Marked facial enlargement in secondary hyperparathyroidism

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Brown tumours in patients with ESRD represent an extreme form of osteodystrophy. They occur most often in the long bones, ribs and pelvis, but can be found in any bone [1,2]. Clinically significant lesions in the jaws are rare [2]. We report a haemodialysis (HD) patient with a rapidly growing maxillary mass diagnosed as a brown tumour. The case is noteworthy because of the patient’s young age, location, large size of the lesion and unusual course of the disease.

A 17-year-old female had been on a regular haemodialysis programme three times a week because of chronic glomerulonephritis since February 2000. She was non-compliant to dietary prescriptions and did not take phosphate binders as prescribed. Therefore, she soon developed hyperphosphataemia (up to 7.4 mg/dl) and hypocalcaemia (7.5 mg/dl). At the start of HD, PTH levels were normal. Overt secondary hyperparathyroidism (HPT) became evident by March 2004, intact PTH increased up to 1483 pg/ml and circulating markers of bone metabolism were suggestive of a high-turnover bone disease. No signs of aluminium intoxication were found. Other clinical problems included pruritus and bone pain. Parathyroidectomy was proposed but was refused by the patient. In September 2004 a hard maxillary swelling appeared. The maxillary tumour was not painful, but its progressive growth caused a marked deformity of the face (Figure 1). A lateral radiograph of the face showed generalized fibrous dysplasia of maxilla and mandible (Figure 2). CT scans revealed that the tumour was almost homogeneous and that it was invading and destroying the maxilla. A surgical biopsy of the maxillary lesion was taken and the histopathological diagnosis was brown tumour. In the context of the patient’s severe HPT, and the rapid growth of the maxillary lesion, a surgical parathyroidectomy was performed in October 2004. After surgery, profound hypocalcaemia and hypophosphataemia developed, with nearly undetectable intact PTH. This ‘hungry bone’ condition required treatment with oral alphacalcidol (2 µg daily) and elemental Ca supplementation (12 g daily). After parathyroidectomy, the bone tumour gradually diminished, diffuse bone pain abated and pruritus became less. Ca x P declined to normal values within one year and PTH levels remained persistently low. Repeated CT examination showed a progressive decrease in the size of the tumour and refilling of the lesions by calcific material.

Brown tumours are probably slightly more frequent in primary than in secondary HPT. Nevertheless, the rate of these lesions in patients with secondary HPT due to chronic renal failure is highly variable, ranging from 1.5% up to more than 13% [1]. Rapid tumour disappearance or regression is well-documented, after parathyroidectomy, but in certain anatomical sites, decompression of the brown tumour is urgently needed because these lesions expand and can cause local destruction [1]. This is particularly true for maxillary lesions, which may lead to serious deformities of the face and even to a lethal outcome [3]. In our case, the surgical excision of the maxillary mass was not established due to the important size of the tumour.

Conflict of interest statement. None declared.

References


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Fig. 1. Lesion causing facial aesthetic deformity. (A) anterior-posterior and (B) oblique views.

Fig. 2. Lateral radiograph of the face showing generalized fibrous dysplasia of maxilla and mandible.