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Original article

Maxillofacial brown tumours: Series of 5 cases

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ABSTRACT

Objectives: Brown tumours are benign bone tumours secondary to hyperparathyroidism. The authors describe the various clinical features, diagnostic methods and treatment modalities for maxillofacial brown tumours.
Material and methods: This multicentre retrospective study comprised 5 patients (four women and one man, between the ages of 29 and 70 years) with one or several maxillofacial brown tumours observed over a 16-year period from January 2000 to December 2016.
Results: Four patients presented secondary hyperparathyroidism in a context of chronic renal failure, one patient presented primary hyperparathyroidism due to parathyroid adenoma. Three patients presented a mandibular brown tumour, and two patients presented a maxillary brown tumour. The diagnosis was based on histological examination and laboratory tests. Brown tumours were treated either surgically or conservatively. A favourable outcome was observed in all cases.
Conclusion: Brown tumours are rare lesions. This diagnosis must be considered in a context of giant cell tumour associated with hyperparathyroidism. Brown tumours should be treated conservatively.

1. Introduction

Brown tumour is a rare entity in otorhinolaryngology, belonging to the group of giant cell tumours and resulting from reorganization of the bone matrix exposed to excessive parathormone secretion in the context of either primary or secondary hyperparathyroidism [1]. Brown tumours are non-neoplastic lesions predominantly arising in long bones and the axial skeleton [2].

The positive diagnosis is based on histological examination and laboratory tests [3]. The main differential diagnosis of brown tumour is central giant cell granuloma [3]. Treatment essentially comprises correction of hyperparathyroidism. These tumours have a favourable prognosis [4,5].

In the light of this case series and a review of the literature, we describe the clinical features, laboratory findings, clinical course and treatment of head and neck and maxillofacial brown tumours.

2. Material and methods

This multicentre retrospective study was conducted at the Grand Ouest University Hospitals over a 16-year period from January 2000 to December 2016.

When suspected clinically, the diagnosis of brown tumour was based on a combination of biopsy with histological examination in favour of giant cell tumour and a laboratory work-up showing hyperparathyroidism (parathormone (PTH) > 38 pg/ml). Personal data were collected for each patient: age, sex, history, including the type of hyperparathyroidism and its aetiology. Data concerning the tumour were also collected: site, number, treatment and postoperative complications. Patient follow-up was mainly clinical, with data recorded one year after the end of treatment.

3. Results

Five patients meeting the inclusion criteria were identified: two at Angers university hospital, two at Tours university hospital and one at Rennes university hospital. Patient characteristics are summarized in Table 1.

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Table 1
Summary of the study population characteristics.

Patients	1	2	3	4	5
Sex/age	F/43	F/70	F/29	M/29	F/30
Tumour site	Mandible	Maxilla	Hard palate	Mandible	Mandible
Time to diagnosis	8 months	4 months	3 months	2 months	3 months
Treatment	Tumour resection	Conservative	Tumour resection	Conservative	Tumour resection
Postoperative complications	Yes	No	Yes	No	No
1-year follow-up	Complete regression	Complete regression	Complete regression	Complete regression	Complete regression
Hyperparathyroidism	II	I	II	II	II
Parathyroid surgery	Not-	Selective resection of the adenoma	Subtotal parathyroidectomy	Subtotal parathyroidectomy	Subtotal parathyroidectomy

M: male; F: female, I: primary; II: secondary.

3.1. Case 1

A 42-year-old woman with treated hepatitis C and dialysed for chronic renal failure of unknown origin, in whom two suspicious lesions of the body of the mandible were discovered on panoramic dental X-rays.

These two lesions were completely resected under general anaesthesia without prior biopsy, but with frozen section examination in favour of giant cell tumours. The postoperative course was marked by right mandibular abscess formation, requiring surgical revision and antibiotic therapy with a subsequent favourable outcome.

In this context of chronic renal failure, a calcium phosphate assessment was repeated, revealing secondary hyperparathyroidism with PTH of 686 pg/ml. The diagnosis of mandibular brown tumours was proposed in the light of these results.

A favourable outcome was observed at 6 months and 1 year with no recurrence or new tumours.

3.2. Case 2

A 70-year-old woman with a history of renal colic and hypertension observed asymmetry of the cheekbones for one year.

On clinical examination, the patient presented a hard swelling of the anterior wall of the right maxilla, filling the vestibule, with no other associated signs. CT scan of the facial bones visualized a destructive bone tumour invading the maxillary sinus with heterogeneous tissue density, with rupture of the cortex (Fig. 1).

Transoral biopsy was performed as an outpatient procedure. Histological examination was in favour of giant cell tumour. In this context, a calcium phosphate assessment was performed, revealing primary hyperparathyroidism (PTH 270 pg/ml, serum calcium 3.48 mmol/l).

The aetiological work-up of this hyperparathyroidism, comprising ultrasound and parathyroid scintigraphy, revealed a 4 cm macro-adenoma in the left lower pole (Fig. 2).

A diagnosis of maxillary brown tumour secondary to primary hyperparathyroidism in a context of parathyroid adenoma was proposed.

Management comprised medical treatment to correct hypercalcaemia by bisphosphonates and intravenous fluid therapy, and surgical resection of the adenoma.

Clinical follow-up 6 months after parathyroidectomy was favourable with marked regression of the tumour and complete disappearance at 1-year follow-up.

3.3. Case 3

A 29-year-old woman, with a history of renal transplantation for lupus nephropathy, consulted for progressive palatine swelling for several months.

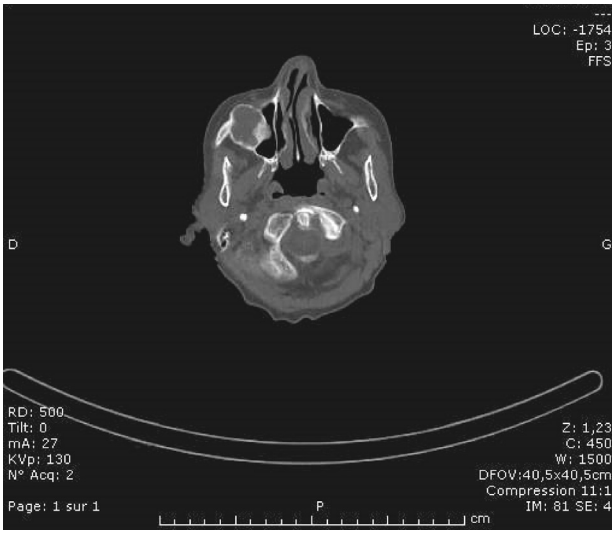

Fig. 1. Axial CT scan of the facial bones, bone window setting, showing a tumour invading the right maxillary sinus, rupturing the cortex.

Clinical examination revealed a swelling of the right hard palate with osteolysis of the floor of the nasal cavities on nasal endoscopy, compressing the inferior turbinate. The rest of the clinical examination was normal. CT scan of the facial bones revealed a heterogeneous lesion with a long axis of 2 cm in the right hard palate, with osteolysis of the floor of the nasal cavities and the base of the septum.

Transoral resection under general anaesthesia was performed with frozen section examination in favour of central giant cell granuloma. The postoperative course was marked by major feeding difficulties requiring maintenance of a nasogastric tube for 7 days. The zone of resection was allowed to heal by secondary healing.

In view of this patient's history of chronic renal failure, a calcium phosphate assessment was performed, revealing features of secondary hyperparathyroidism (PTH 650 pg/ml; serum calcium 2.06 mmol/l). A diagnosis of brown tumour was therefore proposed. A 7/8th parathyroidectomy was performed, completed several years later by left inferior parathyroidectomy due to persistently elevated PTH, allowing correction of calcium phosphate parameters.

Clinical follow-up 1 year after transoral resection revealed no signs of recurrence.

3.4. Case 4

A 20-year-old man with a history of renal transplantation for Alport syndrome with known tertiary hyperparathyroidism due to chronic renal failure. The patient consulted for rib and bilateral mandibular bone tumours.

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