Brown Tumor of Primary Hyperparathyroidism Mimicking a Periapical Lesion

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Abstract: Brown tumors are benign giant cell lesions associated with hyperparathyroidism which can be primary, secondary or tertiary. It usually occurs in the long bones: tibia, femur, clavicle, pelvis rim, ribs and the mandible, but it is rarely involved in the maxilla. It can therefore be the first sign to diagnose an hyperparathyroidism. A rare case of maxillary brown tumor mimicking a periapical lesion in a 32-year-old woman was reported. This case highlights the importance of a detailed examination in case of giant cell lesion, including the oral cavity and the whole body in order to detect eventual systemic problems and to emphasize the necessity of taking into consideration the differential diagnosis of periapical lesions.

Keywords: brown tumor, hyperparathyroidism, giant cell lesion, periapical lesion, parathyroid adenoma.

INTRODUCTION

The oral cavity is often the seat of various manifestations that represent an alerting sign to different general disorders such as brown tumor, which is a benign giant cell lesion that arises as a result of hyperparathyroidism.

We reported a rare case of maxillary brown tumor in a 32-year-old female patient that emphasizes the necessity of a careful examination for osteolytic lesions in order to precisely determine the diagnosis and to adapt the adequate therapeutic approach.

CASE REPORT

A 32-year-old female patient presented to the department of oral medicine and oral surgery at the university clinic of dental medicine of Monastir with a chief complaint of a gradual swelling in the upper left vestibule which evolves for four months. Her family and past medical history were non-significant. The extra oral examination was normal.

The intra oral examination revealed a poor oral hygiene and a swelling on the buccal surface of the left maxillary alveolar ridge adjacent to the left maxillary canine (23), which was nontender to palpation and hard in consistency. The mucosa above the swelling was normal. (Figure 1) It also showed the absence of the first left maxillary premolar and a necrosis of the 23.

The radiographic examination with orthopantomogram reveal a large well-defined solitary radiolucent image in the periapical region of the canine (Figure 2).

The facial computed tomography revealed a solitary hypodense well-demarcated mass in the maxillary left anterior region measuring approximately 2.37 by 1.48 cm, surrounding the apex of the left maxillary canine, and preserving the integrity of the maxillary sinus and the nasal cavity (Figure 3 A, B, C).

Many diagnoses can be proposed in case of such osteolytic lesion: inflammatory periapical lesion related to the necrotic left maxillary canine, aneurysmal bone cyst, giant cell granuloma, giant cell tumor and brown tumor.

First, an endodontic treatment of the left maxillary canine was performed.

Then, this lesion was excised under local anesthesia (figure 4 A, B, C). The specimen appeared as a red brownish friable mass with some hemorrhagic deposits inside (figure 5 A,B).

The histological examination showed a focal giant cell aggregation in loose-textured fibrous tissue with new bone formed, concluding to central giant cell granuloma (figure 6). However, in this case, it is recommended that a lesion secondary to
hyperparathyroidism must be ruled out as a matter of principle by performing a phosphocalcic assessment.

The vitamin D level and the renal function tests were within the normal limits, but there was a high level of calcium, phosphorus and PTH. A brown tumor associated with primary hyperparathyroidism was therefore suspected.

This etiology was confirmed by a neck sonography which showed a parathyroid nodule (figure 7). Other localisations of this nodule were excluded by a parathyroid technetium scintiscan (figure 8).

Surgical excision of the parathyroid nodule was performed and the histological examination showed that it consists in a parathyroid adenoma.

In a two month follow-up, a favorable mucosal healing was noted (figure 9) and the panoramic radiograph showed the beginning of bone remineralization (figure 10). After 11 months, we observed a perfect normal aspect of the mucosa and a complete mineralization of the bone lesion (figures 11, 12).

Fig-1: intra oral view showing swelling in the left side of the maxillary vestibule adjacent to the left canine

Fig-2: panoramic radiograph showing a large well-demarcated polygeodic radiolucent image in the left maxillary alveolar bone

Fig-3 A, B, C: CT scan showing a hypodense mass measuring 2.37 * 1.48 cm surrounding the periapical region of the maxillary left canine preserving the integrity of the nasal cavity and maxillary sinus. A: axial view of CT scan, B: coronal view of CT scan, C: sagittal view of CT scan
Fig-4 A, B, C: Surgical time: excision of the lesion. A: a mucoperiosteal flap rising, B: residual cavity, C: repositionning of the flap and sutures

Fig-5 A, B: specimen: a red brownish mass with hemosiderin deposits. A: specimen, B: specimen after section

Fig-6: Histological examination: Focal giant cell aggregation in loose-textured fibrous tissue with new bone formed (HE*100)

Fig-7: Neck sonography showing a right parathyroid nodule
Fig. 8: Parathyroid technetium scintiscan

Fig. 9: Two months follow up: intra oral view

Fig. 10: Panoramic radiograph after 2 months showing regression of the osteolytic lesion

Fig. 11: 11 months follow up intra oral view
DISCUSSION

Brown tumor is defined as a non-neoplastic giant cell lesion that involves the bone [1]. It is more frequent in patients aged over 50 years with a male to female ratio of 1/3[2-4].

This osteolytic lesion commonly develops in the long bones: ribs, clavicle, pelvic bones, tibia, femur and the mandible, but, it is rarely involved in the maxilla [3, 5, 6, 7]. It can occur only in the maxilla or in the mandible or it can involve the two bones³, uni or bilateral [8, 9]. Some atypical locations are described in the literature such as the maxillary sinus [8].

Clinically, this lesion is asymptomatic at the first stage [10], then as the osteolysis advances, gradually a swelling or a painful exophytic mass can be observed[11]. At the advanced stages, functional problems can be detected such as pathological fractures, teeth migration, root resorption, teeth mobility...

Radiologically, it appears as a well-defined monolocular or multilocular osteolytic lesion with no cortical disruption or periosteal reaction [2, 3, 11]. Histological features are characterized by cellular fibrous tissue containing multiple foci of hemorrhage, aggregations of multinucleated giant cells interspersed with hemosiderin deposits and sometimes reactive trabecules of woven bone [3, 5, 10, 11, 12, 13, 14].

Brown tumor is one of the bone manifestations of hyperparathyroidism, which is an endocrine disorder first described by Sylvanus[9], characterized by overproduction of parathyroid hormone PTH : it is an hormone secreted by the parathyroid glands involved in the regulation of calcium and phosphorus metabolism. In addition, it has a great role in bone mineralization [2, 12, 15].

Hyperparathyroidism may occur in primary, secondary or tertiary forms

For the primary form, hypersecretion of PTH results from adenoma (80%-85%) or hyperplasia (15-20%) or carcinoma of gland (<0.5%)[2,8,15]. Usually, this parathyroid problem is located in the posterior capsule of the thyroid, but it may have ectopic locations (1-3%). The most frequent ectopic one is the mediastinum [16]. For secondary hyperparathyroidism, the PTH excess is due to the low serum calcium levels caused by vitamin D deficiency or chronic renal failure [1, 2, 17]. Finally, the tertiary form is associated with renal failure and autonomous functioning of the glands after untreated long-standing untreated secondary form [1, 2, 12, 18, 19].

In fact, brown tumor results from imbalance between osteoclastic and osteoblastic activity subsequently to hypersecretion of PTH, leading to bone resorption and its fibrous substitution [2, 8, 10, 18]. As it is presented in this case, it may be difficult to diagnose brown tumor because it presents similar clinical, radiological and histological features to many other osteolytic lesions ranging from simple periapical lesion depending on the clinical and radiological aspects to many types of giant cell lesions like giant cell granuloma, cherubism, aneurysmal bone cyst and multiple myeloma, especially the presence of giant osteoclasts in the histological examination leads to confusion, therefore, only the clinical history and the endocrinologic status can confirm the diagnosis[1,2,3,5,20].

For the brown-tumor-treatment, it requires treatment of hyperparathyroidism including medical and surgical therapy [6, 12]. The medical component consists in the regulation of calcium and PTH levels, and the surgical one requires a parathyroidectomy which rapidly decreases the excessive amount of PTH, thus usually achieving complete regression of the bone lesion and remineralization [14].

The excision of brown tumor is not necessary unless the patient presents functional problems or the lesion is too large or if the bone lesion persists even after the parathyroid-lesion-removal [3, 5, 12, 13]. In our case, the excision of osteolytic lesion preceded the treatment of parathyroid problem because this lesion has led to the diagnosis of hyperparathyroidism.

CONCLUSION

Although brown tumor is not frequently the first alert to the early diagnosis of hyperparathyroidism, dental surgeons must be aware of the oral manifestations related to systemic diseases and of available online: http://sasjpjournals.com/sjds
importance and need for detailed clinical, radiological and laboratory tests for diagnosis and therapy success.

REFERENCES