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Incidental Finding of Sialolithiasis in the Sublingual Gland: A Diagnostic Dilemma

Abstract: This case describes a 35-year-old female who presented with an incidental finding of sublingual gland sialolithiasis. The clinical presentation, investigations and management as well as the pathology are described. This case highlights the diagnostic dilemma in determining the anatomical position of sialoliths on radiographs.

Clinical Relevance: To highlight the importance of incidental findings on radiographs and to make clinicians aware that radio-opacities on lower occlusal radiographs do not necessarily imply submandibular sialoliths but, rarely, may be sublingual sialoliths. This may warrant further investigations.

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Sialolithiasis is a common condition and occurs mainly in the submandibular gland (80–90%) and, to a lesser extent, in the parotid gland (5–20%). The incidence of stones in the sublingual gland is much lower, the incidence varying from 0–6.4%.^{1,2} The figure of 6.4% may represent misdiagnosis of stones in the anterior portion of Wharton's duct.

Symptoms associated with sialolithiasis may include swelling and pain of the involved gland, especially during mealtimes. The symptoms tend to subside and recur on a regular basis. Stones, however, may be completely asymptomatic and present as incidental findings on radiographic examination. We describe a patient with multiple sialoliths

in the sublingual gland who was initially thought to have multiple submandibular duct calculi.

Case report

A 35-year-old female was referred to the Oral and Maxillofacial Surgery Department with symptoms suggestive of TMJ dysfunction syndrome. An incidental radio-opaque lesion was seen on the orthopantomogram and hence a standard lower occlusal was taken (Figure 1). This revealed multiple stones in the floor of the mouth. The patient was completely asymptomatic and clinical examination was unremarkable. Both Wharton's ducts were patent with expression of clear salivary flow on gentle manipulation of the submandibular glands. The provisional clinical diagnosis at this stage was multiple sialoliths in the right submandibular gland and duct. Sialography was attempted but was not tolerated. The floor of the mouth was therefore explored under local anaesthesia but, owing to per-operative problems, no stones were recovered at the time. Further exploration was carried out under general anaesthesia at which



Figure 1. Lower occlusal radiograph showing multiple sialoliths in the floor of the mouth.



Figure 2. Surgical specimen of the sublingual salivary gland with multiple calculi.

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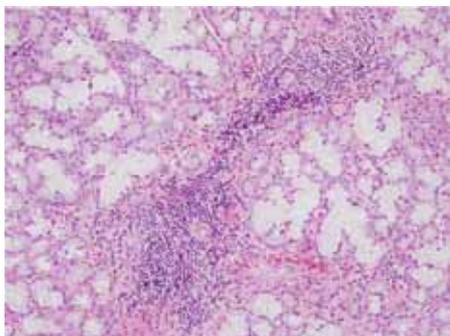


Figure 3. High power view (x20) of the sublingual gland showing chronic sialoadenitis with focal areas of acinar atrophy.²

time multiple sialoliths were found in the sublingual salivary gland. The right sublingual salivary gland, together with the five stones, the largest measuring 7x6x4 mm, were removed by an intra-oral approach (Figure 2). Macroscopic histology sections revealed multiple calculi whilst microscopic views showed chronic sialoadenitis with focal areas of acinar atrophy and duct dilatation (Figure 3). Post-operative recovery was uneventful.

Discussion

Salivary stones may arise from small concretions known as microliths

found within the intraglandular ducts. Intermittent stasis of saliva is thought to change the mucoid concentration, which in turn leads to stone formation.³ The subsequent obstruction of salivary flow may in some cases result in retrograde bacterial colonization.

The greater incidence of stones in the submandibular gland is thought to be related to its physiological characteristics. The secretions tend to be more alkaline and have a higher concentration of calcium and phosphate. However, the latter is not the only important factor in determining whether stones form within the gland. The concentrations of urea and protein also play an important role.

Sialoliths may be single or multiple in any major salivary gland. Lustmann *et al* reported that one sialolith was present in 75.3% of cases, two in 15.6%, three in 2.9% and four to eight (as in this case) in 6.2%.²

Traditional treatment for salivary calculi includes surgery. More recent treatments have used lithotripsy which aims to fragment the stone into small particles which can be expressed along existing ducts.⁴

This case shows the diagnostic dilemma in determining the true anatomical position of the sialoliths on

radiographs. Stones in the floor of the mouth are most commonly located within the submandibular gland or duct but, as in this case, may rarely be located within the sublingual gland. Further investigations, such as ultrasonography or contrast enhanced CT, may sometimes be indicated.

In summary, this was an unusual case of sialolithiasis of the sublingual gland presenting in a 35-year-old female. The gland and sialoliths were dissected and removed with preservation of Wharton's duct and lingual nerve.

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