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Case Report
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Macrognathia secondary to dialysis-related renal osteodystrophy treated successfully by parathyroidectomy

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Abstract. Renal osteodystrophy (ROD) is one of the most common complications affecting patients with chronic renal failure both before and after the initiation of maintenance dialysis, but macrognathia secondary to ROD is rare. Usually, enlarged jaws due to ROD do not return to their normal contours after the treatment of hyperparathyroidism. To the authors' knowledge, this article describes the second case of macrognathia secondary to dialysis-related ROD treated successfully by parathyroidectomy. Immunohistochemical study of the maxilla confirmed that parathyroidectomy could stop maladaptive parathyroid hormone stimulation, which leads not only to the formation of osteoblastic progenitors that become fibroblast-like cells but also to osteoclast formation.

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Case report

A 52-year-old Japanese woman with chronic renal failure was referred to the Department of Oral Surgery of Kawasaki Medical School Hospital chiefly complaining of a painless mass involving the maxilla and mandible. This mass had been present for approximately 6 months and was increasing in size at a steady rate. Jaw enlargement with spacing of dentition and masticatory disturbance had progressed steadily when the patient was referred to us in February 2002. The patient had commenced haemodialysis at 46 years of age, in 1996, and had also been taking a Vitamin D sterol, $1,25(OH)_2D_3$(-calcitriol) since August 2001.

Physical examination disclosed significant diffuse enlargement of both jaws, resulting in facial deformity (Fig. 1A). Maxillary and mandibular involvement caused diffuse smooth swelling of the alveolar ridge, with spacing of the dentition, tooth mobility and increased arch length. The palatal vault disappeared and extended close to the level of the occlusal plane. The mucosa over the mass was normal without ulceration or parasthesia. The interincisal distance at maximum mouth opening was 39 mm. An occlusal radiograph of the anterior maxilla showed loss of the lamina duras and a ground-glass trabecular pattern with a lucent pattern of alveolar bone (Fig. 1B). Radiographs of the skull revealed thickening of the diploic space, loss of the outer table, a diffuse salt-and-pepper appearance and enlargement of both jaws. CT scans of the jaws showed relatively symmetrical enlargement of both jaws with multiple diffuse radiolucency that contained scattered

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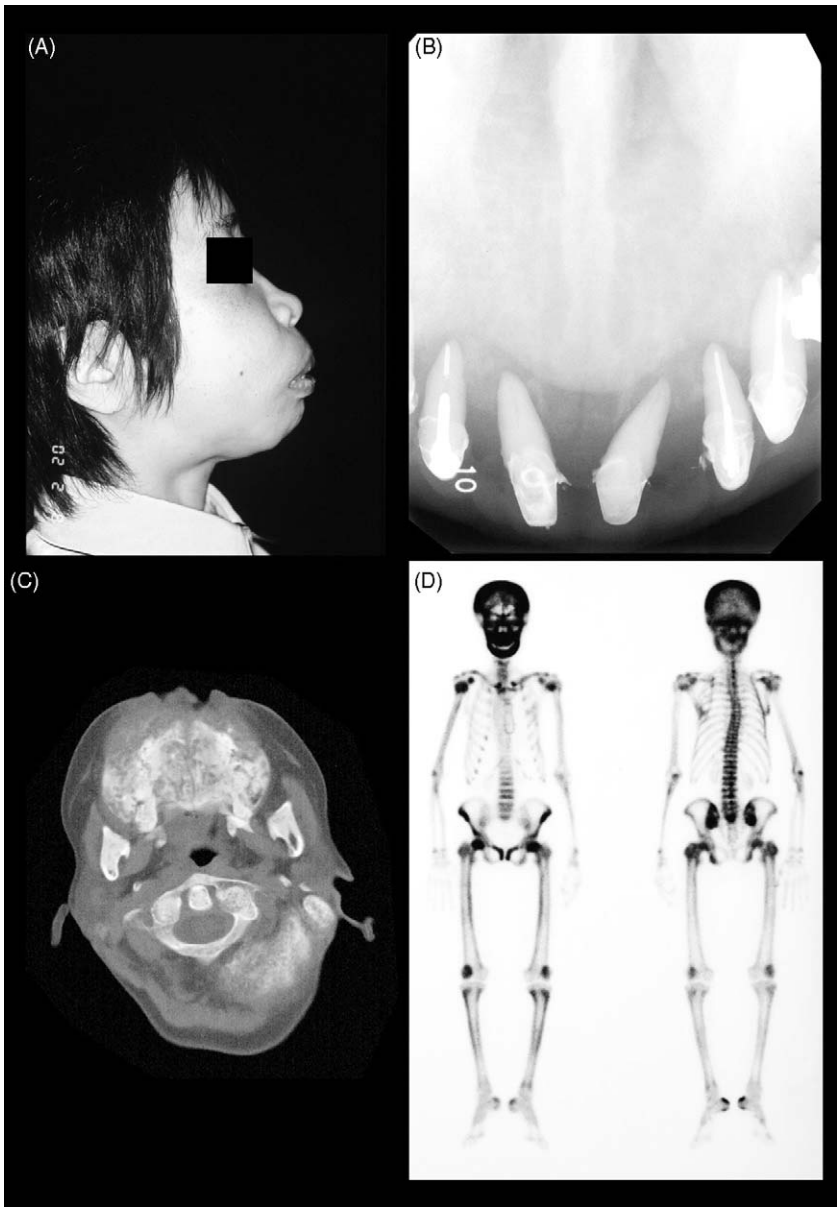

Fig. 1. Clinical photograph and radiographic examinations at the first visit. (A) Right lateral view showing enlargement of both jaws. (B) Occlusal radiograph demonstrating loss of the lamina duras. (C) Axial CT section of the maxilla. (D) Bone scintigram of the whole body.

radio-opacities (Fig. 1C). A radiograph of the sacral spine revealed the sign of rugger-jersey spine. A radiograph of the hands showed subperiosteal erosions. Bone scintigraphy disclosed markedly increased uptake in the jaws, skull and vertebrae (Fig. 1D). Echographic exploration of the neck revealed a hyperplastic parathyroid gland. Parathyroid subtraction scintigraphy with $^{99m}TcO_4^-$ and ^{201}Tl-Cl showed no abnormal accumulation. The patient's metabolic data showed normal levels of calcium (8.6 mg/dl) and calcitonin (39 pg/ml), a high level of phosphorus (7.1 mg/dl), extreme elevation of serum alkaline phosphatase (2530 IU/l) and intact parathyroid hormone (PTH) level (4400 pg/ml).

Under general anaesthesia, a parathyroidectomy (PTX) was done, and a portion of 1 gland was implanted in the right forearm (autografting, AG). The maxilla was easily biopsied by scalpel at the same time. A cut surface of the maxillary mass was whitish. After surgery, recovery was uneventful except for hypocalcaemia. The serum calcium concentration recovered with administration of intravenous calcium hydrate and intraoral alfacalcidol. Serum intact PTH level fell to 15.4 pg/ml. The alkaline phosphatase level was 100 IU/l. These metabolic data were within normal limits.

Three months after surgery, the patient experienced recovery from weakness and regained muscle strength. Both facial and intraoral maxillary deformities slightly diminished in size. The patient could eat solid foods because of improvement in tooth mobility. A postoperative occlusal radiograph of the anterior maxilla disclosed partial regeneration of the lamina duras and partial recovery of the radiolucent areas.

Eight months after surgery, enlargement of the maxilla still remained, but the size and shape of the deformed jaws had markedly improved. CT scans of the jaws disclosed recovery of the cortical line and radiolucencies. Increased uptake in the jaws and skull declined in postoperative bone scintigraphy. Eighteen months after surgery, the patient was satisfied with their improved features (Fig. 2A) and a second biopsy was done to clarify the postoperative state of the jaws.

Histopathologically, irregularly shaped trabeculae of woven bone lay in a cellular fibrous stroma. These assumed a variety of shapes, including small spherical forms, similar to fibrous dysplasia. Osteoblasts uniformly lay along the edges of bony trabeculae, so-called osteoblastic rimming. Clusters of osteoclastic cells were seen in cellular fibrous stroma or along the trabeculae bone (Fig. 2B). These microscopic findings were consistent with osteitis fibrosa. In the postoperative state, extensive cellular fibrous areas decreased, the peripheral portion of bony trabeculae displayed a conspicuous osteoblastic rimming, some of which demonstrating a more mature appearing trabecula structure with osteocytes. Clusters of osteoclastic cells were not seen in stroma. Mature fat cells were present between the bony trabeculae (Fig. 2C).

Formalin-fixed and paraffin-embedded tissue from a biopsied specimen of the maxilla was utilized for immunohistochemical study. The avidin–biotin–peroxidase complex method with an ABC kit (Vector Laboratories, Burlingame, CA, USA) was used for measurement of bone morphogenic proteins (BMPs 2–7, 1:100 dilution, Santa Cruz, CA, USA), BMP-receptor IA (1:100 dilution, Santa Cruz), osteopontin (1:50 dilution, IBL) and collagen type I (1:25 dilution, Fuji Yakuhin). The PAP method was used for osteocalcin (1:100 dilution, LSL), osteonectin (1:1000 dilution, LSL) and bone sialoprotein (BSP, 1:1000 dilution, LSL). These sections were then immersed in methanol containing 0.3% hydrogen peroxide for 30 min to block endogenous peroxidase and rinsed in 0.05 M TBS (5 min, 3 times)