# CASE REPORT

## Dental radiography

### Oral aspects of sickle cell disease

Sir, a 20-year-old male of African descent was referred to our oral surgery clinic by his GDP following a presentation of non-specific, generalised pain affecting various areas in both the maxilla and mandible bilaterally. The pain arose several times each month with varying severity and duration with symptoms ongoing for over one year. The patient described the pain as a throbbing ache affecting the teeth, gingivae and bone. The GDP had taken a periapical radiograph of the lower left second molar (Fig. 1) and was concerned about a generalised radiolucency apical to the teeth in view.

The patient's medical history included sickle cell anaemia (SCA), for which he is under the care of a specialist haematology team. His medication included folic acid, penicillin and hydroxycarbamide. SCA is a severe form of sickle cell disease (SCD), which includes genetic blood disorders characterised by morphological changes to erythrocytes caused by sickle haemoglobin (HbS). These abnormalities can lead to haemolytic anaemia and episodes of micro-vascular vaso-occlusion, resulting in local hypoxia. This is the mechanism behind the characteristic painful vaso-occlusive crises experienced by affected patients.[1]

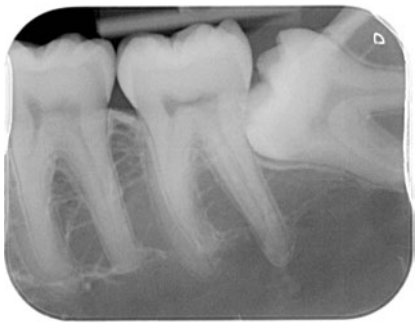

Fig. 1 Periapical radiograph of the lower left second molar

Clinical examination revealed very little of note, with no obvious signs of pathology or distinct abnormalities detected. It should be noted that the patient had excellent oral hygiene. We took an OPG (Fig. 2) in order to identify potential hard tissue sources of pathology in keeping with the patient's presenting complaint. The radiograph demonstrated increased radiolucency of the bone in the inferior body of the mandible, with a distinct, sparse trabecular pattern in this area. Superior to this, coarse trabeculae can be seen. Both of these radiographic signs have been reported in patients with SCA.[1,2,3,4,5] This may be attributed to bone marrow hyperplasia leading to bone resorption and reorganisation of the trabecular architecture.[3] We consider this OPG to be a useful example demonstrating the maxillofacial radiographic features of SCA.

Regarding the patient's dentition, there is no pathology of note. Interestingly, we noted two roots on both lower second premolars. Note that the lower incisors appear foreshortened and blurred due to the anterior mandible being out of the focal plane, secondary to a chin down position. This can be mistaken for external root resorption, which incidentally has increased prevalence in those affected by SCA.[6]

We suspect that the patient has been experiencing multiple episodes of vaso-occlusive events affecting the teeth and supporting structures. We have liaised with the consultant haematologist whom the patient is under the care of in order to make them aware of this aspect of the patient's disease.

We hope that this OPG together with the aforementioned non-specific history of pain provides useful information for clinicians to make reference to as a reminder of the oral effects of SCA which may present in general dental practice or secondary care dental services. Liaison with the patient's consultant haematologist in such cases is recommended and can help ensure effective management.

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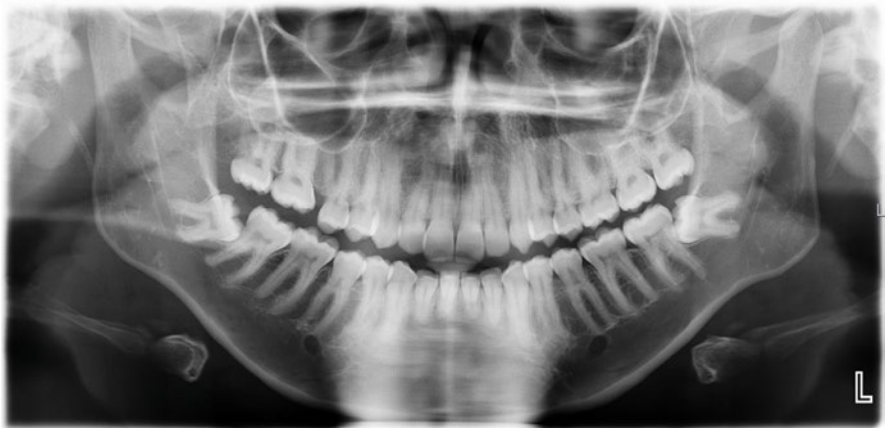

Fig. 2 Radiograph demonstrating increased radiolucency of the bone in the inferior body of the mandible

### References

1. Santos P, Machado P, Passos C, Aguiar M, Nascimento R, Campos M. Prevalence of orofacial alterations in sickle cell disease: a review of literature. *Braz J Oral Sci* 2013; **12:** 153-157.
2. Acharya S. Oral and dental considerations in management of sickle cell anemia. *Int J Clin Pediatr Dent* 2015; **8:** 141-144.
3. Dhir P, David C, Keerthi G. Radiographic manifestations of systemic diseases in jaw bones: a systematic review. *Asian Pac J Health Sci* 2014; **1:** 120-130.
4. Mourshed F, Tuckson C. A study of the radiographic features of the jaws in sickle-cell anemia. *Oral Surg Oral Med Oral Pathol* 1974; **37:** 812-819.
5. Sams D R, Thornton J B, Amamoo P A. Managing the dental patient with sickle cell anemia: a review of the literature. *Pediatr Dent* 1990; **12:** 316-320.
6. Souza S, de Carvalho H, Costa C, Thomaz E. Association of sickle cell haemoglobinopathies with dental and jaw bone abnormalities. *Oral Dis* 2017; **24:** 393-403.

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### Obstructed Stensen's duct

Sir, we present a case from our emergency department when we were asked to see a 65-year-old lady who had presented with a 'facial swelling'. The information that was passed on via the triage nurse was that the dentist had sent the patient in from general practice and the 'dental abscess' was very large. The patient's GDP thought that she had a fascial space infection of dental origin but a five day history of a progressively worsening swelling was reported.

There was no airway compromise but extra-oral examination revealed a significant right facial swelling. This was diffuse and extended from the lower eyelid to the lower border of the mandible. It was extremely tender to touch; the patient had a marked trismus and was clearly distressed. The masseter was grossly inflamed and firm to touch. Upon intra-oral examination the gingivae and teeth were unremarkable however, on the right buccal mucosa there was ulceration and a pinpoint opening; upon palpation of this area it was extremely tender and was draining pus.

Investigations revealed that the patient was apyrexial, her white blood cell count was minimally raised (7.2 x 109/L), however, the C-Reactive protein was very high (136 mg/L). A panoramic radiograph revealed an edentulous posterior right hand segment, and there were no signs of active pathology in the adjacent teeth (Fig. 1). However, a small radio-opaque structure was evident. It was not entirely clear whether this was of relevance.

A provisional diagnosis was that of a facial soft tissue swelling possibly related to obstruction of Stensen's duct and in turn the right parotid gland. Which, we theorised, had caused buccal mucosal swelling that was then traumatised. A dental cause was highly unlikely. However, the size of the facial swelling was still considerable and merited further investigation.

The patient was treated with intra-venous antibiotics and fluids and an urgent CT scan of the neck and face was arranged. This revealed an obstructing 6 x 3.3 x 2.5 mm calculus in Stensen's duct, significant right parotid gland swelling and right masseteric inflammation/ phlegmon accumulation - additionally, the scan revealed multiple reactive lymph nodes in multiple planes of the neck. In short, the patient had a right parotid sialadenitits secondary to an obstructed Stensen's duct.

Upon review the following day, the patient reported that a small hard ball like stone had fallen out into the mouth - this seemed to confirm the diagnosis of a salivary stone. Salivary stones are calcified masses within the salivary glands and/ or ducts,[1] they account for over 50% of salivary gland disease,[2] and the majority present in the submandibular glands, it has been reported that less than 10% of stones present in the parotid glands,[3] so this is an unusual case. Parotid stones are an equal mixture of both organic and inorganic substances with minimal bacterial involvement.[2] Pain is often secondary to salivary duct obstruction, bacterial ingress occurs with no protection from salivary fluid and the duct becomes a suitable environment for bacterial colonisation and proliferation,[3] as in this case. One must rule out other non-dental causes of facial swelling such as dehydration or radiation induced sialadenitis, mumps and an acute presentation of Sjögren's syndrome. With the correct radiographic investigations and exclusion of the above a definitive diagnosis can be achieved.

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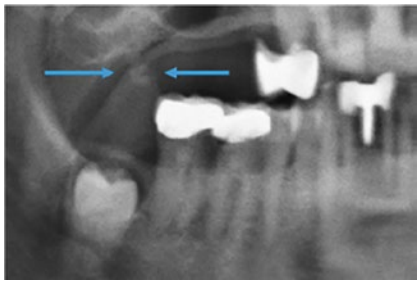

Fig. 1 Radio-opague mass visible in URQ

## References


1. Neville B W, Damm D D, Allen C A, Bouquot J E. *Oral and maxillofacial pathology.* 2nd ed. pp 393–395. Philadelphia: W.B. Saunders, 2002.
2. Zenk J, Benzel W, Iro H. New modalities in the management of human sialolithiasis. *Minimally Invasive Ther* 1994; **3:** 275–284.
3. Tepan M G, Rohiwal R L. Multiple salivary calculi in Wharton's duct. *J Laryngol Otol* 1985; **99:** 1313–1314.


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