

Case Report Article

Bilateral maxillary osteoplasty with esthetic purpose in patient with secondary hyperparathyroidism-related brown tumor: case report

Bruna da Fonseca Wastner¹
Vitor Hugo Candido Ferreira¹
Juliana Lucena Schussel^{1, 2}
Roberta Targa Stramandinoli-Zanicotti¹
Laurindo Moacir Sassi¹

Corresponding author:
Bruna da Fonseca Wastner
Erasto Gaertner Hospital
Rua Guilherme Tragante, 377 – Tarumã
CEP 82800-090 – Curitiba – PR – Brasil
E-mail: bru.wastner@hotmail.com

¹ Department of Oral and Maxillofacial Surgery, Erasto Gaertner Hospital – Curitiba – PR – Brazil.

² Department of Stomatology, Universidade Federal do Paraná – Curitiba – PR – Brazil.

Received for publication: September 15, 2015. Accepted for publication: December 18, 2015.

Keywords: brown tumour; secondary hyperparathyroidism; chronic kidney disease.

Abstract

Introduction and Objective: To report a case of a patient with secondary hyperparathyroidism that developed brown tumour in bilateral maxilla surgically managed with bilateral osteoplasty for aesthetic reasons. **Case report:** A 46 years-old woman patient with chronic kidney disease with complain of a painless bilateral growth on the maxilla, beginning 10 years before that ceased after surgical removal of the parathyroid eight years before. A CT scan showed a diffuse and heterogeneous bone lesion, with lytic areas and increased multilobulated volume on the maxilla, bilaterally. The surgery was an osteoplasty of the maxilla bone and performed under general anaesthesia. At 16-month follow-up, no recurrence was observed and the patient is satisfied with aesthetic and functional results. **Conclusion:** Secondary hyperparathyroidism related with oral brown tumour is a rare condition that affects function and aesthetic. The case reported in this paper was appropriately managed, achieving patient expectation and improving her life quality.

Introduction

Brown tumours are erosive bone lesions caused by rapid osteoclast activity and trabecular fibrosis related to primary or secondary hyperparathyroidism, resulting in a local destructive phenomenon [7].

Secondary hyperparathyroidism occurs due chronic renal failure that leads to an excessive secretion of parathyroid hormone (PTH) by the parathyroid glands in response to hypocalcaemia, hyperphosphatemia and hypertrophy of those glands [3]. These lesions develop mainly on the maxillomandibular complex, usually presented as well-defined lesions, circumscribed and osteolytic [9].

Histologically, they are characterized by giant cells in a fibre-vascular stroma with cyst-like spaces lined by connective tissue and *foci* of haemorrhage. These areas appear as a red-brown crumbly mass, which gives the name "brown tumour" [2]. Thus, this study aims to report a case of brown tumour of hyperparathyroidism surgically managed in a patient with chronic renal failure in dialysis, reviewing relevant concepts on the subject.

Case report

Female patient, brown, 46 years old, attended the Department of Oral and Maxillofacial Surgery

of Erasto Gaertner Hospital with complain of a painless bilateral growth on the maxilla, beginning 10 years before that ceased after surgical removal of the parathyroid eight years before because of an adenoma [6]. The patient has chronic kidney disease diagnosed in 1996 and is in dialysis 3 times a week ever since, besides hepatitis C virus and hypertension treated with medication. Oral examination revealed tumefaction with bone consistency, covered by normal mucosa, with approximately 10cm in bilateral maxilla (figure 1). Pre-surgical laboratory tests showed altered urea, creatinine and phosphorus. The calcium levels, PTH, and alkaline phosphatase were within normal limits. A CT scan of the facial sinuses showed a diffuse and heterogeneous bone lesion, with lytic areas and increased multilobulated volume on the maxilla, bilaterally, being more evident on the right side (figure 2). The treatment option was a bilateral osteoplasty performed under general anaesthesia (figures 3 and 4). The histopathological examination showed bony trabeculae surrounded by fibrous connective tissue, compatible with bone neoformation. The patient is in regular follow-up; no recurrence was observed 16 months after the treatment and the patient is satisfied with aesthetic and functional results (figure 5).



Figure 1 - Initial face and intraoral aspect

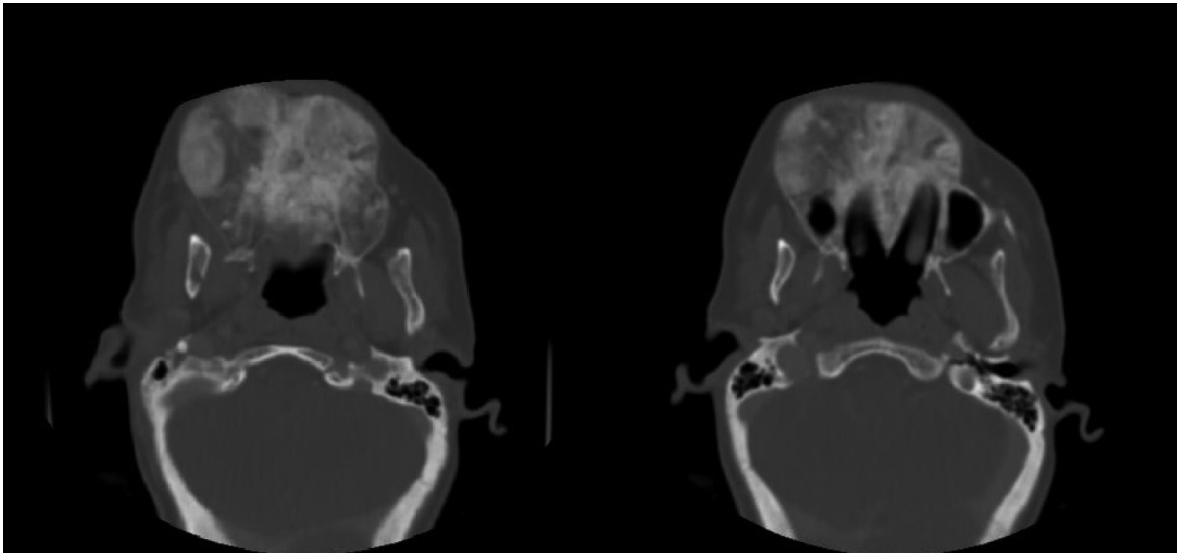


Figure 2 - CT at axial view showing the extension of the lesion

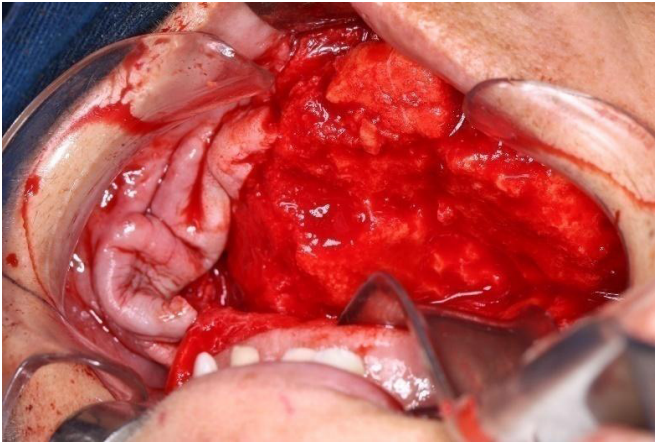


Figure 3 - Trans-operative aspect



Figure 4 - Immediate post-operative aspect



Figure 5 - Face and intraoral view at 16-month follow-up

Discussion

According to the Brazilian Society of Nephrology (SBN), worldwide, 500 million people suffer from kidney problems and 1.5 million of them are on dialysis. Statistics also reveal that one in ten people in the world suffer from chronic kidney disease [6].

Failing kidneys do not convert enough vitamin D to its active form, and do not adequately excrete phosphate. When this happens, insoluble calcium phosphate forms in the body and removes calcium from the circulation. Both processes lead to hypocalcaemia and secondary hyperparathyroidism [3]. Over time, the parathyroid glands become hyperplastic and less susceptible to treatment with calcium and calcitriol, producing a continuous secretion of PTH and resulting secondary hyperparathyroidism [3].

All individuals with chronic kidney disease have some degree of hyperparathyroidism, since changes in the parathyroid gland occur early in the development of renal impairment, even before the patient requires dialysis. Secondary hyperparathyroidism is a frequent problem and must be monitored [8].

Brown tumours affect especially females above 50 years old and the preferred site is maxilla, similar to our patient, with varying degrees of aggressiveness and risks of recurrence. Usually, brown tumors are painless, unless their dimensions are large enough to compress nerve structures [5]. The asymmetry frequently caused by the brown tumour in the jaws also causes a negative impact on patient's social life and can compromise breathing and swallowing [9]. In a same way, the patient described in this paper reported difficulty on swallowing.

The clinical management of this lesion aims primarily to reduce the elevated parathyroid hormone levels through pharmacological treatment [3], although surgical removal of parathyroid is considered to be curative. Surgical removal of the tumour may be required sometimes due to large size or when lesions do not resolve completely after PTH control [1]. The case reported here presented a lesion with large proportions that stopped growth after removal of the parathyroid glands, according to what was previously described by Pinto *et al.* [6], but not sufficiently regressed in size, resulting in aesthetic and functional sequelae for the patient. Histopathological analysis showed a tissue in process of regeneration.

In certain anatomical sites, decompression of the brown tumour is recommended to control

lesion expand and prevent destruction of adjacent structures. This is particularly true for maxillary lesions, which may lead to serious deformities of the face and even a lethal outcome [4]. On the other hand, it is necessary to consider that the surgical approach may cause loss of bone segments, teeth, neural lesions and more aesthetical and functional alterations. On the case reported in this paper, the lesion was no longer active and an osteoplasty was proposed to improve aesthetic and functional aspects of the patient.

In conclusion, secondary hyperparathyroidism related with oral brown tumour is a rare condition that affects functional and aesthetic aspects of the patient. The case reported was appropriately managed, achieving patient expectation and improving life quality.

References

1. Daniels JS. Primary hyperparathyroidism presenting as a palatal brown tumor. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2004;98:409-13.
2. Di Daniele N, Condò S, Ferrannini M, Bertoli M, Rovella V, Di Renzo L et al. Brown tumour in a patient with secondary hyperparathyroidism resistant to medical therapy: case report on successful treatment after subtotal parathyroidectomy. *Int J Endocrinol.* 2009.
3. Kangfeng L, Zhou C, Xuezhi Z, Yukun Z, Dechang Z, Guanhu Y et al. Secondary hyperparathyroidism associated with multiple brown tumor: a case report. *Journal of Medical Colleges of PLA.* 2011;26:360-66.
4. Pinar Sumer A, Arik N, Sumer M, Karagoz F. A rare complication of secondary hyperparathyroidism. Brown tumor of the maxilla and mandible. *Saudi Med J.* 2004;25:2010-20.
5. Pinto MC, Sass SMG, Sampaio CPP, Campos DS. Tumor marrom em paciente com hiperparatireoidismo secundário à insuficiência renal crônica. *Braz J Otorhinolaryngol.* 2010;76(3):404.
6. Sociedade Brasileira de Nefrologia (SBN). [cited 2015 Jul 20]. Available from: URL:<http://www.sbn.org.br>.
7. Suarez-Cunqueiro MM, Schoen R, Kersten A, Klisch J, Schmelzeisen R. Brown tumor of the mandible as first manifestation of atypical parathyroid adenoma. *J Oral Maxillofac.* 2004.

8. Tominaga Y. Parathyroidectomy for patients with renal osteodystrophy. *Clin Calcium*. 2003;13:340-3.

9. Triantafyllidou K, Zouloumis L, Karakinaris G, Kalimeras E, Iordanidis F. Brown tumors of the jaws associated with primary or secondary hyperparathyroidism. A clinical study and review of the literature. *American Journal of Otolaryngology - Head and Neck Medicine and Surgery*. 2006;27:281-6.