Abstract

Objectives: Brown tumors are a definitive feature of hyperparathyroidism, whether primary or secondary. They are well-documented osteolytic lesions commonly in the appendicular skeleton. They are composed of giant cells within a fibrovascular stroma with foci of hemorrhage from which they get their color and name. Primary hyperparathyroidism at present is most often suggested by hypercalciemia and hypophosphatemia on routine lab tests. Much more rarely do these cases present with a craniofacial mass. Here we detail a case presenting with a growing mass along the maxilla associated with loosened dentition.

Study Design: A case report and literature review.

Methods: We investigate a unique presentation of terminal stage primary hyperparathyroidism emphasizing the importance of a broad differential diagnosis and thorough work-up.

Results: 45 year old female with several months of a growing mass of the left maxilla now with loose teeth, pain, and bleeding. She details a 2-year history of diffuse bone pain, headaches and mood swings. Labs include calcium of 14.4 mg/dL, phosphorous 2.0 mg/dL, and PTH of 1200 pg/mL. 4D parathyroid CT scan demonstrates 1.5 cm hyperenhancing mass subjacent to the right thyroid gland and multiple lytic lesions within the mandible and maxilla with arterial phase hyperenhancement. Biopsy of the maxilla was suggestive of a brown tumor. Parathyroidectomy retrieved a 1.5 gram parathyroid adenoma with subsequent laboratory and clinical improvement.

Conclusions: Hyperparathyroidism can present in very unique ways. As an otolaryngologist in the frontline we must think beyond just tissue diagnosis so that appropriate and expedited care may be implemented.

Case Presentation

45 year old Hispanic female with arthritis presents with enlarging mass of her maxillary alveolar ridge. The mass has been growing for several months and is now associated with loose dentition. She is now experiencing pain in the region as well as intermittent bleeding. Further, her gingiva are nubm. She notes diffuse bone pains since 2013 mostly in the knees. She has problems with constipation, headaches and mood swings. She denies nephrolithiasis or overt psychosis / delusions. She has a history of cholecystectomy. Takes Norco for pain. Denies any tobacco or ETOH. Her family history is unremarkable.

PHYSICAL EXAM:
Firm mass on buccal surface of maxillary alveolar ridge adjacent to #16. Loose dentition of right mandibular canine, #27. Neck is without lymphadenopathy.

CLINICAL PRESENTATION: Some maxillary and malar prominence (< L < R). 1 cm exophytic, erythematous mass centered around root of tooth #16 in the left maxilla. Tender to touch with some bleeding.

CT FACE WITH CONTRAST: Coronal and axial sections demonstrate multiple mandibular and maxillary expansile, lucent enhancing lesions with extraosseous extension and some with internal matrix calcification. With a multi-focal presentation, the differential diagnosis may include metastases, myeloma with plasmacytomas, multiple odontogenic tumors, or brown tumors (osteitis fibrosa cystica), although with the markedly elevated calcium, brown tumors (osteitis fibrosa cystica) was the main differential consideration.

Lab tests:
Ca: 14.4 (9.0 – 10.3) mg/dL.
Phos: 2.0 (2.4 – 4.7) mg/dL.
Mg: 1.6 (1.8 – 2.5) mg/dL.
Albumin: 3.3 (3.5 – 4.8) gm/dL.
PTH 1200 (15 – 65) pg/mL.

Discussion

Primary hyperparathyroidism is caused by a single parathyroid adenoma 80% of the time, with multiple adenomas seen in 2-4% of cases. 15% of cases are secondary to 4 gland disease, or hyperplasia. 4D parathyroid CT imaging has dramatically improved our ability to pre-operatively differentiate between these conditions. Bony changes are a characteristic feature of PHPT with cortical bone affected more than cancellous bone. Brown tumors, occur in a very small percentage of patients and may prompt their presentation.

Treatment of brown tumors consists of reversal of hypercalcemia via parathyroidectomy or medical therapy. Natural progression of brown tumors following correction of primary or secondary hyperparathyroidism is to regress. Rosende-Ceballos et al followed 22 patients with brown tumors resulting from primary or secondary hyperparathyroidism. 18 cases had complete regression of bony lesions by 10 months on average following treatment. More cystic changes and bony destruction tends to portend a worse prognosis for complete resolution and older age slows recovery.

Curettage or local excision for improved aesthetics may be warranted down the road. If the lesion is symptomatic, deforming, or fails to regress after correction of hypercalcemia, surgical resection of the brown tumor should be reconsidered. Intralosseal corticosteroid injections or systemic steroids as a means of reducing tumor size and easing surgical resection has been reported. In this case, correction of hypercalcemia via removal of parathyroid adenoma led to near complete resolution of the clinically visible maxillofacial lesions and symptoms by 6 months.

Conclusion

As a clinician we are trained to recognize patterns of data. In regards to brown tumors as a primary presentation of PHPT it is critical to consider the entire data set. No one piece of information is singularly adequate. We desire to hasten treatment but must ensure we hasten what is right and best for the patient.

References


Answer: None. All are Brown Tumors.

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Which is cancer?
See below references for answer.

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