Case Report

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Brown tumor of hyperparathyroidism of anterior maxilla: case report & literature review

Jahangir Hammad, Farhat Gul Babar, Kashaf-ud-doja Tariq, Muhammad Ayoub

[1]Associate professor, oral and Maxillofacial surgery, Sandeman Provincial hospital, BUMHS, Quetta
[2-4]Resident oral and Maxillofacial surgery, Sandeman Provincial hospital, BUMHS, Quetta

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Address of Correspondent
Dr. Kashaf-ud-doja Tariq
Resident oral and Maxillofacial surgery, Sandeman Provincial hospital, BUMHS, Quetta
memahmood1945@gmail.com

A B S T R A C T

Hyperparathyroidism is characterized by the overproduction of parathyroid hormone (PTH). Hyperparathyroidism can be Primary, secondary and tertiary. In all these forms of hyperparathyroidism there is osteoclast mediated bone resorption which is caused by excess Parathyroid hormone levels that produce bone lesion called brown tumor of hyperparathyroidism. Brown tumor of Hyperparathyroidism is a benign non-odontogenic giant cell lesion of the jaw with multinucleated giant cells. The term brown tumor is derived from the brown color of the lesion as seen on surgical exploration. The characteristic discoloration is result of hemosiderin production. The brown tumor regress usually after hyperparathyroidism has been successfully corrected. Its surgical excision is usually not recommended until the lesion is largely symptomatic, at risk of pathological fracture and recurrence. All the giant cell lesion are identical histologically and must be distinguished based on clinical, radiographic and biochemical means. Here we present a case of Brown tumor that reoccurred in 13 years old girl in the anterior maxilla.

Keywords: Brown Tumor, Hyperparathyroidism, Resection.

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Introduction

Brown tumor of hyperparathyroidism derived its name from the color of tissue that is due to hemosiderin deposition, it is also called osteitis fibrosa cystica because of fibrous displacement of bone marrow.[1,2] Most common sites of brown tumor are ribs, clavicle, pelvic girdle and in maxillofacial region palate, nasal cavity, paranasal sinuses, [3,1] orbital, and temporal bones, mandible and uncommonly maxilla.[3,4] In case of jaws patient presents with complain of facial asymmetry, localized jaw pain, swelling, bleeding from gingiva and loose teeth.[5]

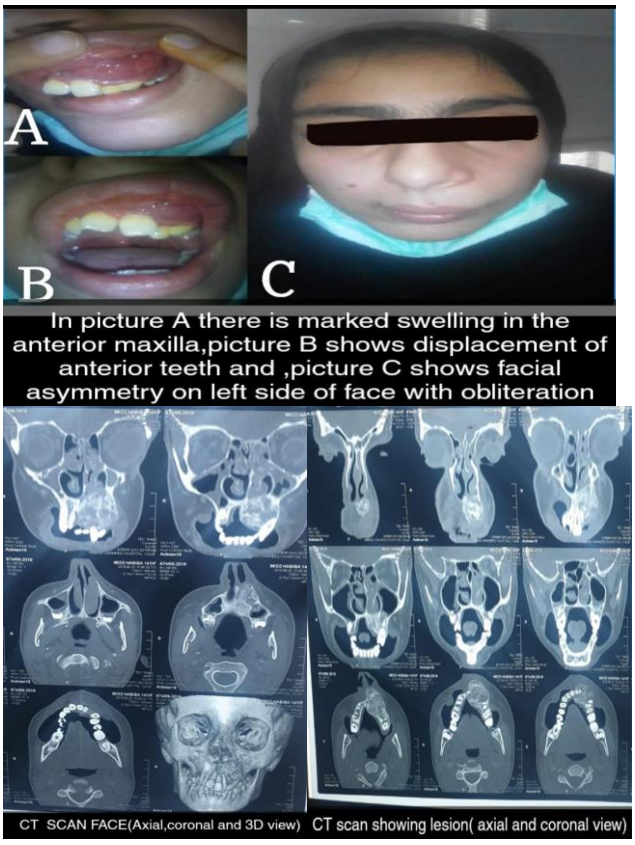
Primary hyperparathyroidism is due to parathyroid adenoma, hyperplasia, and adenocarcinoma associated with hypercalcemia. Its clinical features are weakness, fatigue, depression. Bone manifestations are bone pain, bone resorption, cortical bone loss if facial bone involves then facial swelling and asymmetry. renal manifestations include renal stones, polyuria, gastrointestinal features are abdominal pain, peptic ulcer, pancreatitis[1,6] Whereas secondary hyperparathyroidism is due to physiological demand for hormone in response to hypocalcemia in chronic renal failure, vitamin D deficiency, malabsorption and rickets. Initially there is hypocalcemia due to lack of absorption of vitamin D that activate the parathyroid gland to secrete PTH and its continuous stimulation increase PTH and cause hypercalcemia thus patient develops secondary hyperparathyroidism.[6] Tertiary hyperparathyroidism is due to the development of parathyroid tumor against a background of prolonged secondary hyperparathyroidism.[7]

Case Report

A 13 years old girl came to oral and maxillofacial surgery department 1 year back with the complaint of painless swelling on anterior maxilla for the last 6 months although inflammatory features such as fever, chills, etc,

were absent. Past medical, drug, surgical, dental and family history was insignificant. In general, the physical examination patient was vitally stable, with no signs of anemia, jaundice, clubbing, and cyanosis present. Extra orally there was no significant findings related to swelling, no palpable lymph nodes and cranial nerves were intact. Intraorally hard, and non-tender swelling of approximately 2 cm in size on anterior maxilla with normal overlying mucosa was present. Investigations for Parathyroid hormone calcium & vitamin D level estimation, parathyroid Scintigraphy and, face 3D CT scan were advised. Incisional biopsy was done, in which sample was taken from the maxillary vestibule, histopathological findings were a variable number of multinucleated giant cell, hemosiderin within the vascular and proliferative fibroblastic background that confirmed Brown tumor of hyperparathyroidism Her PTH ,calcium levels were raised with deficiency of vitamin D. i.e. 184ng/L(PTH), 9.5mg/dl Ca and D3 4ng/ml. The patient was informed about the disease, its consequences, medical treatment and surgical excision. Patient was not willing for any surgical procedure and opted for medical treatment. The patient was referred to the general physician to manage hyperparathyroidism, injection calcitonin, and cap Indrop - D was prescribed to decrease PTH, increase vitamin D level & reduce swelling to some extent. The patient was kept on observation and was convinced by the physician for surgical intervention when swelling continue to increase in size.

The patient again came after 1 year with the same complaint of painless swelling on the same site that was progressively increased in size. After general physical examination patient was thoroughly examined extra orally and intraorally. There was marked facial asymmetry on left side along with bulging upper lip and obliteration of nasolabial fold. Intraorally mouth opening was adequate, hard, non-tender swelling of approximately 4cm size was present extending superiorly to the floor of nasal cavity, inferiorly palate, anteriorly to central incisor and posteriorly to the second premolar, associated teeth were displaced. The same investigations advised previously, was repeated Ultrasound neck was done to exclude tumor of the parathyroid gland and with patient consent, surgery was planned. For surgery, PTH level was controlled up to normal with the help of endocrinologist. Under General anesthesia incision was made in left maxillary vestibule extending from central incisor to mesial side of the last molar, blunt dissection was done, the lesion was explored as the patient was young the visible lesion was excised whereas for remanant of lesion curettage and peripheral ostectomy was done Associated teeth were extracted. BIPP Dressing was placed and wound was then left open for secondary healing No intraoperative complication was reported. Analgesics and antibiotics were prescribed, vitamin D, calcium level maintained to normal level. Patient was advised to take soft diet and was recalled weekly for 3 months for follow up. Patient was satisfied with her surgical treatment as there was only mild postoperative pain that subsided with time and normal facial symmetry no other late postoperative complications reported, rehabilitation with obturator was done.



Discussion

Here we report a case of brown tumor of maxilla due to secondary hyperparathyroidism that was reoccurred after 1 year as initially no surgical intervention was done due to patient unwillingness.so in that case, surgical removal of the brown tumor became essential as it took a long time to resolve and to prevent its complication. Brown tumor of hyperparathyroidism may involve any part of the skeleton. but in maxillofacial region mandible, palate, nasal cavity, paranasal sinuses, maxilla orbital and temporal bone may be affected. The mandible is more commonly affected than the maxilla[5], unlike this case.

these tumors are usually asymptomatic except when large. Brown tumors may cause facial asymmetry, difficulty in mastication, talking, failure to socialize, Other complications include headaches, visual impairment, proptosis of the eyes, displacement and mobility of the teeth, and nasal or intraoral bleeding depending upon the site involved.[5] Brown tumor of hyperparathyroidism is histologically identical to cherubism, giant cell tumor, aneurysmal bone cyst, central giant cell granuloma.[8] radiographically resembles ossifying fibroma, jaw osteoma of Gardner syndrome and multiple odontogenic keratocyst as part of nevoid basal cell carcinomas.[6]

The diagnosis of the brown tumor should be based on elevated serum calcium, alkaline phosphatase, and parathyroid hormone levels, low serum phosphate levels, and histological features. Computed tomography or bone scans help make a differential diagnosis and identify the extent of the lesion.[9] Serum calcium, serum PTH, serum phosphate helps to differentiate Brown tumor of hyperparathyroidism from other giant cell lesions. Parathyroid Scintigraphy, neck ultrasound can be used to diagnose parathyroid adenoma.

There are different treatment options for brown tumor of hyperparathyroidism depending upon the type of hyperparathyroidism, but in all types first, step involve treatment of hyperparathyroidism, and normalizing PTH level with drugs, dialysis, parathyroidectomy, or kidney transplantation depending upon the cause.[4] Mostly brown tumor will regress after hyperparathyroidism has been corrected.[6] Other options are curettage, resection or radiotherapy to prevent reoccurrence of tumor that are large and symptomatic.

The Brown tumor of primary hyperparathyroidism is treated with surgical removal of abnormal parathyroid tissue and normalization of abnormal parathyroid levels.[6,9] The significant bone disease associated with secondary hyperparathyroidism may be prevented or reduced by medical treatment, such as calcium carbonate, vitamin D, and aluminum hydroxide antacids for hyperphosphatemia.[10] For our case of secondary Hyperparathyroidism with brown tumor in the maxilla, we decided to remove the tumor because that had enlarged intraoral swelling leads to functional problems like difficulty in mastication and speech and facial asymmetry.

Conclusion

Brown tumor is an expansile osteolytic lesion caused by hyperparathyroidism. this case was unique considering the unresponsiveness to medication, recurrence and site that is ncommonly involve in the Brown tumor of hyperparathyroidism**.** It also emphasizes on the importance of history, investigations and patient compliance in proper diagnosis and management of Brown tumor as it resembles other giant cell lesions.

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