J. Oral Diagn 2025;10:e316
https://doi.org/10.5327/2525-5711.316

CASE REPORT

Renal osteodystrophy in the oral and maxillofacial region: a case report and literature review

Laura Marina Demenech[1], Isadora Vilas Bôas Cepeda[1], Bernardo da Fonseca Orcina[1], Clarissa Fôlha Nachtigal[1], Caroline de Oliveira Langlois[2], Marcos Antonio Torriani[3], Antonio Cesar Manentti Fogaça[3], Adriana Etges[4,*]

Abstract:

This study presents a case report of a patient with chronic kidney disease (CKD) with secondary hyperparathyroidism and the presence of manifestations in the oral-maxillofacial complex with Renal osteodystrophy (ROD). Also, a literature review in the PubMed was carried out using the MeSH Terms descriptors (((jaws) AND (treatment)) AND (secondary hyperparathyroidism)) AND (case reports). The search was limited to articles written in English and published in the last 10 years. Also, this article presented the clinical case of a 49- year-old caucasian female patient, with volumetric increase in jaws after dental extraction. The patient reported having CKD for 10 years. Clinical examination of the patient revealed enlargement of the alveolar bone ridge in the maxilla and mandible. The maxilla showed an evident loss of palatal concavity, and the mandible loss of space at the bottom of the vestibular groove. The diagnosis of ROD was consolidated after the analysis of imaging tests, laboratory tests and histopathological evaluation of the bone tissue. The manifestation of systemic diseases in the face demands a systemic look, to establish treatments with the best prognosis. We reinforce the need for multidisciplinary work for the correct diagnosis and management of patients with systemic diseases.

Keywords: Renal osteodystrophy; Chronic kidney disease; Maxillary diseases.

INTRODUCTION

Chronic Kidney Disease (CKD) affects 752 million individuals worldwide[1] and 0,10% of the Brazilian population that is attended by the National Health System (SUS)[2]. Patients with CKD present organic dysfunction in the balance of ions circulating in the blood. The loss of homeostasis tends to occur with a decrease in the concentration of calcium ion (Ca^{2+}) and an increase in phosphate ion (PO_4^{3-}). This imbalance induces an increase in the excretion of parathyroid hormone (PTH), which plays a role in stimulating osteoclasts that reabsorb the inorganic bone matrix and increase the levels of Ca^{2+} circulating in the plasma[3]. This metabolic change, if untreated, tends to progressive degradation of the bone structure or osteodystrophy[4].

Renal osteodystrophy (ROD), is a lesion characterized by alteration of bone tissue with replacement of fibrous tissue[5]. Although it is relatively common among patients with end-stage renal disease, its manifestation in the maxillofacial region is rare[6]. The definitive diagnosis of ROD requires the association of many factors, such as: laboratory tests, imaging tests and histopathological analysis. When it involves maxillary bones, this condition most frequently affects the mandible, female individuals, in the fourth decade of life[6,7].

Statement of Clinical Significance
Renal osteodystrophy (ROD) can cause significant oral-maxillofacial alterations in patients with chronic kidney disease (CKD) and secondary hyperparathyroidism. This study highlights the need for dentists to recognize these manifestations for early diagnosis and treatment. A multidisciplinary approach is essential to ensure proper management, improving patient care and quality of life.

ROD manifests itself by promoting an increase in bone volume and consequent deformity in the jaws and face. Radiographically, bone demineralization is observed with loss of the trabecular pattern and the integrity of

[1]Federal University of Pelotas, Postgraduate Program in Dentistry – Pelotas (RS), Brazil.
[2]Federal University of Pelotas Department of Semiology and Clinic, School of Dentistry – Pelotas (RS), Brazil.
[3]Federal University of Pelotas, Department of Traumatology and Maxillofacial Surgery – Pelotas (RS), Brazil.
[4]Federal University of Pelotas, School of Dentistry, Diagnostic Center for Oral Diseases – Pelotas (RS), Brazil.
***Correspondence to:** Email: aetges@gmail.com
Received on February 10, 2025. Accepted on April 25, 2025.
https://doi.org/10.5327/2525-5711.316

the cortical bone, revealing a ground glass appearance[6]. Histologically, these lesions present a variety of osteitis fibrosa, osteoclast mediated resorption of bone trabecula and replacement by fibrous connective tissue[8].

Usually, the treatment of ROD is systemic, since the alterations are caused by an imbalance in the parathyroid glands caused by chronic kidney disease, but in some cases, parathyroidectomy is necessary. Also, sometimes bone plasty is needed after the treatment to re-establish the normal function of the maxillary bones[6,7].

Considering the above, the objective of the present work is to present a case report of a patient with chronic renal failure with secondary hyperparathyroidism and the presence of manifestations in the oral-maxillofacial complex and to carry out a brief review of the literature, in order to establish the most common clinical characteristics of this entity, seeking to assist in its understanding and, in this way, collaborate with the diagnosis of this nosological entity.

CASE REPORT

Female patient, caucasian, 49 years old, attended in the Diagnostic Center of Oral Diseases, School of Dentistry (DCOD) of the Federal University of Pelotas (UFPel), in Pelotas, Rio Grande do Sul, Brazil, complaining of a painless volumetric increase in the jaws after performed dental extraction. The patient was seen in person in conjunction with the Residency in Oral and Maxillofacial Surgery and Traumatology (RCTBMF) of the Faculty of Dentistry of the UFPel, signing a Free and Informed Consent Form.

During the anamnesis, the patient reported having CKD for 10 years, with the installation of a peritoneal bag in 2014 and removal and reinstallation in 2021. Current treatment includes Sevelamer Hydrochloride and Calcium Carbonate, prescribed for hyperphosphatemia related to CKD, and Omeprazole, used as a gastrointestinal protective agent.

Clinical examination revealed generalized, painless bone expansion in the maxilla and mandible, with the overlying mucosa appearing normal in color. Palpation of the affected areas elicited no pain. The alveolar bone ridges showed significant enlargement, resulting from diffuse bony overgrowth. The maxilla demonstrated marked loss of palatal concavity (Figure 1A), rendering the upper complete prosthesis unusable.

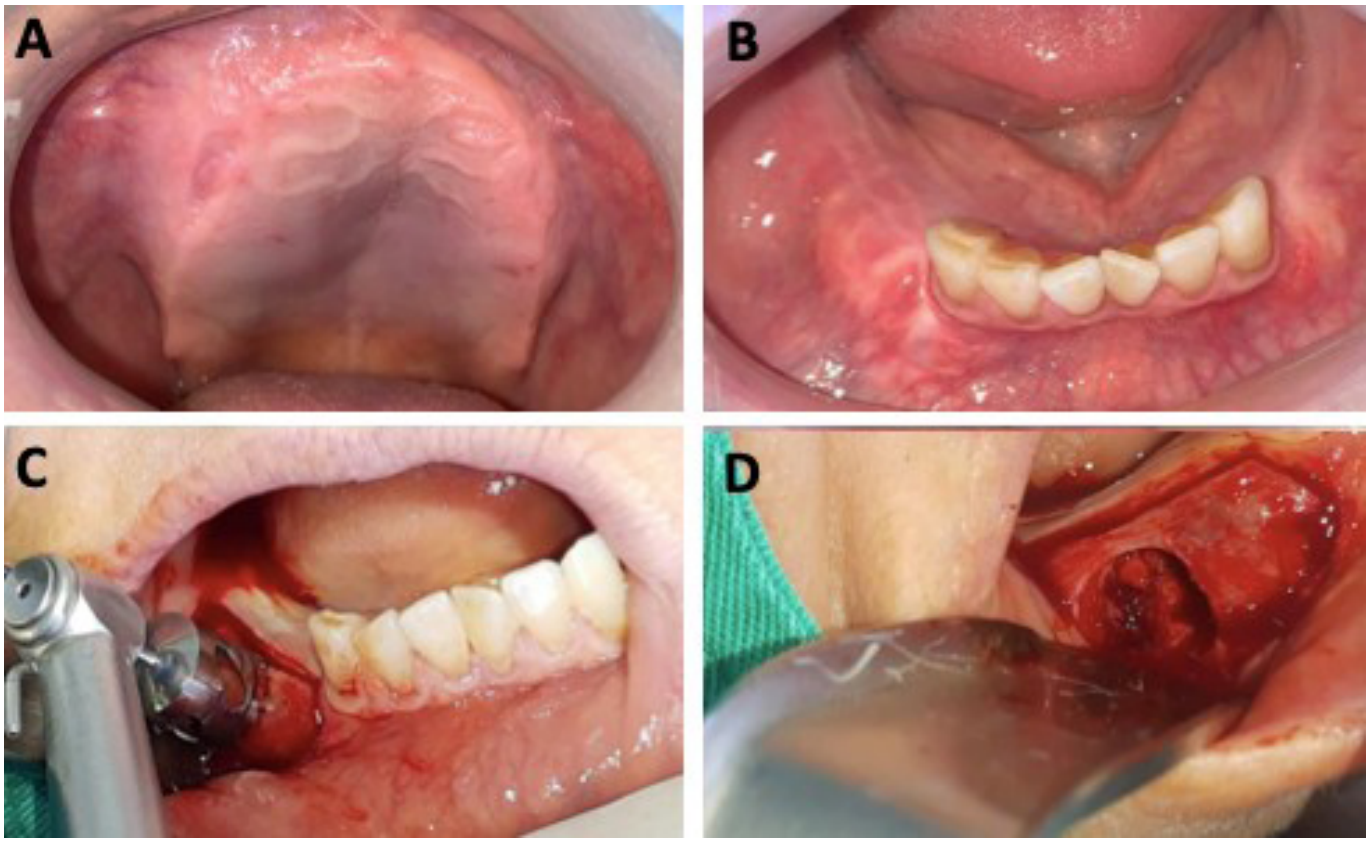

Figure 1. Clinical appearance of the maxilla, showing loss of palate concavity **(A)**, and the mandible, with loss of anatomy at the base of the vestibule **(B)**. The rotary instrument used for collecting the material **(C)** and its bone stock **(D)**.

In the mandible, the lower anterior teeth (incisors and canines) remained without mobility or displacement (Figure 1B). The alveolar ridges exhibited buccolingual expansion, and palpation revealed firm, non-tender bony surfaces with obliteration of the vestibular sulcus depth. Despite these extensive osseous changes, facial symmetry and proportions were maintained. Additional laboratory tests were ordered, with results detailed in Table 1.

On the periapical radiographs (Figure 2), there was a loss of the radiopaque pattern of the alveolar ridges of the mandibular bones, loss of integrity of the cortical bone and the multifocal presence of radiolucent spots similares to the pattern described in the literature as "salt and pepper".

In the Waters examination (Figure 3A), changes in the radiopacity of the contours and volume of the bones of the face and skull are observed.

Table 1. Laboratory tests results.

Blood test	Results
Creatinine	9,6 mg/dl
Calcium	11,0 mg/dl
Alkaline phosphatase	491 U/l
Parathyroid hormone	3.500 pg/mL
Glucose	101 mg/dl
Potassium	4,8 mEq/L
Alanine transaminase	10 U/l
Pre-dialysis urea	165 mg/dl
Post-dialysis urea	33 mg/dl

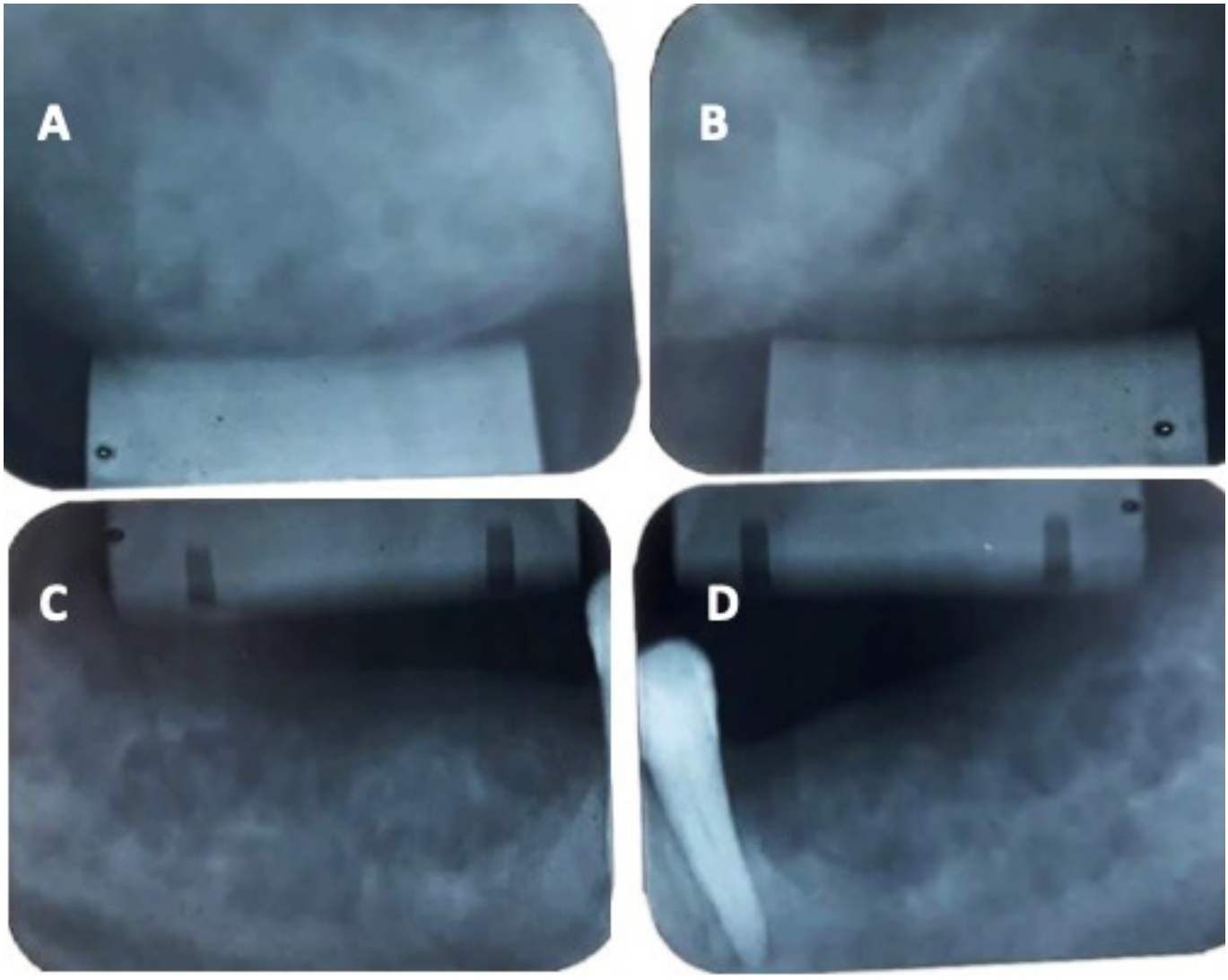

Figure 2. Periapical radiographs showing the posterior region of the right maxilla **(A)**, posterior region of the left maxilla **(B)**, posterior region of the right mandible **(C)**, and posterior region of the left mandible **(D)**. All images show a loss of cortical bone integrity and the multifocal presence of radiolucent spots, resembling the "salt and pepper" pattern described in the literature.

The computed tomography (Figure 3C/3D) demonstrated that the maxilla showed generalized buccopalatal expansion and loss of the palatal concavity. In the posterior region of the mandible, buccal cortical expansion was observed, with preservation of the lingual cortical plate. Within the lesion, in both jaws, an irregular trabecular bone pattern was noted, with projections oriented in multiple directions.

In scintigraphy (Figure 4), hyper uptake of the radiopharmaceutical is observed, corresponding to parathyroid adenomas. The ultrasonography report of the posterior surface of the middle and lower third of the thyroid region reveals solid, hypoechoic, nodular images with regular, well-defined margins and, due to their location, related to enlargement of the parathyroid glands. The left lower third measured 2.18 x 1.60 x 1.32cm and had calcifications inside.

Laboratory tests (Table 1) values agreed with a dysfunctional picture of calcium regulation, which when associated with images of nodules in parathyroid glands and the history of chronic kidney disease suggested a brown tumor. Therefore, an incisional biopsy

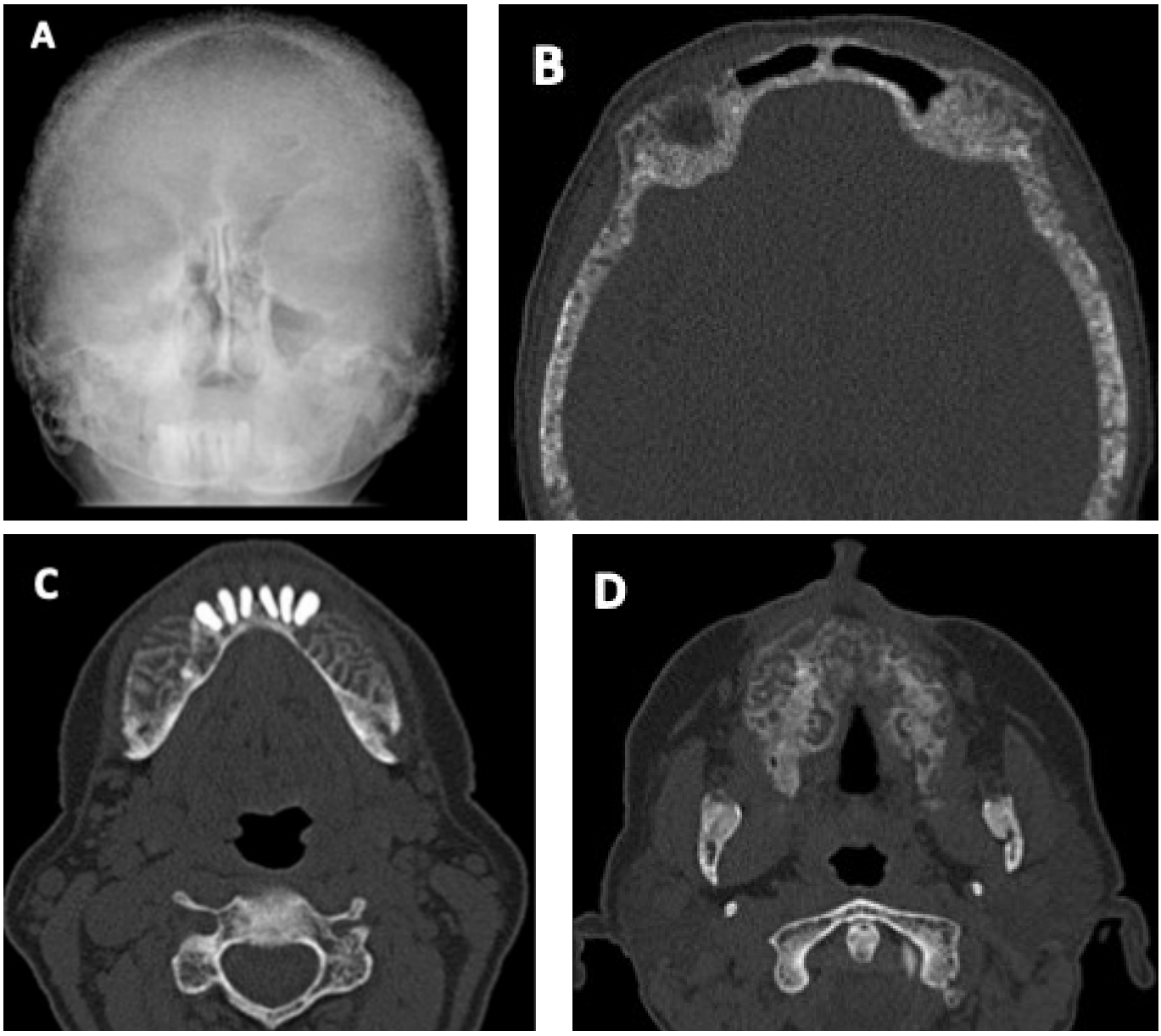

Figure 3. Waters X-ray showing altered radiopacity in the contours and volume of the facial and skull bones, as salt and pepper **(A)**. The axial sections of the CT scan **(B–D)** reveal altered trabecular bone in the face, displaying a hypodense pattern, as well as an infiltrate in localized areas of the skull, with loss of continuity and integrity of the cortical bone.

was performed through a mucoperiosteal access in the mandible alveolar ridge region, using a bone trephine (Figure 1C) covering the cortical and medullary bone (Figure 1D). The removed material was sent for histopathological analysis.

Macroscopic analysis revealed two fragments of hard tissue, collectively measuring 15×12×10 mm, exhibiting a firm consistency, whitish-reddish coloration, and an ellipsoidal morphology. Histopathological analysis showed a fragment of bone tissue, with an area of intense tissue replacement by fibrous connective tissue, moderately vascularized and cellularized by spindle cells, forming bone matrix and a periphery of bone trabeculation, some with the appearance of Chinese characters (Figure 5A, B). The newly formed bone had osteoblasts containing osteocytes inside and some multinucleated giant cells were present (Figure 5C, D).

The diagnosis was consolidated as ROD, with secondary manifestation in the bones of the craniofacial complex. After the diagnosis, the patient returned to the endocrinologist to evaluate the altered laboratory levels and the possibility of surgical intervention and removal of the parathyroid adenomas.

DISCUSSION

We presented the case report of a 49-year-old female patient, with a diagnosis of ROD and conducted a literature review to determine the most affected anatomical location and the most commonly used treatment, in PubMed using in PubMed using the MeSH Terms descriptors: (((jaws) AND (treatment)) AND (secondary hyperparathyroidism)) AND (case reports). The search was limited to case reports written in English and published in the last 10 years, which resulted in 20 articles, then, 13 articles were selected and reviewed (Figure 6).

This case stands out for the involvement of both the maxilla and the mandible, a less frequent finding

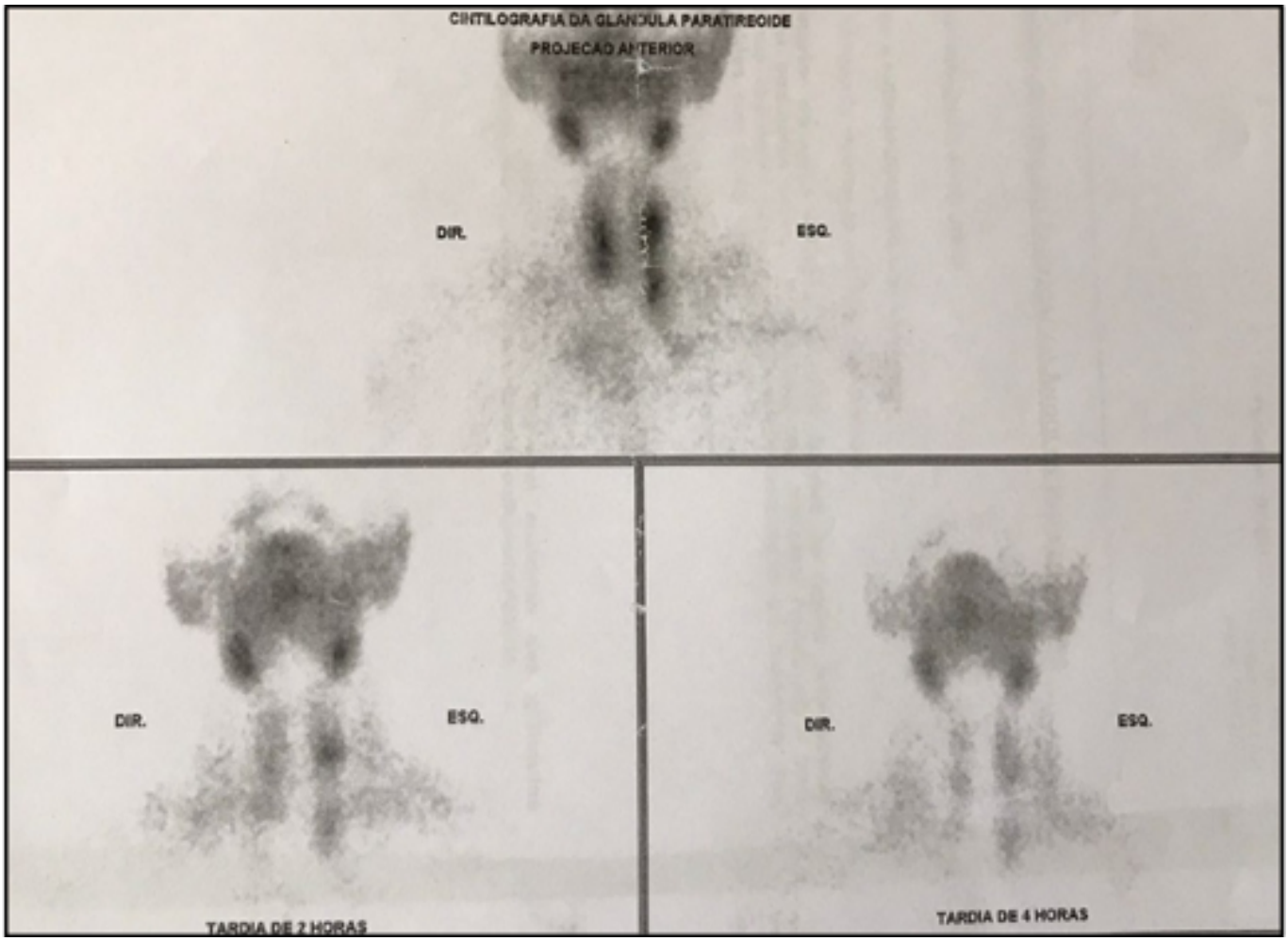

Figure 4. Scintigraphy showing abnormal radiopharmaceutical uptake in the left-sided parathyroid region.

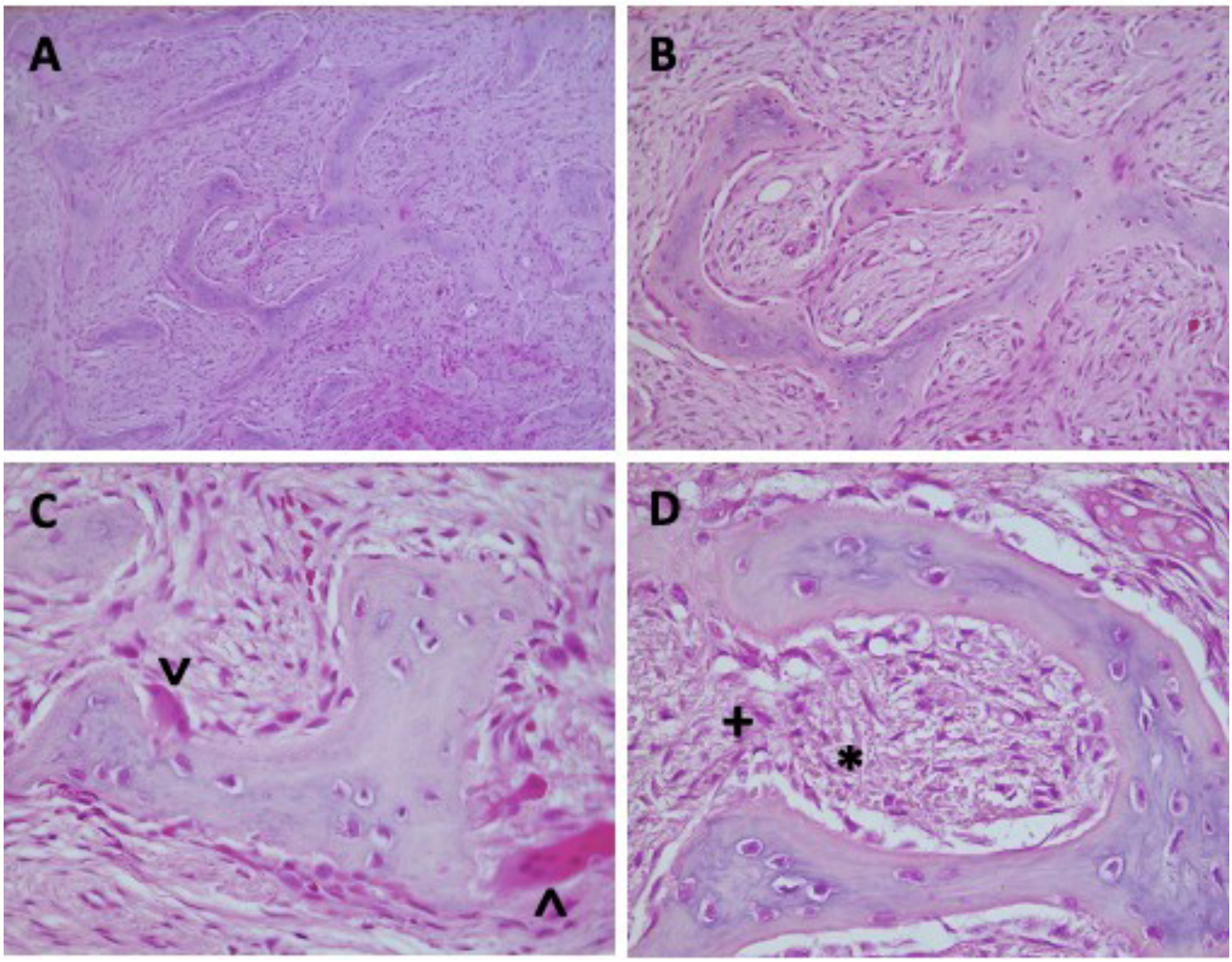

Figure 5. Histopathological images (H&E: A- 40x, B- 100x, C- 200x, D- 400x) showing bone trabeculation in Chinese characters, as well as medullary replacement by fibrous connective tissue, densely arranged and cellularized **(A–B)**. Multinucleated giant cells (^) are present near the trabecular bone **(C)**. Spindle cells (*) and polyhedral cells (+) are observed in the fibrous connective tissue **(D)**.

in the literature, as seen in Table 2[7,9-19], which demonstrates that the majority of diagnosed cases of ROD develop in just one of the maxillary bones. The patient in the present case was also in an age group above that most frequently identified in the literature at the time of diagnosis, while the patient in this case was 49 years old, in the literature review[7,9-19], most of the studies present cases of patients in the third and fourth decades of life, with an average age of 35 years, although some studies report cases that deviate from the rule, such as that of Hakkou et al.[7], who reported the case of two patients, one aged 55 and the other aged 71 (Table 2).

The presence of expansive lesions, whose tomographic aspects reveal diffuse images, in which it is not possible to verify the limits between cortical and medullary, can be confused with fibrous dysplasias[4]. The differential diagnosis of ROD with other expansive bone pathologies, such as brown tumor, cystic fibrous osteitis, cherubism, central giant cell lesion and ossifying fibroma, should always be considered[6,7]. The presence of a bulging hard palate and clinical and radiographic characteristics in more than one facial bone may initially be confused with an isolated case of fibrous dysplasia[5-9]. A radiographic finding called the "salt and pepper" pattern, revealing the loss of continuity of the external surface of the double layer and the skull, is characteristic of ROD[9]. In this case, these changes in the radiopacity of the contours and volume of the bones of the face and skull were observed, contributing to the diagnosis.

Microscopically, the histopathological pattern observed in this case may resemble fibrous dysplasia, which can lead to diagnostic confusion[13]. However, the integration of histopathological findings with a thorough anamnesis, laboratory results, and the patient's systemic condition was essential to establish an accurate diagnosis[6]. Interdisciplinary communication — particularly between the surgeon, oral pathologist and the nephrologist — and full access to the patient's medical history are crucial in such scenarios. In the present case, histological examination revealed replacement of normal bone architecture by a fibro-osseous matrix, along with scattered multinucleated giant cells. These findings, when interpreted alongside imaging and biochemical data, confirmed the diagnosis of renal osteodystrophy with expansive characteristics.

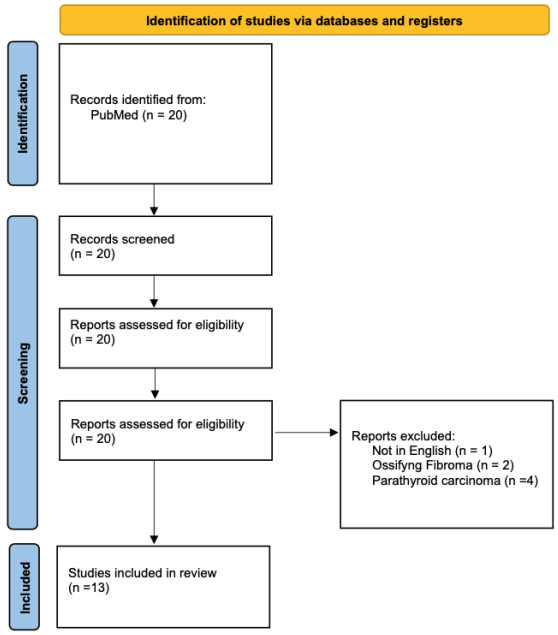

Figure 6. PRISMA flow chart illustrating the selection process for case reports included in the qualitative synthesis.

In this case, laboratory tests reveal a PTH greater than 3,500 pg/ml and alkaline phosphatase with 491U/L, findings that are of a decompensated patient[14], that deserve immediate evaluation by the attending physician. Under these conditions, the treatment of secondary hyperparathyroidism aims to balance circulating Ca^{2++} and PO_4^{3-} levels through diet and the use of PO_4^{3-} binders, control PTH and Vitamin D using calcimimetics and maintain them within acceptable standards[4].

Despite the available treatment options, there is no standardized protocol for managing this condition. In the reviewed literature, therapeutic approaches varied considerably:

Table 2. Cases of expansive renal osteodystrophy of the articles reviewed.

Author(s)	Year	Country	Sex	Age	Lesion location	Treatment and follow up
Hoornenborg et al.[9]	2014	Netherlands	F	38	Maxilla and Mandible	Not specified, no follow-up
Hussain and Hammam[10]	2016	Saudi Arabia	F	21	Mandible	Oral vitamin D, 36 months follow-up
Aerden et al.[11]	2018	Belgium	M	32	Mandible	Oral vitamin D, 6 months follow-up
Lim and Thevandran[12]	2017	Malaysia	M	21	Maxilla	Total parathyroidectomy, 17 months follow-up
Andrade et al.[13]	2018	Brazil	F	24	Maxilla and mandible	PTH, Ca, alkaline phosphatase stabilization and osteoplasty, 96 months follow-up
Guéroult and Cameron[14]	2019	United Kingdom	M	42	Mandible	Surgical enucleation, follow-up every 6 months
Wilt et al.[15]	2019	United States	M	28	Maxilla and mandible	Osteoplasty, 1 month follow-up
Shavlokhova et al.[16]	2021	Germany	F	41	Mandible	Radical resection and graft, 60 months follow-up
Gaballah et al.[17]	2021	Australia	F	12	Mandible	Oral vitamin D, 18 months follow-up
Ruddocks et al.[18]	2022	United States	M	21	Maxilla and mandible	Palliative care, no follow-up information
Priyanthan et al.[19]	2022	Denmark	F	54	Mandible	Subtotal parathyroidectomy, no follow-up
Hakkou et al.[7]	2023	Morocco	M	55	Maxilla	Referred to an endocrinologist for management of hyperparathyroidism, no follow-up
Hakkou et al.[7]	2023	Morocco	F	71	Mandible	Surgical removal, no follow-up

three patients received vitamin D supplementation[10,11,17]; one underwent stabilization of PTH, calcium, and phosphate levels combined with osteoplasty[12]; one received surgical enucleation[14]; one underwent total parathyroidectomy[12]; one had partial parathyroidectomy[19]; one was treated with osteoplasty alone[15]; one received palliative care[18]; and one was referred to an endocrinologist[7] (Table 2). Palla et al.[5] suggested parathyroidectomy as the first-line treatment for controlling bone dystrophy. However, in cases with expansive bone alterations and facial deformities, these changes may persist even after surgical intervention. Therefore, osteoplasty and complementary dental procedures may be necessary to restore function and aesthetics[13,21].

Although long-term follow-up data were not consistently reported across all reviewed cases, some studies described favorable outcomes after treatment. Andrade et al.[13] reported no evidence of recurrence and stable bone structure eight years after initial surgery and systemic stabilization, with the patient progressing to orthodontic treatment and achieving good facial aesthetics. Similarly, Shavlokhova et al.[16] documented a five-year follow-up with no recurrence after radical resection and bone grafting, highlighting the potential for long-term stability following definitive treatment. These reports reinforce the importance of systemic control and interdisciplinary care in achieving both functional and aesthetic recovery. However, the limited number of cases with comprehensive follow-up underscores the need for future studies to include long-term clinical and radiographic monitoring to better understand recurrence patterns and bone remodeling potential.

CONCLUSION

The diagnosis of ROD is not common and usually presents itself as a challenge. Because of its similarity with other lesions that affect the jaws, the systemic history of patients and the association of clinical and imaging characteristics are crucial to the correct diagnosis of this injury. The treatment approach depends on the patient's systemic status and is contingent upon resolution of the underlying predisposing condition. This highlights the critical importance of multidisciplinary collaboration for accurate diagnosis and comprehensive management of patients with systemic diseases.

AUTHORS' CONTRIBUTIONS

LMD: Conceptualization, Data curation, Methodology, Writing – original draft. IVBC: Conceptualization, Data curation, Methodology, Writing – original draft, Writing review – editing. BFO: Conceptualization, Data curation, Writing review – editing. CFN: Conceptualization, Data curation, Writing review – editing. COL: Data curation, Writing review – editing. MAT: Data curation, Supervision, Validation, Writing review – editing. ACMF: Data curation, Supervision, Validation, Writing review – editing. AE: Data curation, Supervision, Validation, Writing review – editing.

CONFLICT OF INTEREST STATEMENT

Funding: The authors declare that no funds, grants, or other support were received during the preparation of this manuscript.

Competing interests: The authors have no relevant financial or non-financial interests to disclose.

Ethics approval: Not applicable.

REFERENCES

1. Kidney Disease: Improving Global Outcomes (KDIGO) CKD Work Group. KDIGO 2024 clinical practice guideline for the evaluation and management of chronic kidney disease. Kidney Int. 2024;105(4S):S117-S314. https://doi.org/10.1016/j.kint.2023.10.018
2. Brasil. Ministério da Saúde. Secretaria de Vigilância em Saúde e Ambiente. Cenário da doença renal crônica no Brasil no período de 2010 a 2023. Boletim Epidemiologico [Internet]. Brasília: Ministério da Saúde; 2024 [cited 2024 Dec 16]. Available from: https://www.gov.br/saude/pt-br/centrais-de-conteudo/publicacoes/boletins/epidemiologicos/edicoes/2024/boletim-epidemiologico-volume-55-no-12.pdf
3. Silva BC, Costa AG, Cusano NE, Kousteni S, Bilezikian JP. Catabolic and anabolic actions of parathyroid hormone on the skeleton. J. Endocrinol Investig. 2011;34(10):801-10. https://doi.org/10.3275/7925
4. Saliba W, El-Haddad B. Secondary hyperparathyroidism: pathophysiology and treatment. J Am Board Fam Med. 2009;22(5):574-81. https://doi.org/10.3122/jabfm.2009.05.090026
5. Palla B, Burian E, Fliefel R, Otto S. Systematic review of oral manifestations related to hyperparathyroidism. Clin Oral Investig. 2018;22(1):1-27. https://doi.org/10.1007/s00784-017-2124-0
6. Pérez-Guillermo M, Acosta-Ortega J, García-Solano J, Ramos-Freixá J. Cytologic aspect of brown tumor of hyperparathyroidism. Report of a case affecting the hard palate. Diagn Cytopathol. 2006;34(4):291-4. https://doi.org/10.1002/dc.20434
7. Hakkou F, Benjelloun L, Hallab L, Chbicheb S. Brown tumor of the jaw as a rare manifestation of hyperparathyroidism: two case reports and literature review. Int J Surg Case Rep. 2023;111:108823. https://doi.org/10.1016/j.ijscr.2023.108823
8. Raubenheimer EJ, Noffke CE, Mohamed A. Expansive jaw lesions in chronic kidney disease: review of the literature and a report of two cases. Oral Surg Oral Med Oral Pathol Oral Radiol. 2015;119(3):340-5. https://doi.org/10.1016/j.oooo.2014.11.002

9. Hoornenborg E, van Delft F, Vogt L. Jaw enlargement in a haemodialysis patient. Secondary hyperparathyroidism. Neth J Med. 2014;72(4):229, 233. PMID: 24829182.
10. Hussain M, Hammam M. Management challenges with brown tumor of primary hyperparathyroidism masked by severe vitamin D deficiency: a case report. J Med Case Rep. 2016;10:166. https://doi.org/10.1186/s13256-016-0933-4
11. Aerden T, Grisar K, Nys M, Politis C. Secondary hyperparathyroidism causing increased jaw bone density and mandibular pain: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol. 2018;125(3):e37-e41. https://doi.org/10.1016/j.oooo.2017.11.020
12. Lim CTS, Thevandran TK. Resolution of rare palatal brown tumour after parathyroidectomy. Clin Exp Nephrol. 2017;21(2):352-3. https://doi.org/10.1007/s10157-016-1292-6
13. Andrade GS, Carvalho ACGS, Magalhães TG, Cetira Filho EL, Cavalcante RB, Nogueira RLM. Expansive renal osteitis fibrosa: a case report. Oral Maxillofac Surg. 2018;22(3):323-7. https://doi.org/10.1007/s10006-018-0697-7
14. Guéroult AM, Cameron M. Rare brown tumour of the mandible secondary to tertiary hyperparathyroidism in a renal transplant recipient. BMJ Case Rep. 2019;12(9):e231231. https://doi.org/10.1136/bcr-2019-231231
15. Wilt DC, Kim B, Zaid W. The use of virtual surgical-guided osteoplasty for maxillofacial brown tumors. J Craniofac Surg. 2019;30(6):e551-e553. https://doi.org/10.1097/SCS.0000000000005507
16. Shavlokhova V, Goeppert B, Gaida MM, Saravi B, Weichel F, Vollmer A, et al. Mandibular brown tumor as a result of secondary hyperparathyroidism: a case report with 5 years follow-up and review of the literature. Int J Environ Res Public Health. 2021;18(14):7370. https://doi.org/10.3390/ijerph18147370
17. Gaballah K, Kenz S, Anis R, Kujan O. Can vitamin D therapy contribute to the conservative resolution of osteolytic lesions of the jaws? Case Rep Dent. 2021;2021:5510724. https://doi.org/10.1155/2021/5510724
18. Ruddocks LA, Nascimento AF, Bhattacharyya I, Islam MN, Cohen DM. Central odontogenic fibroma in association with brown tumor of hyperparathyroidism in a patient with neurofibromatosis type 1. Oral Surg Oral Med Oral Pathol Oral Radiol. 2022;133(1):e26-e32. https://doi.org/10.1016/j.oooo.2021.07.004
19. Priyanthan T, Hermann AP, Bojsen JA, Krøigaard AB, Bistrup C, Pedersen EB. Multiple focal brown tumors (osteitis fibrosa cystica) in a renal transplant recipient. Case Rep Nephrol. 2022;2022:4675041. https://doi.org/10.1155/2022/4675041
20. Raubenheimer EJ, Noffke CE, Hendrik HD. Recent developments in metabolic bone diseases: a gnathic perspective. Head Neck Pathol. 2014;8(4):475-81. https://doi.org/10.1007/s12105-014-0580-2
21. Hata T, Irei I, Tanaka K, Nagatsuka H, Hosoda M. Macrognathia secondary to dialysis-related renal osteodystrophy treated successfully by parathyroidectomy. Int J Oral Maxillofac Surg. 2006;35(4):378-82. https://doi.org/10.1016/j.ijom.2005.08.006