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A rare occurrence of primary hyperparathyroidism with brown tumor in the left maxilla

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ABSTRACT

Brown tumors are rare skeletal manifestations of hyperparathyroidism. It is a form of osteitis fibrosa cystica caused by hyperparathyroidism due to excessive secretion of parathyroid hormone. Then, hypercalcemia results in excessive osteoclastic activity in the bones. Brown tumors are considered a rare complication of hyperparathyroidism, as their reported existence is only about 1.5-4.5%. Herein, we report a rare case of brown tumor arising from the left maxilla due to primary hyperparathyroidism, its challenge in diagnosis with giant cell tumor, and its literature review.

Introduction

A brown tumor is a rare, non-neoplastic, reactive bone lesion resulting from excessive parathyroid hormone (PTH) secretion. Histologically, brown tumors are characterized by masses of giant cells surrounding the fibrovascular stroma, cystic spaces in connective tissue, foci of hemorrhage associated with microfracture, and subsequent release of hemosiderin, which gives them their name (1). One of the difficulties encountered in dealing with brown tumors is that they may mimic giant cell tumors (GCT), yet both are different in management. In our cases, the initial workout diagnosis was a GCT of the left maxilla, which raised suspicion when her routine blood investigation revealed hypercalcemia, and the investigation later showed a raised PTH level.

Case Presentation

A 47-year-old female patient with no known comorbidities was admitted with a 1-month history of left maxillary swelling, redness, and tenderness. She had no episode of arrhythmia because her cardiac workout was normal. There were no

gastrointestinal manifestations or any evidence suggesting nephrolithiasis.

Clinical examination revealed a hard left maxillary swelling of approximately 7x6 cm with central ulceration involving the left hard palate crossing the palatal midline. There was no palpable neck swelling.

Subsequent skull X-rays showed an opacified mass in the left maxillary region up to the temporal region. Computed tomography scan revealed a well-defined heterogeneously enhancing soft tissue mass extending into the left maxillary sinus cavity associated with the destruction of the maxillary sinus floor and the inferior part of the nasal septum.

Hyperparathyroidism came to our attention when her blood test revealed a high calcium level of 3.18 mmol/L. Subsequent investigation of intact PTH (i-PTH) revealed a raised level of 79.43 pmol/L.

However, her renal profile was normal, with a urea level of 6.1 mmol/L and a creatinine level of 74 µmol/L. Her phosphate level was 0.52 mmol/L. Thus, we concluded that our patient had primary hyperparathyroidism.

Unfortunately, her left maxillary lesion caused ulceration and recurrent bleeding. Thus, the patient underwent left maxillectomy. Histopathological examination revealed hypercellular proliferation of plump spindled and fibroblastic cells with aggregates of multinucleated giant cells in the presence of large blood-filled cystic spaces, hemorrhage, abundant hemosiderin deposits, and lymphoplasmacytic infiltrates.

Postoperatively, we performed an ultrasound on the neck, which showed a multinodular goiter with a TIRADS-4 nodule at the isthmus, sized at 1.2x2.5x2.2 cm. We then performed a technetium-99m sestamibi scan, which showed evidence of a functioning adenoma at the superior thyroid lobe. The patient was counseled for total thyroidectomy and superior parathyroidectomy; however, she refused to proceed with the suggested surgical intervention and lost follow-up.

Discussion

Primary hyperparathyroidism is more common in women and African Americans than in men and other racial groups. Most cases (>80%) of primary hyperparathyroidism are asymptomatic and are incidentally detected during routine blood investigations. Bandeira et al. (2) reported in a study of 124 patients at their center that 25% showed severe skeletal involvement and osteitis fibrosa cystica (OFC) and 25% had nephrolithiasis. However, a recent cross-sectional study by Eufrazino et al. (3) found that only 6.1% of patients manifested OFC, whereas 18.2% had nephrolithiasis.

Histologically, the present case showed hypercellular proliferation of spindled and fibroblastic cells, multinucleated giant cells, an area of blood-filled cystic spaces, hemorrhage foci, and abundant hemosiderin deposition. This histological manifestation may also mimic GCT, as it shows aggregates of multinucleated GCTs along with a degree of hemosiderin deposition, and some may exhibit a degree of atypia (4).

Clinical and radiological brown tumors and GCTs are difficult to differentiate as both may exhibit clinically exophytic brownish mass and expansile lytic bone lesions with intralesional trabeculation in radiological findings. GCT may favor its growth toward long bones, whereas brown tumors are more favored toward the pelvis, ribs, clavicles, or extremities (5). Thus, increased calcium and PTH levels remained a hallmark of differentiated brown tumors from GCTs.

In our case, the patient presented with a rare brown tumor in the left maxilla. Oral manifestation of brown tumors is considered rare, and tumors involving the oral cavity may exhibit exophytic growth with a high tendency to bleed. Lajolo et al. (6) described in a systematic review that the mandible was the most affected side of intraoral brown tumors (66.2%), with most cases presenting as single lesions (59.5%). When maxillary involvement is detected, it is important to consider the risk of multiple oral lesions.

Patients with tumors that affect their function or quality of life should undergo surgical treatment. There was less evidence-based discussion regarding whether to proceed with parathyroidectomy first, then tumor regression, or undergo tumor resection along with parathyroidectomy in the same setting. There have been cases reported, such as those by Nabi et al. (7), that reported regression of maxillary brown tumors after total parathyroidectomy. Other cases reported by Oliveira et al. (8) showed a large left orbital cavity and part of a brown nasal tumor measuring 8.1 cm x 6 cm x 5.1 cm that regressed after removal of the parathyroid adenoma.

In our case, tumor progression caused episodes of ulceration, obstructing the maxillary sinuses, and recurrent bleeding, requiring multiple hospital admissions. Thus, we first decided to perform a left maxillectomy to improve her quality of life.

Conclusion

Primary hyperparathyroidism with a brown color remained a challenge in management. Early detection and intervention for primary hyperparathyroidism significantly decreased the incidence of brown tumors. The existence of bone mass associated with hypercalcemia should raise suspicion of hyperparathyroidism.

Ethics

Informed Consent: Consent form was filled out by a participant.

Authorship Contributions

Surgical and Medical Practices - Concept - Design - Data Collection or Processing - Analysis or Interpretation - Literature Search - Writing: M.A.M.F., S.R.H.I.M., M.M.Y., M.F.O.

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References

1. Triantafillidou 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. Am J Otolaryngol. 2006;27:281-286.
2. Bandeira F, Griz L, Caldas G, Bandeira C, Freese E. From mild to severe primary hyperparathyroidism: The Brazilian experience. Arq Bras Endocrinol Metabol. 2006;50:657-663.
3. Eufrazino C, Veras A, Bandeira F. Epidemiology of Primary Hyperparathyroidism and its Non-classical Manifestations in the City of Recife, Brazil. Clin Med Insights Endocrinol Diabetes. 2013;6:69-74.
4. Rosenberg AE, Nielsen GP. Giant cells containing lesions of bone and their differential diagnosis. Curr Diagn Pathol. 2001;7:235-246.
5. Rossi B, Ferraresi V, Appetecchia ML, Novello M, Zoccali C. Giant cell tumor of bone in a patient with diagnosis of primary hyperparathyroidism: a challenge in differential diagnosis with brown tumor. Skeletal Radiol. 2014;43:693-697.
6. Lajolo C, Patini R, Limongelli L, et al. Brown tumors of the oral cavity: presentation of 4 new cases and a systematic literature review. Oral Surg Oral Med Oral Pathol Oral Radiol. 2020;129:575-584.
7. Nabi Z, Algailani M, Abdelsalam M, Asaad L, Albaqumi M. Regression of brown tumor of the maxilla in a patient with secondary hyperparathyroidism after a parathyroidectomy. Hemodial Int. 2010;14:247-249.
8. Oliveira FM, Makimoto TE, Scalissi NM, Marone MM, Maeda SS. Regression of orbital brown tumor after surgical removal of parathyroid adenoma. Arch Endocrinol Metab. 2015;59:455-459.