



Case report

Palatal swelling revealing a secondary hyperparathyroidism: A case report

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ABSTRACT

Introduction and importance: Secondary hyperparathyroidism is a frequent complication of end-stage renal diseases. It is characterized by elevated secretion of parathyroid hormone (PTH). This excessive secretion of PTH induces the formation of brown tumors frequently on long bones and axial skeleton but rarely in head and neck region.

Case presentation: We report a 31-year-old woman with a history of end-stage renal disease on dialysis who eating difficulties. Physical examination revealed a palatal swelling. Imaging and biology confirmed the diagnosis of palatal brown tumor due to secondary hyperparathyroidism.

Clinical discussion: Brown tumors are rare benign tumors caused by excessive parathyroid hormone secretion during hyperparathyroidism. Head and neck locations are unusual.

Conclusion: With better management of secondary hyperparathyroidism, brown tumors have become a rare entity. However, physicians should suspect them in order to prevent in time their possible mass effect.

1. Introduction

Secondary hyperparathyroidism is a frequent complication of end-stage renal diseases. It is characterized by elevated secretion of parathyroid hormone (PTH) to maintain a normal calcium homeostasis. In fact, people with chronic kidney disease tend to have high serum inorganic phosphorus and low serum calcium and calcitriol, which triggers the production and secretion of PTH [1]. Brown tumors are rare benign tumors due to reorganization of the bone matrix caused by exposure to excessive secretion of PTH, they usually occur on long bones and axial skeleton. Head and neck locations are unusual [2].

This article has been reported in line with SCARE criteria [3].

2. Presentation of case

A 31-year-old woman presented to our department with a 9-month history of an increasing palatal swelling which had become painful. She reported moderate eating difficulties causing slight weight loss. The patient's medical history was significant for an end-stage renal disease on dialysis three times a week for 7 years. She did not report a history of bone fractures.

Physical examination revealed a firm homogeneous palatal swelling, covered by a normal looking mucosa causing a deformation of the upper

dental arch (Fig. 1A). However, other bone deformities have not been found.

A head and neck CT scan was performed (Fig. 1B) showing a well-defined expanding osteolytic lesions in the palate, clivus, mandible, and odontoid, and a diffused decreasing bone density.

Laboratory examination showed a normal calcium level: 2.52 mmol/L (2.25–2.6), hyperphosphatemia: 1.83 mmol/L (0.74–1.52), an increased total alkaline phosphatase level: 1550 U/L (40–150) and an elevated parathyroid hormone level: 2580 pg/mL (26.5–96.5).

Based on physical examination, laboratory findings, and CT scan, we concluded that it was a brown tumor due to secondary hyperparathyroidism with low impact on eating and the patient was scheduled for a subtotal parathyroidectomy.

3. Discussion

Secondary HPT is a late complication of chronic renal disease principally induced by hyperphosphatemia and vitamin D deficiency. In fact, in case of renal failure, phosphorus clearance decreases leading to phosphorus retention which directly stimulates PTH secretion, causes moderate hypocalcemia, stimulates FGF-23 leading to severe inhibition of 1-alpha hydroxylase and a decrease in 1,25 dihydroxyvitamin D [1,4].

Brown tumors are rare benign tumors due to reorganization of the

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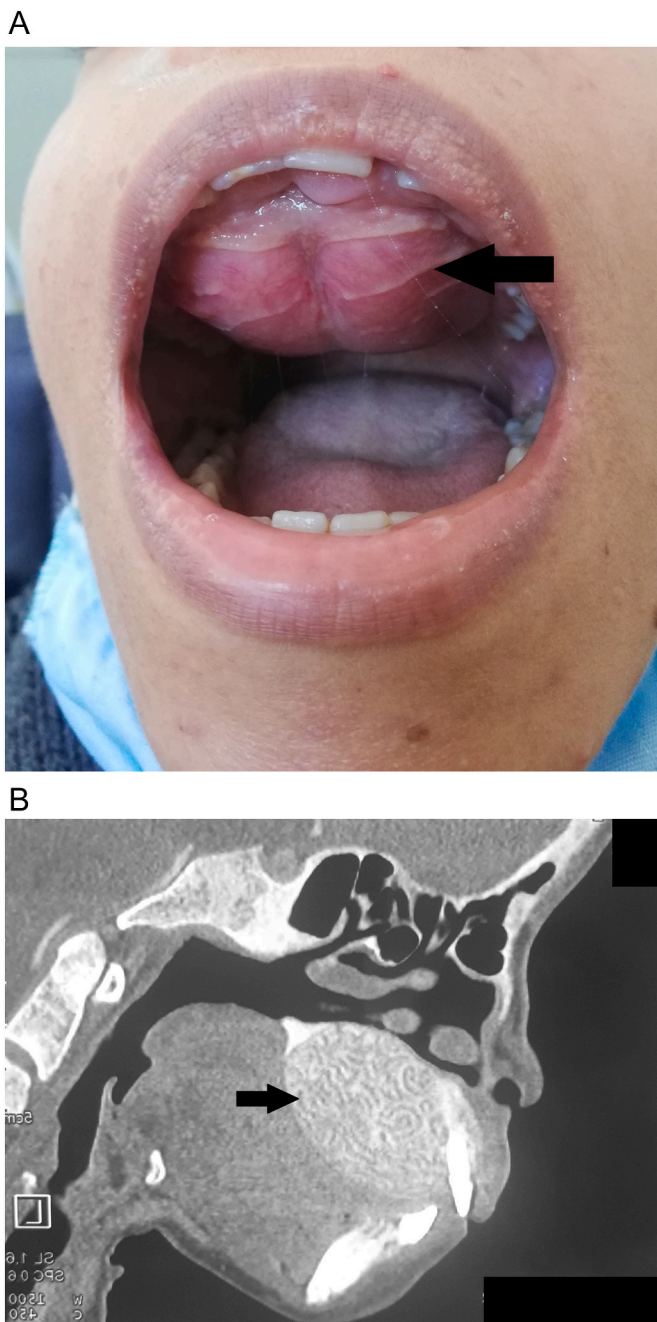


Fig. 1. A (Above) palatal swelling(arrow) B (Below) Sagittal CT scan of palatal brown tumor with well-defined expanding osteolytic lesions (arrows).

bone matrix caused by exposure to excessive parathyroid hormone secretion during hyperparathyroidism, either primary, secondary or tertiary. It was first described by Recklinghausen in 1891 as Osteitis fibrosa cystica. It occurs most frequently in the long bones and axial skeleton. Head and neck locations are unusual and not well known by physicians [5].

Clinically, palatal brown tumors can be completely asymptomatic or cause pain, facial deformity impairing normal functions such as chewing, speaking and breathing [6].

The radiological characteristics of brown tumors on plain radiograph and computed tomography consisted of well-defined single or multilobular osteolytic lesions with bone expansion and may be associated with bony destruction or a pathological fracture [7].

Differential diagnoses of a palatal swelling are benign and malignant

salivary gland tumors, lymphomas, immune-mediated diseases such as immunoglobulin G4 related disease and peripheral nerve tumors [8].

Brown tumors regress spontaneously, completely or at least partially after parathyroidectomy. Resection of the tumor can therefore be discussed if the patient wishes a rapid regression of the tumor, in case of facial deformity, if it compromises a body function by compression or if it does not regress after 2 years of follow-up [6,9].

4. Conclusion

With better management of hyperparathyroidism, brown tumors have become a rare entity. However, physicians, especially in cases of head and neck localization, should suspect them in order to prevent in time their possible mass effect on the upper aerodigestive tract.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

This study is exempt from ethical approval at our institution.

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Author contribution

Makram Tbini: Writing - Original draft.
Emna Essefi: Writing - Original draft.
Housseem Eddine Kamel: Writing - Review & editing.
Ines Riahi: Writing - Review & editing.
Mamia Bensalah: Supervision.

Guarantor

Makram Tbini.

Registration of research studies

Not applicable.

Declaration of competing interest

Not applicable.

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