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CASE REPORT

Oral Manifestations of Renal Osteodystrophy in a Patient with Systemic Lupus Erythematosus with Chronic Renal Failure and Secondary Hyperparathyroidism: A Case Report

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ABSTRACT

Systemic lupus erythematosus (SLE) is one of the chronic autoimmune diseases that leads to multiple manifestations in several organ systems, including chronic renal failure (CRF). Renal osteodystrophy (ROD) is one of the most common complications of CRF associated with secondary hyperparathyroidism (HPT). A combination of CRF and HPT can result in broad-spectrum disorders in the metabolism of minerals such as calcium, phosphorus, and vitamin D, which can affect bone turnover, mineralization, and volume. Radiographic alterations of ROD are considered as one of the earliest signs of CRF. **Objective:** To describe the oral manifestations in both the maxilla and mandible of a patient with ROD resulting from SLE with CRF and secondary HPT. **Case report:** A 37-year-old female patient visited the Faculty of Dentistry, Khon Kaen University, for the management of traumatic ulcers on the left and right buccal mucosa and generalized maxillary and mandibular bony hard swellings with mild paresthesia in the lower left mandible for a period of 4 months. The patient had been suffering from SLE for approximately 20 years. However, secondary HPT and CRF as consequences of refractory SLE were being treated concomitantly. Intraoral examination showed several nonpainful ulcers on the left and right buccal mucosa. Radiographic findings revealed generalized loss of lamina dura with thinning of cortical outlines of the maxilla and mandible. Due to complications associated with treatment and the general conditions of this patient, she was referred for treatment to a palliative care unit. **Conclusion**: Systemic symptoms and complications of patient with ROD with CRF and secondary HPT are seriously problematic. The role of the dentist in this case is to detect the disease, lessen the complications, and make the patient experience minimal pain and side effects in palliative care.

Key words: chronic renal failure, hyperparathyroidism, renal osteodystrophy, systemic lupus erythematosus

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INTRODUCTION

Systemic lupus erythematosus (SLE) is one of the autoimmune diseases that causes chronic inflammation in several organ systems, including vasculitis, pleuritis, pericarditis, and nephritis. Long-term renal inflammation can develop into chronic renal disease (CRD) and chronic renal failure (CRF).[1] The effects of CRF can result in broad-spectrum disorders of minerals such as vitamin D, phosphate, and potassium and anemia that can induce abnormal production of parathyroid hormone and bone malformation.

Renal osteodystrophy (ROD) is a bone malformation disease and one of the most common complications of CRF associated with secondary hyperparathyroidism (HPT). However, there are only few cases of ROD that affect jaw bones.[2,3]

This article reports and discusses about the case of a patient with ROD in both the maxilla and mandible with multiple chronic systemic conditions resulting from SLE for 20 years, with CRF and secondary HPT being present for 7 years.

CASE REPORT

A 37-year-old female patient was referred to Khon Kaen University Dental Hospital with a primary complaint of gradual expansion of her maxilla and mandible along with generalized spacing of teeth and traumatic ulcers around the buccal mucosal area near the left and right second molars for 4 months. Her medical history revealed that she had SLE, which was treated with systemic corticosteroids for 20 years.

Consequently, she had developed CRF due to SLE for the past 7 years. She had also been diagnosed with secondary HPT caused due to CRF stage V for 4 months. She was also on hemodialysis on alternate days and on medications such as alphacalcidol 1.5 µg/day, calcium carbonate 1800 mg/day, losartan potassium 50 mg/day, hydralazine 100 mg/day, simvastatin 20 mg/day, folic acid 5 mg/day, and ferrous fumarate 200 mg/day.

General physical examination revealed that the patient experienced weakness and fatigue when she walked. Malformation of bone, both in the maxilla and mandible, was also detected. Extraoral examination revealed facial asymmetry and bony hard swelling on the maxilla and mandible (Figure 1), limited mouth opening (20 mm), no lymphadenopathy, no tenderness, and noncompressible on palpation.

Intraoral examination demonstrated generalized spacing of teeth, generalized 2nd degree teeth mobility, bony hard swelling on the maxilla and mandible, shallow palatal vault, and traumatic ulcers measuring 10 mm in diameter on bilateral buccal mucosa near the left and right second molars due to trauma (Figure 2-4). The patient had good oral hygiene, generalized pale pink gingiva with mild plaque, and calculus deposition. Her salivary excretion was normal. There was no sign of intraoral infection or inflammation.

The radiographic examination was done using panoramic and cone beam computed tomography (CBCT). The panoramic radiograph showed osteolytic areas, trabelcular pattern and general appearance of the lamina dura and cortical outlines of the jaw bones of the patient (Figure 5). The CBCT showed bone algorithm, expansion of the maxilla and mandible with multiple radiolucency and decreasing trabecular density (Figure 6).

Laboratory investigations demonstrated that the levels of BUN, creatinine, alkaline phosphatase, and RDW were higher than the normal range, whereas the levels of chloride, total bilirubin, Hb, and Hct were lower than the normal values. (Table 1)

The patient was monitored by the physician and treated with oral medications such as alphacalcidol 1.5 µg/day, calcium carbonate 1800 mg/day, losartan potassium 50 mg/day, hydralazine 100 mg/day, simvastatin 20 mg/day, and folic acid 5 mg/day for her systemic diseases and with ferrous fumarate 200 mg/day as a mineral supplement. For secondary HPT, the physician made an

Table 1. Laboratory investigation (* indicates abnormal value) Based on history and clinical examination, a diagnosis of renal osteodystrophy (ROD) was made.

Investigations Chronic renal failure	Observed values	Normal values
BUN	25.9 mg/dL*	5.8–19.1 mg/dL
Creatinine	6.5 mg/dL*	0.5–1.5 mg/dL
Chloride	91 mEq/L*	96–107 mEq/L
Total bilirubin	0.3 mg/dL*	0.3–1.5 mg/dL
Hyperparathyroidism		
Alkaline phosphatase	532 U/L*	42–121 U/L
Anti-HBS	Negative	
Anti-HCV	Negative	
Cytomegalovirus	Negative	
Anti-HIV	Negative	
AST	13 U/L	12–32 U/L
ALT	6 U/L	4–36 U/L
Calcium	9.6 mg/dL	8.4–10.2 mg/dL
Others		
RDW	0.211%*	11.9–14.8%
Hb	10.6 g/dL*	12.0–14.3 g/dL
Hct	34.6%*	36.0–47.7%
MCV	90.9 fL	80.0–97.8 fL

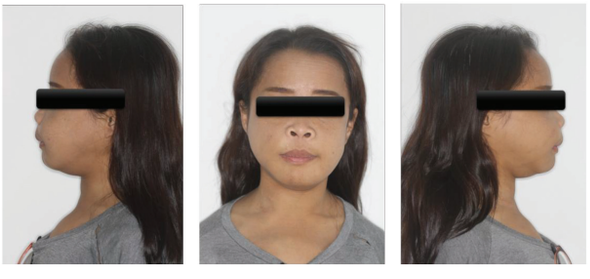

Figure 1. Extraoral examination demonstrated facial asymmetry, convex profile, and swelling of the maxilla and mandible.

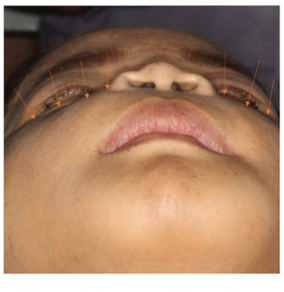

Figure 2. Swelling of the maxillary bone and saddle nose due to bone expansion.

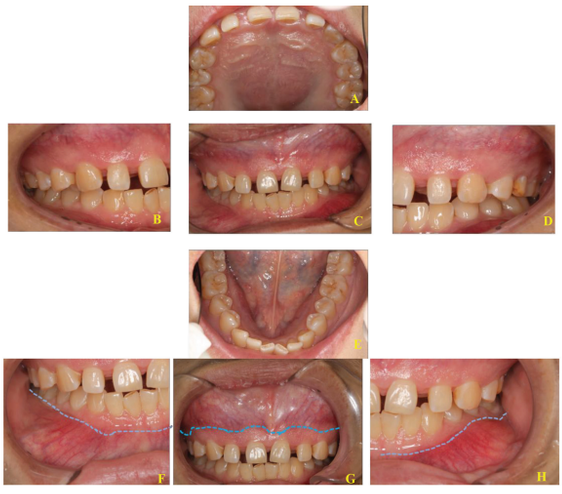

Figure 3. (A-H) Generalized tooth spacing, bone swelling on the alveolar ridge (blue line), shallow buccal vestibule, and palatal vault.

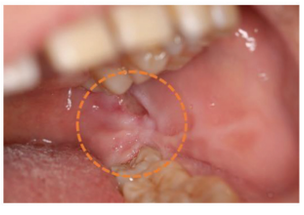

Figure 4. Traumatic ulcers measuring approximately 10 mm in diameter on the left and right buccal mucosa caused by the drifting and migration of left and right second molars.

appointment for further investigations and referred the patient to the endocrinologist for appropriate treatment.

Regarding dental management, the dentist prescribed 0.1% triamcinolone acetonide in orabase three times per day as a topical treatment for the traumatic ulcers, gave oral hygiene instructions to maintain good oral condition and scheduled to follow up the patient for 2 weeks.

The conditions of generalized tooth spacing, tooth drifting, tooth mobility, and paresthesia were treated symptomatically due to her complex systemic condition.

DISCUSSION

SLE is a chronic autoimmune disease that can affect several organs of the body. It can cause various symptoms such as malar rash, joint pain and swelling, severe fatigue, headache, hair loss, anemia, and blood clotting problems. When long-term chronic SLE still persists, it can cause morbidities such as vasculitis, pericarditis, heart attack, pleuritis, decreased renal function, and renal failure.[1]

CRF is a multifactorial syndrome characterized by progressive and irreversible loss of renal function. The most common causes of CRF are diabetes mellitus,

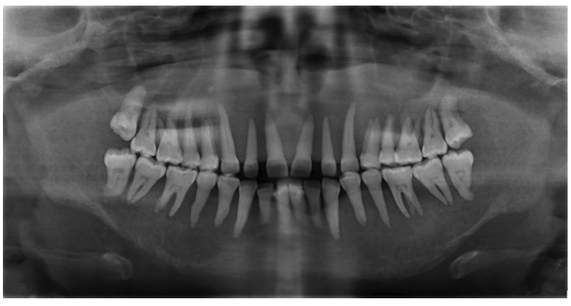

Figure 5. Panoramic radiograph demonstrated generalized osteolytic area, reduction of trabecular pattern, ground-grass appearance, and loss of lamina dura with thinning of cortical outlines of the maxilla and mandible. Left and right inferior alveolar canals were not detected.

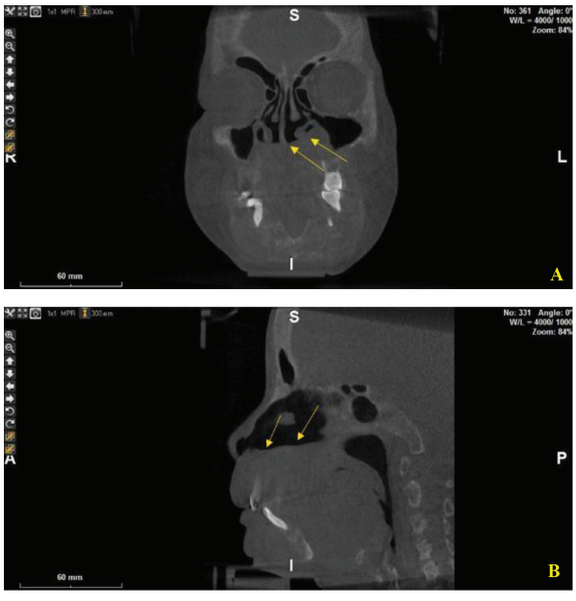

Figure 6. A. Coronal view; B. Sagittal view of cone beam computed tomography images with bone algorithm of this case showed expansion of the maxilla and mandible and multiple radiolucent areas in the maxilla and body of the mandible. Decrease in trabecular density was also observed (yellow arrows)

SLE, hypertension, and chronic glomerulonephritis. The symptoms of CRF consist of gout, anemia, hyperkalemia, heart disease, fluid buildup, broad spectrum of disorders of mineral metabolism, and hyperphosphatemia.[2, 4]

HPT is caused due to an excessive production of parathyroid hormone (PTH) by parathyroid glands. HPT can be categorized into the following four types: primary, secondary, tertiary, and quaternary. HPT manifests with several symptoms such as osteoporosis, fatigue and weakness, joint pain, and abdominal pain. Secondary HPT is one of the most common types of HPT. It can occur as a complication of CRF as a result of ROD.[4-11]

ROD is a frequent long-term complication of renal disease that can induce a broad spectrum of bone metabolism disorders associated with different pathogenic pathways. The symptoms of ROD comprise bone demineralization with trabeculation and cortical loss, giant cell radiotransparencies, or metastatic calcifications of the soft tissues. The risk of bone fracture and obstruction of airways due to deformation of the hard palate are the important issues that require concern during dental treatment.[4-6, 12]

This patient was diagnosed with SLE and treated with systemic corticosteroids for 13 years. Long-term treatment of this disease induced renal inflammation that later developed into CRD and CRF. Consequently, the patient had to stop systemic corticosteroids due to her complex systemic conditions for 7 years. Furthermore, she was diagnosed with secondary HPT because of long-term CRF, which led to hyperphosphatemia and induced the parathyroid gland to release high levels of PTH that in turn led to secondary HPT as detected by alkaline phosphatase levels.[13] In this patient, ROD was one of the complications of CRF combined with secondary HPT.[2] The symptoms of ROD included bone malformation that appeared in several bones in the body.[6]

A few cases of osteodystrophy involving jaw bones have been reported in the literature.[14] Asaumi et al reported cases of diffuse swelling of both jaws in hemodialysis patients with long-standing CRD.[15] Damm et al described seven cases of diffuse swelling of both jaws secondary to ROD.[16] Furthermore, Kakade et al in 2015 reported a case of ROD in a patient with secondary HPT.[4] Radiographic findings of all cases demonstrated similar clinical and radiographic features such as ground-grass appearance, loss of trabecular pattern, and diffuse swelling of the maxilla and mandible. However, the majority of cases of severe jaw enlargement associated with HPT failed to return to normal contours even after renal transplantation.[17]

CONCLUSION

Despite the limited reports of patients with ROD with SLE, CRF, and secondary HPT, systemic symptoms and complications of these diseases are seriously problematic, which must be concerned by the doctors. Early diagnosis and appropriate treatment of these diseases are the best strategy to reduce the complications. The role of the dentist in this case is to detect the disease, lessen the complications, and make the patient experience minimal pain and side effects in palliative care.

REFERENCES

1. Cojocaru M, Cojocaru IM, Silosi I, Vrabie CD. Manifestations of systemic lupus erythematosus. Maedica (Buchar). 2011;6(4):330-6.
2. Ketteler M, Block GA, Evenepoel P, Fukagawa M, Herzog CA, et al. Diagnosis, evaluation, prevention, and treatment of chronic kidney disease-mineral and bone disorder: Synopsis of the kidney disease: Improving global outcomes 2017 Clinical Practice Guidelines Update. Ann Intern Med. 2018;168(6):422-30.
3. Bartter FC. Bone as a target organ: Toward a better definition of osteoporosis. Perspect Biol Med. 1973;16(2):215-31.
4. Kakade S, Gogri A, Umarji H, Kadam S. Oral manifestations of secondary hyperparathyroidism: A case report. Contemp Clin Dent. 2015;6(4):552-8.
5. Parthiban J, Aarthi Nisha V, Asokan GS, Prakash CA, Varadharaja MM. Oral manifestations in a renal osteodystrophy patient - a case report with review of literature. J Clin Diagn Res. 2014;8(8):ZD28-30.
6. El-Kishawi AM, El-Nahas AM. Renal osteodystrophy: Review of the disease and its treatment. Saudi J Kidney Dis. 2006;17(3):373-82.
7. Avram MM. Symposium proceedings. Perspective: irreversible progressive chronic kidney disease - An overview. Kidney Int Suppl. 2010(117):S1-2.
8. Davis JS, Liles AM, Hain DJ. NPs in nephrology. An overview of chronic kidney disease. Adv Nurse Pract. 2010;18(7):30-2.
9. Galvao TF, Araujo ME, Penha AP, Silva MT. Statins for early stage chronic kidney disease: an overview of reviews. Cardiovasc Hematol Disord Drug Targets. 2014;14(3):205-11.
10. Jain N, Simoyi P. An overview of chronic kidney disease management and CAPD in the home. Br J Community Nurs. 2008;13(5):213-8.
11. Liyanage T, Ninomiya T, Perkovic V, Woodward M, Stirnadel-Farrant H, Matsushita K, et al. Chronic kidney disease in Asia: Protocol for a collaborative overview. Nephrology (Carlton). 2017;22(6):456-62.
12. Diaz-Corte C, Naves Diaz ML, Gomez Alonso C, Barreto S, Cannata Andia JB. Prevention, diagnosis and treatment of renal osteodystrophy in Spain. Preliminary results from a multicentre enquiry. Nephrol Dial Transplant. 1998;13 Suppl 3:51-6.
13. Diethelm AG, Adams PL, Murad TM, Daniel WW, Whelchel JD, Rutsky EA, et al. Treatment of secondary hyperparathyroidism in patients with chronic renal failure by total parathyroidectomy and parathyroid autograft. Ann Surg. 1981;193(6):777-93.
14. Ergün S, CE. Localized jaw enlargement caused by renal osteodystrophy in an adolescent patient.

Marmara Dent J. 2013;2:87-9.
15. Asaumi J, Aiga H, Hisatomi M, Shigehara H, Kishi K. Advanced imaging in renal osteodystrophy of the oral and maxillofacial region. Dentomaxillofac Radiol. 2001;30(1):59-62.
16. Damm DD, Neville BW, McKenna S, Jones AC, Freedman PD, Anderson WR, et al. Macrognathia of renal osteodystrophy in dialysis patients. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1997;83(4):489-95.
17. Tarrass F, Benjelloun M, Bensaha T. Severe jaw enlargement associated with uremic hyperparathyroidism. Hemodial Int. 2008;12(3):316-8.

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