CASE REPORT

Acquired Wharton's duct stenosis after dental radiographs treated with sialendoscopy

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Accepted 3 April 2015

SUMMARY

Salivary gland trauma may result in ductal stenosis and chronic sialadenitis. We describe a case of an 81-year-old woman with a history of intermittent left submandibular swelling that began after recent dental examination and radiographs. Diagnostic sialendoscopy was performed and demonstrated a near complete distal stenosis. It was determined that trauma experienced during dental radiography may have resulted in the patient's ductal obstruction and subsequent sialadenitis. This case illustrates an unusual presentation of non-sialolith-related ductal obstruction in the submandibular gland, treated with sialendoscopic-assisted sialodochoplasty.

BACKGROUND

The most common causes of benign salivary gland disease relate to infection and inflammation.¹ Sialadenitis is a cause of salivary gland swelling that most commonly develops from a sialolith obstruction of the ductal outflow tract but also may develop from ductal stenosis or stricture.^{2,3} As a pathological entity, salivary duct stenoses are relatively rare, as they are reported to be the inciting source of only approximately 15–25% of cases of sialadenitis overall.³ Observed to be most commonly located in the parotid ducts of women in the fourth to sixth decades of life,² stenoses occur in the submandibular duct in only a mere 25% of cases.⁴

Stenoses have shown associations with a variety of processes, including surgical manipulation of the oral cavity, presence of allergies, sialoliths, autoimmune disease and postinflammatory, but the true aetiology may be unclear.^{3,4} Stenoses typically cause symptomatology of painful glandular swelling that is cyclical in nature, flaring when the patient eats.³

Imaging studies are fundamental in the diagnosis of salivary gland disease. High-resolution ultrasonography is often the first-line imaging modality due to affordability, ease of access, safety and great spatial resolution. It is able to identify and help localise areas of stenosis based on hypoechogenic signals, which commonly present over areas on the masseter muscle.³

Sialendoscopy is a diagnostic and therapeutic modality for salivary gland pathology, including stenosis or strictures. By indirectly visualising the ductal system, sialendoscopy can assess tissue quality in addition to location, number and extent of obstruction. It is also useful as a means of intervention, and allows for procedural dilation of strictures.

CASE PRESENTATION

An 81-year-old woman with a medical history of hypothyroidism and hypertension presented with 4 months of left submandibular swelling that began after a particularly traumatic episode while obtaining dental radiographs using a standard intraoral digital radiographic sensor imaging apparatus (figures 1 and 2). She reported a foul salty purulent discharge that was aggravated on deep palpation of the gland and described symptoms of xerostomia but denied trismus and history of bruxism. She denied recent use of oral appliances. Her symptoms were intermittent and she had a temporal relationship with eating. Physical examination was significant for left submandibular gland swelling and overlying erythema, without tenderness; but otherwise unremarkable. The patient was started on a course of empiric antibiotics by her primary provider and referred for further evaluation. Prior serology and history were negative for autoimmune disorders such as Sjögren's disease, systemic lupus erythematosus and rheumatoid arthritis. History was specifically negative for any prior salivary gland symptoms preceding the dental trauma, prior history of radioactive iodine administration, external beam radiation or previous neck surgery.

Physical examination of the anterior floor of the mouth demonstrated a firm area of scar 1–1.5 cm proximal to the submandibular papilla. The mucosa was slightly pale in this region but otherwise normal appearing. Bimanual palpation of the left submandibular gland was performed and the duct was 'milked' forward. No saliva was noted to be coming from the papilla in the anterior floor of the mouth.

Figure 1 Close-up view of intraoral digital radiographic sensor.

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To cite: Kieliszak CR, Shokri T, Joshi AS. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-209117

Figure 2 Demonstration of the proper positioning of an intraoral digital radiographic sensor in a standardised patient.

INVESTIGATIONS

An in-office ultrasound was performed assessing grey-scale appearance and colour Doppler flow, and revealed atrophy of the left submandibular gland with homogeneous echotexture of the parenchyma, and dilation of the main ductal system

Figure 3 Transverse B-Mode ultrasound view of right submandibular gland demonstrating an example of ductal stenosis. Long arrow points to the posterior edge of mylohyoid muscle; short arrow points to area of stenosis. Note hypo/anechoic dilated duct just adjacent to the stenosis. While this image was taken from a different patient, it illustrates a similar situation.

(figure 3). No calculi were identified. Stenosis in the distal aspect of the submandibular duct was suspected, and a surgical plan for submandibular sialendoscopy, dilation and stent placement was discussed.

During sialendoscopy and floor of mouth exploration under local anaesthesia in the office, the papilla was positively identified with a 0.015-inch guidewire, but failed to effectively pass through Wharton's duct (figure 4). Attempts to dilate the duct using the Seldinger technique and salivary ductal dilators failed to dilate through the presumed area of stenosis. Diagnostic sialendoscopy with a 1.1 mm Erlangen sialendoscope was then attempted to visualise the lumen of the duct, and a near complete stenosis of the distal ostium of the duct was noted, roughly 1 cm from the papilla. The 0.015-inch guidewire was then introduced through the working channel of the sialendoscope and the stenosis was probed but could not pass (figure 5). At that point, with the sialendoscope in place, a proximal dochothomy to the stenosis was then performed and the duct was unroofed. Sialendoscopy was then performed through the proximal dochothomy and copious mucus plugging was noted and irrigated. No calculi were encountered. The ductal system was irrigated until the secondary and tertiary branches could clearly be identified, and again no sialolith was identified; the duct was irrigated and stented with an 18-gauge angiocath sutured to the floor of the mouth to help mature the dochothomy.

OUTCOME AND FOLLOW-UP

At 1-week follow-up, the stent was removed and the patient denied any further swelling of the left submandibular gland. Repeat sialendoscopy at 2 weeks demonstrated ductal patency with clear saliva emanating with bimanual pressure on the gland. The patient did not require any further procedures for treatment and the submandibular gland was entirely preserved. Long-term follow-up demonstrates the patient to be symptom-free at 18 months.

DISCUSSION

Salivary duct obstruction has been reported to be caused by sialoliths, fibrosis, allergic disorders, medication side effects, tumours, dehydration and radioiodine.⁵ Salivary duct stenoses, or strictures, are another important cause of salivary duct obstruction. Based on a 10-year retrospective study of 1362 sialograms, stenoses have been found to occur in nearly 25% of

Figure 4 Ductal probing performed in-office demonstrating stenosis. While this image was taken from a different patient, it illustrates a similar situation.

Figure 5 Sialendoscopy performed on the 81-year-old case report patient demonstrating near complete distal stenosis.

cases of salivary duct obstruction.² As a mechanical impairment of the ductal outflow tract, stenosis results in stagnation of salivary flow and propagation of infection, leading to clinical symptomatology. Stenoses require thorough evaluation and often surgical treatment.

While the exact aetiology of stenosis may be unknown, several associations have been reported,^{3 4} suggesting an association with any inflammatory process or trauma. Conservative measures such as hydration, sour candies, warm compresses and gland massages are not always effective for management of obstruction caused by ductal stenosis.⁴ Surgical treatment for recalcitrant cases involves dilation of the stenotic segment or bypassing the stenosis altogether. The choice of technique depends on the location of the stenosis and the degree of narrowing. The ultimate goal is to facilitate salivary flow and restore gland function.^{6 7} Surgical repair of the duct with either dilation or bypass offers an attractive, minimally invasive gland preserving alternative to en bloc resection.

Balloon ductoplasty can be used to dilate the duct at areas of stenosis and has been shown to be an effective technique, and in one prospective cohort, has shown success in greater than 90% of patients.⁸ Sialendoscopy offers the ability to localise stenosis as well as to deliver the balloon through the working channel and dilate the stenotic segment. Unfortunately, current technology requires a 1.7 mm sialendoscope for the delivery of the endoscopic balloon, which is often not effective for clinical use. Additionally, stenoses that are complete or near complete (as seen in this case) cannot be treated using a balloon technique, as the balloon cannot traverse the stenosis. An alternative is the use of serial salivary ductal dilators, although these tools are also limited for treatment of near complete stenoses.

If balloon or other dilation techniques are not feasible, transoral surgical bypass of stenosis may still be possible. A proximal doctotomy may be useful for strictures in the middle third or distal third of the duct, but proximal strictures have been found to be more amenable to sialendoscopic techniques.⁴

Marsupialisation of the duct proximal to the stricture helps to create a neo-ostium during transoral techniques. This may also be utilised to prevent stricture formation in cases of salivary gland trauma.⁹

Stents can be left in to help mature the doctotomy and aid in preventing re-stenosis. In this case, a simple angiocatheter was left in place, sutured to the floor of the mouth. There are also other commercially available products that can be used to accomplish similar goals. Examples include Cook salivary dilators, Schaitkin Salivary Duct Cannulas and Walvekar Salivary Stents. Other tools that are utilised include Cook introducer sheaths and infant feeding tubes, which can be left in safely to promote healing of the doctotomy. These latter two, however, are not dedicated nor validated instruments for stenting.

Learning points

- ▶ Sialadenitis can result from a variety of obstructive causes, including stenosis or stricture. This case presents a unique association of distal ductal stenosis treated with sialendoscopic-assisted sialodochoplasty presumably caused by trauma from dental radiographs, given the clinical history and presentation. No previous association has been documented in the medical literature to the best of our knowledge.
- ▶ A careful history, coupled with a thorough physical examination and proper procedural interventions may unearth the source of the obstruction.
- ▶ Revaluations are also important in ensuring the proper underlying aetiology of sialadenitis, and reassessment after procedural intervention ensures the success of a treatment.
- ▶ Early specialist referral in cases of sialadenitis is of paramount importance.

Acknowledgements Zachary C Riesenberger, DDS, for providing clinical images that were used in this report.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Jones RH, Findlay GJ. The management of benign salivary disease: a case series. *Aust Dent J* 2013;58:112–16.
- 2 Ngu RK, Brown JE, Whaites EJ, *et al.* Salivary duct strictures: nature and incidence in benign salivary obstruction. *Dentomaxillofac Radiol* 2007;36:63–7.
- 3 Kopec T, Szyfter W, Wierzbicka M, *et al.* Stenoses of the salivary ducts-sialendoscopy based diagnosis and treatment. *Br J Oral Maxillofac Surg* 2013;51:e174–177.
- 4 Koch M, Iro H, Kunzel J, *et al.* Diagnosis and gland-preserving minimally invasive therapy for Wharton's duct stenoses. *Laryngoscope* 2012;122:552–8.
- 5 Gillespie MB, Intaphan J, Nguyen SA. Endoscopic-assisted management of chronic sialadenitis. *Head Neck* 2011;33:1346–51.
- 6 Marchal F, Dulguerov P, Becker M, *et al.* Submandibular diagnostic and interventional sialendoscopy: new procedure for ductal disorders. *Ann Otol Rhinol Laryngol* 2002;111:27–35.
- 7 Marchal F, Kurt AM, Dulguerov P, *et al.* Histopathology of submandibular glands removed for sialolithiasis. *Ann Otol Rhinol Laryngol* 2001;110:464–9.
- 8 Drage NA, Brown JE, Escudier MP, *et al.* Balloon dilatation of salivary duct strictures: report on 36 treated glands. *Cardiovasc Intervent Radiol* 2002;25:356–9.
- 9 Lazaridou M, Iliopoulos C, Antoniadis K, *et al.* Salivary gland trauma: a review of diagnosis and treatment. *Craniomaxillofac Trauma Reconstr* 2012;5:189–96.

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