarpals and metatarsals; also other parts of the skeleton may be affected [1, 7, 9]. In some published illustrations [7, 9, 11], like in the present case, the ulnae demonstrate periosteal reactions while the radii do not. The bony changes are usually more pronounced at the insertions of tendons and ligaments [7, 9]. In advanced cases, osteoporosis may be seen [1, 7, 9].

HOA can develop in a period of months and hence can occur in the first half year of life [8]. Very often bony changes, and complaints, disappear with cure of the primary disease [8, 9, 12], sometimes within days after removal of a primary lung cancer [12]. Only rarely [12] have haematogenous metastases to the lungs been recorded in association with HOA, as is the case in our patient. Usually, even after subsidence of HOA following elimination of a primary malignancy, HOA does not recur when lung metastases develop [12].

Many authors mention the predictive value of HOA, especially in association with



malignant tumours [1, 3, 6, 10, 12]. Vogl et al. [12] state that the symptoms of HOA may precede any pulmonary symptom by 1–18 months in cases of neoplastic disease of the chest (in contrast to suppurative lung processes). With regard to the present case, one may only speculate about the predictive value of HOA. In this respect the worsening general condition of our patient after onset of HOA may be of interest.

HOA in children is very rare, especially in association with malignancy, all retrieved cases demonstrating a tumour mass within the thorax [1, 3, 7, 8, 10]. To the best of our knowledge our case is the second description of HOA in association with an epithelial cell carcinoma of the nasopharynx in a child.

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