Case Report

Upper alveolar brown tumor as initial presentation of parathyroid adenoma

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ABSTRACT

Parathyroid adenoma usually presents with the signs and symptoms of hypercalcemia due to hyperparathyroidism. Brown’s tumor is the late clinical consequence of untreated hyperparathyroidism and is rarely seen now-a-days due to improved screening biochemical tests. These tumors are characterized by high orthoclastic activity and bone resorption. These are usually located in the pelvis, ribs, clavicles, and extremities. This manuscript highlights a rare presentation of brown’s tumor of upper alveolus in a 35-year-old female with a previously undiagnosed case of hyperparathyroidism due to a parathyroid adenoma. Blood analysis depicted high levels of serum calcium and parathyroid hormone. The patient was subjected to selective left lower parathyroidectomy and will receive follow-up to prevent further developments of the disease. Clinicians should be aware of such rare presentation of parathyroid adenomas so that unnecessary extensive surgery of brown’s tumor is avoided. This case also highlights the need of detailed work up to arrive at an exact diagnosis and direct surgical intervention to the adenoma rather than the secondary manifestation of the disease.

Key words: Brown’s tumor, parathyroid adenoma, parathyroid hormone

INTRODUCTION

A parathyroid adenoma is the most frequent cause of primary hyperparathyroidism and usually presents as signs and symptoms of hypercalcemia. Patients with primary hyperparathyroidism are usually diagnosed incidentally as hypercalcemia, but less frequently, the clinical presentation includes renal calculi, osteoporosis, and neuropsychiatric symptoms, and rarely, peptic ulcer disease or pancreatitis.1,2 Bone involvement in primary hyperparathyroidism is usually a late manifestation and due to improved screening techniques such as serum calcium and parathormone levels is rarely seen now-a-days in the developed world.3,4 The bony manifestation of prolonged hyperparathyroidism results in osteitis fibrosa cystica or brown tumor formation characterized by high osteoclastic activity and bone resorption and are usually located in the pelvis, ribs, clavicles, and extremities. This may mimic as primary bone neoplasm or metastatic disease and might induce confusion between the two diagnoses.5-8 Brown tumors are named so due to their characteristic brown appearance of hemosiderin and fibroblastic tissue penetrating into the gaps created in the bone matrix by the increased osteoclastic activity. This increased osteoclastic activity causes expansion of the bone beyond its normal contours. We report a rare case of previously

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undiagnosed primary hyperparathyroidism due to a parathyroid adenoma with the initial presentation as brown’s tumor of the upper anterior alveolus. This case highlights the importance of awareness in clinicians of such rare presentation of parathyroid adenomas. This will avoid unnecessary extensive surgery of the brown tumor, which responds to the parathyroid adenoma removal as it normalizes the parathyroid hormone (PTH) levels.

**CASE REPORT**

A 35-year-old female presented to our head and neck unit with a progressive swelling of the upper anterior alveolus of 6 months duration. She denied any history of trauma, toothache, and fever. The patient did not have any facial numbness but described mild pain and heaviness in the upper jaw, which compelled her to seek medical attention. She was, otherwise, systemically well and reported no other associated symptoms such as abdominal pain, renal colic, or pain in other bones. No cervical lymphadenopathy could be palpated. Local examination revealed brownish mass involving anterior upper alveolus and hard palate causing mal occlusion of the jaws [Figure 1]. The swelling was noted to be firm, but upon palpation did not appear to be of bony origin. Maximal mouth opening was normal for the patient. Serum biochemistry revealed a serum calcium level of 2.94 mmol/L (normal range: 2.2–2.6 mmol/L), PTH level of 101 pg/ml (normal range: 10–65 pg/ml) with alkaline phosphatase, renal, and liver profiles within normal limits. Ultrasonography examination showed 1.2 cm × 0.8 cm well-defined solid hypoechoic lesion inferior to the left lobe of the thyroid gland and medial to the carotid artery. On Tc-MIBI double phase parathyroid scintigraphy, the initial image showed homogenous tracer distribution in both lobes of the gland and a focal intense tracer uptake inferior to the left lower pole of thyroid [Figure 2]. Delayed images after 2 h showed normal washout of MIBI from thyroid bed with significant tracer retention in the inferior parathyroid region suggestive of an adenoma [Figure 3]. The patient underwent a selective excision of the left parathyroid adenoma [Figure 4]. Histology revealed a circumscribed tumor composed of nests and islands of mainly chief cells without evidence of mitoses. Postoperative period was uneventful. Serum calcium on the first postoperative day was 2.32 mmol/L (normal range: 2.2–2.6 mmol/L) and PTH level 45 pg/ml (normal range: 10–65 pg/ml). At the 9 months follow-up clinic review, blood tests revealed normal range calcium and PTH levels. Clinical examination showed a good resolution of the jaw tumor. She is regularly followed in our head and neck clinic.
DISCUSSION

The brown tumor is not a neoplasm but may be mistaken as a primary bone tumor and subjected to unnecessary surgical intervention. It is rare to find a brown tumor as the first clinical manifestation of primary hyperparathyroidism before the onset of general manifestations—like our case. A biopsy may be needed to exclude malignancy. The brown tumor was suspected on the basis of the appearance of the lesion and serum biochemistry revealed elevated serum calcium and PTH level. Therefore, the knowledge of such an entity will not only help clinicians to diagnose such cases but also avoid unnecessary surgical intervention.

The management of brown tumor due to parathyroid adenoma is purely surgical directed to the removal of the parathyroid adenoma rather than surgical intervention at the tumor site. Spontaneous complete regression of brown tumors has been reported in 90% of the cases within 4–20 months after parathyroidectomy. Regression is slower in case of patients with age more than 60 years and if brown’s tumors are in cancellous bones due to slower bone turnover. However, the surgical intervention of the brown tumor may be necessary in some exceptional situations, especially in case of skull base involvement with compression of vital structures such as an optic nerve.

CONCLUSION

We present a case of brown tumor in a young woman whose long-standing hyperparathyroidism had not been recognized in time. The brown tumor may be mistaken for neoplasm and diagnosis of underlying hyperparathyroidism gets delayed. Extensive surgery of the brown tumor is not indicated except in a situation of compression of vital structures such as optic nerve at the skull base. Blood tests for calcium and PTH levels are essential to reaching the correct diagnosis.

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Conflicts of interest
There are no conflicts of interest.

REFERENCES

1. Fraser WD. Hyperparathyroidism. Lancet 2009;374:145-58.
2. Rodgers SE, Lew JJ, Solórzano CC. Primary hyperparathyroidism. Curr Opin Oncol 2008;20:52-8.
3. Damjanov I, editor. The endocrine system. In: Pathology for the Health-related Professions. 2nd ed. Philadelphia: W.B. Saunders; 2000. p. 403-20.
4. Su AW, Chen CF, Huang CK, Chen PC, Chen WM, Chen TH. Primary hyperparathyroidism with brown tumor mimicking metastatic bone malignancy. J Chin Med Assoc 2010;73:177-80.
5. Pai M, Park CH, Kim BS, Chung YS, Park HB. Multiple brown tumors in parathyroid carcinoma mimicking metastatic bone disease. Clin Nucl Med 1997;22:691-4.
6. Lu G, Shih WJ, Xiu Y. Technetium-99m MIBI uptake in recurrent parathyroid carcinoma and brown tumors. J Nucl Med 1995;36:811-3.
7. Miyakoshi M, Kamoi K, Takano T, Nishihara M, Kawashima T, Sudo N, et al. Multiple brown tumors in primary hyperparathyroidism caused by an adenoma mimicking metastatic bone disease with false positive results on computed tomography and Tc-99m sestamibi imaging: MR findings. Endocr J 2007;54:205-10.
8. Meng Z, Zhu M, He Q, Tian W, Zhang Y, Jia Q, et al. Clinical implications of brown tumor uptake in whole-body 99mTc-sestamibi scans for primary hyperparathyroidism. Nucl Med Commun 2011;32:708-15.
9. Reséndiz-Colosia JA, Rodríguez-Cuevas SA, Flores-Díaz R, Juan MH, Gallegos-Hernández JF, Barroso-Bravo S, et al. Evolution of maxillofacial brown tumors after parathyroidectomy in primary hyperparathyroidism. Head Neck 2008;30:1497-504.
10. Emin AH, Süoğlu Y, Demir D, Karatay MC. Normocalcemic hyperparathyroidism presented with mandibular brown tumor: Report of a case. Auris Nasus Larynx 2004;31:299-304.
11. Daniels JS. Primary hyperparathyroidism presenting as a palatal and mandibular brown tumor. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;98:409-13.
12. Diamanti-Kandarakis E, Livadas S, Tseleni-Balafouta S, Lyberopoulos K, Tantalaki E, Palioura H, et al. Brown tumor of the fibula: Unusual presentation of an uncommon manifestation. Report of a case and review of the literature. Endocrine 2007;32:345-9.