Maxillary Brown Tumor as Manifestation of Renal Osteodystrophy

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Dear Sir,

Current management of chronic renal failure (CRF) usually prevents the development of serious forms of renal osteodys-trophy. However, treatment noncompliance may give rise to severe and almost forgotten manifestations of bone involvement in uremic children with active osseus metabolism. We report a case of brown tumor in the maxilla of a 3-year-old female with end-stage renal disease (ESRD).

A 3-year-old female with ESRD due to bilateral malformative uropathy and undergoing continuous ambulatory peritoneal dialysis from the 6th month of life was admitted to our hospital because of swelling of the left side of her face. Chronic medical treatment included subcutaneous erythropoietin and oral administration of calcium carbonate, calcium citriol, sodium bicarbonate, multivitamin supplements, and antihypertensive drugs. However, biochemical control of hyperparathyroidism had been poor because of the family's persistent noncompliance with medical therapy and dietetic recommendations. Two months before the admission, she had received a cadaver renal transplant which was extracted 1 month later because of renal vein thrombosis. Physical examination revealed a firm, fixed, painful, 5 × 5 cm mass in the left maxilla. Serum chemistry analysis showed depressed values of 1,25-dihydroxyvitamin D₃ (26 nmol/l) and marked elevations of N-terminal parathyroid hormone (339 ng/l) and alkaline phosphatase concentrations (2,144 U/l). X-ray films of the skull revealed radio-lucent lesions affecting the left maxilla and orbit with the salt-and-pepper bone appearance characteristic of hyperparathyroidism.

Fig. 1. Axial computerized tomography showing an homogenous, soft-tissue density mass in the left cheek and antrum.

Radiological signs of renal osteodystrophy were also present in long bones. Axial computerized tomography demonstrated a homogeneous, soft-tissue density mass, affecting the left cheek and antrum, and extending towards the nasal cavity and ethmoidal posterior spaces (fig. 1). Fine-needle aspiration cytology showed aggregates of multinucleated giant cells within a fibrovascular hemorrhagic stroma. Three weeks after admission, the patient

died as a result of bronchoaspiration and congestive cardiac failure. Although the dosage of calcitriol had been doubled (50 ng/kg/ day), no signs of maxillary mass regression were noticed. The patient's family did not authorize a postmortem study.

Brown tumor is a variant of osteitis fibrosa [1] rarely reported in the hyperparathyroidism

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secondary to CRF and exceptional in infants. Before the extended clinical use of active vitamin D metabolites, its frequency had been estimated to be from 1.5 to 1.7% of adult patients [2, 3]. To our knowledge, this is the 4th case described in children with CRF, always in females [4-6]. The reasons why a very limited number of children with severe renal osteodystrophy develop brown tumors is unknown. In our case, the possibility that erythropoietin treatment could be involved in the occurrence of the tumor cannot be ruled out.

However, this undesirable side effect of eryth-

ropoietin therapy has not been reported to date. Brown tumor treatment should include appropriate management of renal osteodystrophy and curettage or surgical mass removal [7]. Total or subtotal parathyroidectomy has been useful in all reported cases but one [8]. Renal transplant solved the problem in 1 child [6]. No surgical procedure was performed in our patient because of her poor general condition.

This report serves to emphasize that brown tumor should be included in the differential diagnosis of a bone mass in children with CRF. The prolonged survival of infants with ESRD may lead to a higher incidence of this unusual form of renal osteodystrophy in poorly controlled patients.

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