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Brown Tumors: A Case Report and Review of the Literature

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Key Words
Brown tumors · Secondary hyperparathyroidism · Craniofacial involvement · Treatment

Abstract
Brown tumors are focal bone lesions, encountered in patients with uncontrolled hyperparathyroidism. They can be located in any part of the skeleton. Clinically significant lesions in the craniofacial bones are rare. Craniofacial involvement may cause facial disfiguration and compromise social ease of the patient and normal functions, such as chewing, talking, and breathing. In this case report, we present a patient with a brown tumor of the craniofacial bones provoked by secondary hyperparathyroidism and review the last 10 years of craniofacial brown tumors associated with secondary hyperparathyroidism in the English literature.

Introduction

Brown tumors are focal bone lesions, caused by increased osteoclastic activity and fibroblastic proliferation, encountered in patients with uncontrolled hyperparathyroidism (HPT). They can be located in any part of the skeleton, but are most frequently encountered in the ribs, clavicles, extremities, and pelvic girdle. Clinically significant lesions in the craniofacial bones are rare [1].

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Here, we present a patient with a brown tumor of the craniofacial bones provoked by secondary HPT. When we searched PubMed for English literature of the last 10 years, there were 26 cases of craniofacial brown tumors associated with secondary HPT. Of the 26 cases, full texts of 24 cases were available and a summary of these cases is shown in table 1.

Case Report

A 30-year-old Albanian female patient was referred to our hospital for living donor kidney transplantation. She was unable to walk without help and was restricted to a wheelchair. Her medical history included chronic renal insufficiency, hypertension, anemia, and hepatitis C. She had been undergoing dialysis three times per week for 19 years. A tumor was protruding through her oral cavity; it had appeared insidiously 10 years ago and had increased gradually in size. The lesion was fixed to the underlying mandible, was tender to the touch, and was covered with oral mucosa. The teeth were severely affected, and bleeding sites were observed on the fragile mucosa (fig. 1). The patient had functional problems with chewing and speech. Serum chemistry revealed an elevated parathyroid hormone (PTH) level of 2,930.6 pg/dl (normal range, 15–65 pg/dl), serum calcium 9.08 mg/dl (normal range, 8.8–11 mg/dl), phosphorus 4.2 mg/dl (normal range, 2.5–5.0 mg/dl), and alkaline phosphatase (ALP) 1,753 IU/l (normal range, 65–300 IU/l).

A sagittal magnetic resonance imaging scan of the head showed a brown tumor originating from the mandibular and maxillary bones (fig. 2). Ultrasonography of the neck revealed no parathyroid mass; however, a scan with 20 mCi Tc-99-sestamibi showed increased uptake by the parathyroid glands (fig. 3). Based on the medical history, clinical manifestations, and laboratory tests, the final diagnosis was brown tumor with HPT. A whole-body scan with 20 mCi Tc-99m-HDP was performed to look for multifocal disease, which may have been missed. However, only maxillary and mandibular accumulation was observed.

A total parathyroidectomy was performed without implantation of a parathyroid fragment into the forearm muscle. PTH was monitored intraoperatively, and the PTH level decreased (355 pg/dl). A histopathological examination of the mass confirmed the diagnosis of parathyroid adenoma. Serum PTH level continued to decrease after surgery, and the laboratory findings 1 month postoperatively were PTH 127.2 pg/dl, ALP 247 IU/l, calcium 8.4 mg/dl, and phosphorus 2 mg/dl.

The lesion failed to regress 6 months postoperatively, despite optimal treatment for HPT. Kidney transplantation was performed with a living donor kidney from her cousin and creatinine levels decreased to normal postoperatively. No complications were observed during the first postoperative week, and the patient was discharged from the hospital.

Discussion

Here, we report a case of a huge brown tumor originating from the mandible and protruding through the oral cavity, which was provoked by secondary HPT. This tumor is of the longest duration reported among brown tumors provoked by secondary HPT that affected craniofacial bones (table 1). In fact, these tumors are not malignant, and there are options for management. Treating the HPT is the first step, and normalizing PTH level with drugs, dialysis, parathyroidectomy, or kidney transplantation will often cause the tumor to regress or resolve [1]. Surgical resection of a brown tumor is generally not recommended and should only be considered if the patient wants quick resolution, if the bony lesion is com-

promising body functions or promoting facial deformation, or if the lesion fails to regress after 1–2 years of follow-up [2, 3]. The 10-year duration of our case was too long, reflecting the inadequacy of preventing and managing brown tumors in underdeveloped countries.

Brown tumors may be located in any part of the skeleton. Involvement of the mandible, maxilla, palate, nasal cavity, paranasal sinuses, and orbital and temporal bones has been reported [1–15]. The mandible is more commonly affected than the maxilla, and these tumors are usually asymptomatic except when large. Brown tumors may cause facial disfiguration and compromise social ease of the patient and normal functions, such as chewing, talking, and breathing. Other complications include headaches, visual impairment, proptosis of the eyes, displacement and mobility of the teeth, and nasal or intraoral bleeding [1–15]. The huge tumor in our case originated from the mandible and caused teeth to fall out and severely affected chewing. Talking and the appearance of our patient were also affected. This tumor caused surgical difficulties due to its location. Nasotracheal intubation and a tracheotomy were necessary for the surgery, but our team completed the procedure successfully.

Conclusion

This case was unique considering the severity and the absolute lack of response to parathyroidectomy. Despite the parathyroidectomy and optimal control of calcium/phosphate metabolism, the brown tumor did not decrease in size during follow-up, which was confirmed by computed tomography of the lesion. The mass remained also unchanged 1 year after transplantation and the surgical option will be assessed during the follow-up of the patient.

Statement of Ethics

The authors have no ethical conflicts to disclose. Written informed consent was obtained from the patient for publication of this case report and the accompanying images.

Disclosure Statement

There are neither conflicts of interest nor financial support to declare.

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Table 1. Cases of craniofacial brown tumors associated with secondary HPT

Study (first author)		Age, years	F/M	HD duration, years	Tumor age, months	Location	iPTH	Ca^{2+}	ALP	PO_4	Treatment	Response
1	Leal 2006 [2]	31	F	9	8	Maxilla	3,086	8.4	1,333	7.1	Total parathyroidectomy and 2 years later local excision of the tumor	Tumor regressed, able to breathe and feed, improvement of patient's appearance
2	Zwick 2006 [3]	29	M	–	1	Frontal calvarium and orbital wall	450	9.6	–	7.3	Complete excision of the tumor	–
3	Triantafillidou 2006 [4]	70	F	–	–	Mandible	412.5	–	–	–	Local excision of the lesion	No recurrence
4	Triantafillidou 2006 [4]	68	F	–	–	Mandible	389.2	–	–	–	Local excision of the lesion	No recurrence
5	Triantafillidou 2006 [4]	21	F	–	–	Mandible	481.61	–	–	–	Local excision of the lesion	Recurrence after 1 year, 7 years after kidney transplantation there was no tumor
6	Tarrass 2008 [1]	18	M	6	2	Mandible	1,335	8.2	568	5.7	Subtotal parathyroidectomy	Progressive decrease in the size of tumor
7	Karabekmez 2008 [5]	11	M	4	–	Maxilla and mandible	2,528	8.4	1,869	9.7	–	Died before operation
8	Monteiro 2009 [6]	40	F	7	24	Orbit					Removal of parathyroid glands	Symptoms and signs disappeared
9	Di Daniele 2009 [7]	40	F	–	–	Maxilla	1,700	10.5	319	5.3	Total parathyroidectomy and implantation of parathyroid fragment	Regression of the tumor, but with residual hyperostosis
10	Fatma 2010 [8]	19	F	144	–	Mandible	870	2.27	2,706	2.08	Subtotal parathyroidectomy	Regression of the tumor
11	Fatma 2010 [8]	37	F	120	–	Mandible	3,687	2.29	–	1.8	Subtotal parathyroidectomy, local excision of the lesion	Insufficient regression, so excision of the tumor
12	Fatma 2010 [8]	57	F	88	–	Maxilla	1,500	2.25	945	1.98	Subtotal parathyroidectomy	Regression of the tumor
13	Fatma 2010 [8]	30	F	CKD	–	Mandible	1,115	2.13	2,493	2.38	Total parathyroidectomy, implantation of parathyroid fragment	Regression of the tumor
14	Fatma 2010 [8]	29	M	84	–	Maxilla	1,450	2.63	628	2.63	Subtotal parathyroidectomy	Regression of the tumor
15	Fatma 2010 [8]	32	F	84	–	Maxilla	1,142	2.56	318	2.56	Subtotal parathyroidectomy	Regression of the tumor
16	Fatma 2010 [8]	52	F	216	–	Maxilla	1,700	2	568	2	Subtotal parathyroidectomy, local excision of the lesion	Insufficient regression, so excision of the tumor
17	Pinto 2010 [9]	37	F	8	4	Maxilla and mandible	1,927	–	1,831	–	Total parathyroidectomy	Regressed significantly but at 18 months remains stable
18	Nabi 2010 [10]	24	F	10	2	Maxilla and ipsilateral paranasal sinus	1,591	3.26	352	0.88	Total Parathyroidectomy	Significant regression
19	Jakubowski 2011 [11]	49	F	10	12	Mandible	–	–	–	–	–	–
20	Pechalova 2013 [12]	19	M	6	–	Maxilla and mandible	1,409.3	–	–	2,204	Local excision	–
21	Pechalova 2013 [12]	19	F	6	–	Maxilla	2,595.8	–	–	3,227	Local excision	–
22	Artul 2013 [13]	46	F	11	No	Maxillary frontal bone	1,282	8.5	406	4.1	Medical	–
23	Verma 2014 [14]	31	F	CKD	13	Mandible	234.1	14.3	1,963	–	Referred for treatment in higher medical center	–
24	Jafari-Pozve 2014 [15]	29	M	8	3	Mandible, zygoma, maxilla, palate	3,552	8.7	2,800	6.3	Parathyroidectomy	Symptoms relieved

F = Female; M = male; HD = hemodialysis; CKD = chronic kidney disease; iPTH = intact PTH; Ca^{2+} = calcium; PO_4 = phosphorus.

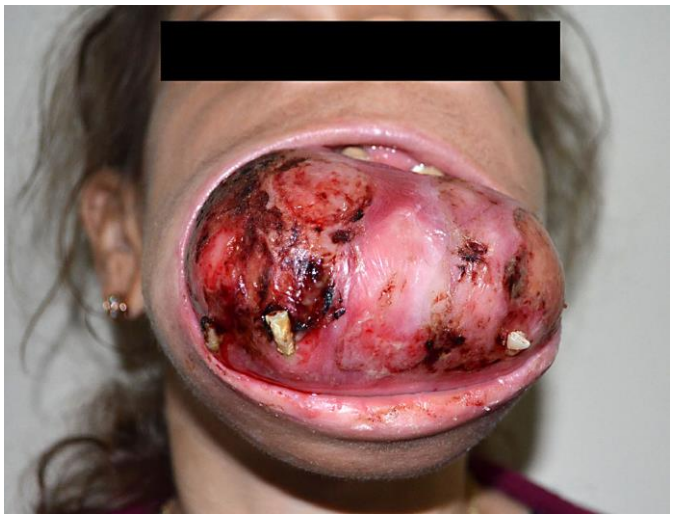

Fig. 1. Apperance of the brown tumor before surgery.

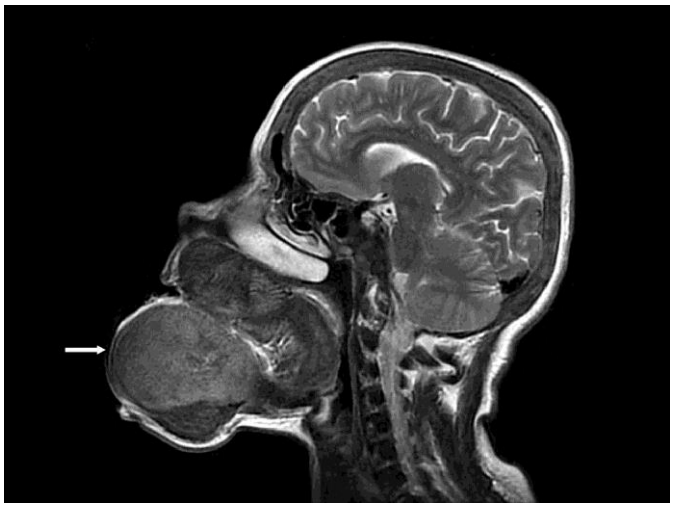

Fig. 2. Sagittal magnetic resonance imaging scan showed that the brown tumor originated from the mandibular and maxillar bones (white arrow).

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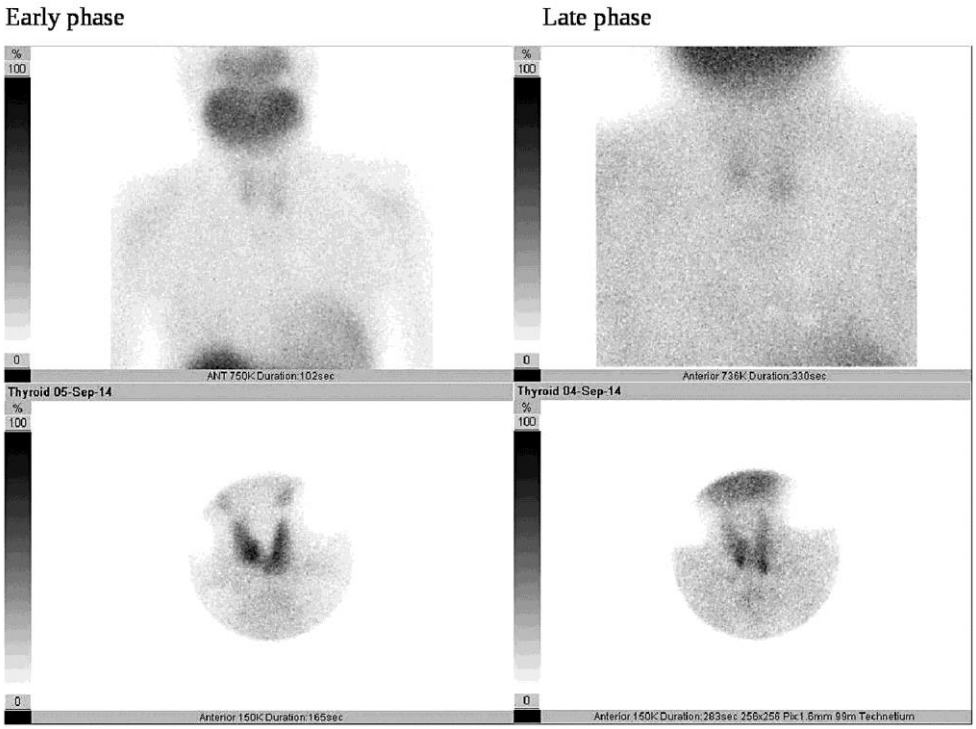

Fig. 3. Parathyroid scan showed increased uptake in the parathyroid glands.