

Brown Tumor of the Palate as First Manifestation of a Primary Hyperparathyroidism

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Abstract Brown tumors represent a late-stage bone change caused by primary hyperparathyroidism, presenting in only 2-3 percent of the cases. Therefore as primary manifestations there is even a smaller percent. The common sites of brown tumor are the long bones, ribs, clavicle or pelvic girdle. The solitary palate brown tumor as initial presentation of a primary hyperparathyroidism is rare and often accompanied by tumors of other facial bones. We present the case of a 67 year old woman with initial presentation of a large tumor of the right palate, with no extension to the orbits. The first histological diagnosis was of a giant cell tumor, which delayed the diagnosis of the underlying cause for about 6 months, when the patient's relatives requested a second opinion on the initial histological findings, the second analysis revealed the same diagnosis but recommended that a primary or secondary hyperparathyroidism should be excluded as the brown tumor is very similar to the one with giant cells. We received the patient at this moment when we immediately diagnosed a primary hyperparathyroidism and sent the patient to surgery. Unfortunately in this case the long progression of high levels of PTH affected a lot the mineral density of the bones and now the patient is highly osteoporotic. The particularity of the case is the location of the brown tumor and the delayed diagnosis of a first primary hyperparathyroidism.

Keywords: brown tumor, giant cell tumor, hyperparathyroidism, osteoporosis

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1. Introduction

Brown tumors also called osteoclastomas represent rather a reparative process than a neoplastic process that is attributed to hyperparathyroidism. Histologically they are practically identical to giant cell tumors so they can be easily misdiagnosed.

In primary hyperparathyroidism their incidence is about 2-3% but with the new diagnosis methods to identify hypercalcemia and hypophosphatemia we rarely see now this type of tumor.

The common sites of this tumor are the mandible, long bones, the clavicle or the pelvic girdle whereas the giant cell tumors are more common close to the knee joint either on the lower part of the femur or the proximal part of the tibia. The giant cell tumor are more common in women aged 20-40 years old, the brown tumors on the other side are also slightly more common in women but usually older than 40 years and they tend to increase with age.

The brown tumors can appear in both primary and secondary hyperparathyroidism and because secondary hyperparathyroidism is more common we tend to see more brown tumors in this second case of hyperparathyroidism.

Therefore the solitary palate brown tumor as initial presentation of a primary hyperparathyroidism is rare and often accompanied by tumors of other facial bones.

2. Case Presentation

We present the case of a 67 year old women that addressed her dentist in 2016 for a bump on her palate. The ortopantomography revealed a lesion on the right maxilla that implied the maxillary sinus. The exploration was completed by computer tomography which showed a large mass of 5.5 cm enhancing the maxillary sinus and the adjacent teeth. The patient was recommended immediate surgery.

A 5.5/4/4 cm maxillary bone fragment covered with skin on one side was excised, under the skin a brown solid tumor of 4.5/4 cm and also two adjacent teeth.

The histopathological exam showed either central giant cell lesion or granuloma. The microscopy revealed only giant cells and reactive bone tissue.

Although the brown tumor is very similar to the one with giant cells the anatomopathologist didn't recommend at that time that a hyperparathyroidism should be excluded as a possible cause.

Six months later the patient's relatives requested a second opinion on the initial histological findings, the

second analysis revealed the same diagnosis but recommended that a primary or secondary hyperparathyroidism should be excluded as the brown tumor is very similar to the one with giant cells.

That was practically the moment the patient addressed our service where she was found to have hypercalcemia (11.49 mg/dl, nv: 8.6-9.8 mg/dl) with normal phosphate, an elevated PTH of 334 pg/ml (nv: 12-67 pg/ml), normal alkaline phosphatase, and also a vitamin D deficit of 16.01ng/ml (nv: >30 ng/ml).

We were able to identify a small hypoechoic tumor of 1/1.2 cm above the left thyroidal lobe that was confirmed to be a parathyroid adenoma by the scintigraphy with Tc⁹⁹.



Figure 1. ultrasound of the left inferior parathyroid nodule

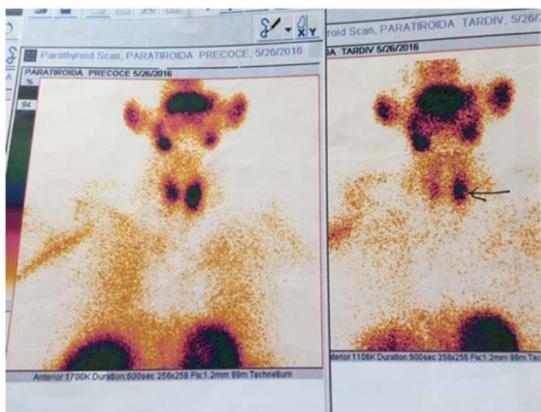


Figure 2. Scintigraphy using Tc⁹⁹ showing parathyroid adenoma

We also performed a osteodensitometry bone test which was consistent with our supposition of hyperparathyroidism showing typical bone resorption greater on the arm (Ts=-3.9) than on the vertebrae (Ts=-2.2) or the neck of the shoulder (Ts=-2).

The patient underwent surgery for the parathyroid adenoma with immediate normalization of the calcium levels and inhibition of PTH <3 pg/ml.

Three months later her PTH levels started to increase again 88 pg/ml but with low calcium levels and also a very low vitamin D status. So we were now facing a potentially secondary hyperparathyroidism so we decided to correct the vitamin d levels with a high Vitamin D dose of 4000U/day. 3 months later she had normal lab tests.

She also received recommendation treatment for osteoporosis with bisphosphonates and calcium supplement together with a high intake of phosphorus-rich aliments.

One year and a half later the patient readdressed our service. Her lab tests were typical for secondary hyperparathyroidism and she also had progression of osteoporosis.

Ca	8,4 mg/dl	8,6-9,8
Mg	1,4 mg/dl	2,5-4,5
P	3,9mg/dl	2,5-4,9
PTH	102 pg/ml	12-67
25 OH vit D	10,8 ng/ml	> 30

Figure 3. lab tests oct 2018

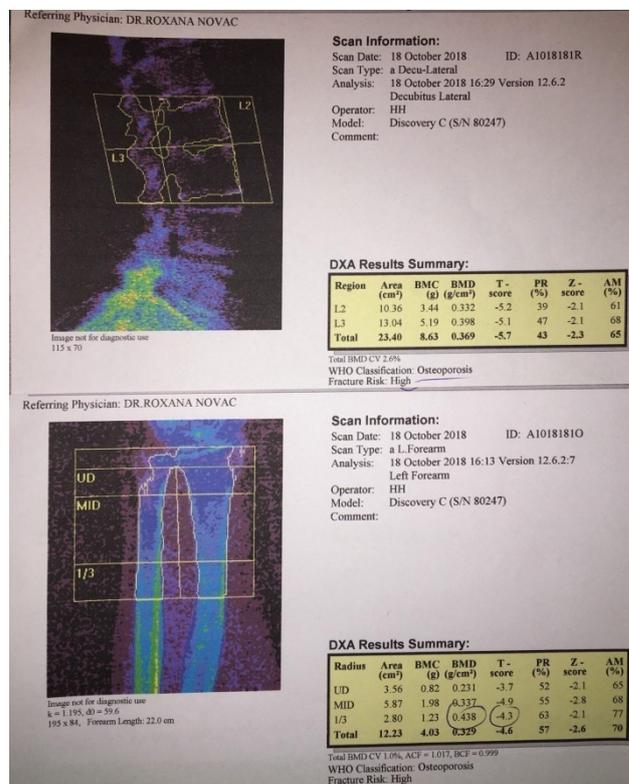


Figure 4. BMD-DXA of the lumbar spine (lateral-view) and arm

She admitted having stopped all treatment for one year and a half, so she was again recommended vitamin D supplements, bisphosphonates treatment and calcium supplements.

3. Discussions

We present the case of a brown tumor of the palate as first manifestation of a primary hyperparathyroidism in which the diagnosis of the hyperparathyroidism was delayed due to the confusion on histopathology with the giant cell tumor. [1]

This is a common confusion considering the resemblance between the two lesions, both of them showing giant cells and reactive bone tissue [2].

What was really interesting in this case was the development of a secondary hyperparathyroidism due to vitamin D deficit which is probably the most common form of secondary hyperparathyroidism and which we think in this case conducted to the progression of bone resorption [3,4,5].

4. Conclusion

We think it is mandatory to consider a form of hyperparathyroidism whenever facing a giant cell lesion on histopathological findings to avoid other complications due to the primary cause as advanced osteoporosis and renal impairment.

Conflict of Interest

No conflict of interest.

References

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