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CASE REPORT

Corrosive injury of oral cavity — a rare presentation

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Summary Corrosive injury of the upper gastrointestinal tract has been frequently reported in medical literature. The ingestion of acid or alkali may be accidental or intentional. The spectrum of injury varies from involving the oesophagus, stomach, and duodenum either individually or together. The oral cavity is rarely involved. The sequelae of oral cavity caustic injury that have been reported include microstomia, shallow vestibule, ankyloglossia, speech impairment, loss of teeth and impairment of facial expression. We report a case of corrosive injury of the oral cavity following accidental ingestion of caustic alkali, who presented to us with bilateral submandibular gland enlargement following the development of corrosive stricture of the submandibular ducts.

The scar tissue in the anterior floor mouth was excised with the submandibular salivary glands. Concomitant release of the contracture in the gingivo-buccal sulcus was also done and the defect was covered with a radial forearm free flap. The presentation of corrosive oral injury as salivary duct obstruction and enlarged salivary glands has not been reported. Free tissue transfer for corrosive scarring in the oral cavity helps to restore pliable tissue.

Case history

A 35-year-old male presented to our outpatient clinic in December 2004 with a history of having sustained burns in the oral cavity following accidental ingestion of a caustic alkali solution which he did not swallow. He presented to our

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outpatient clinic 2 months after this incident with difficulty in opening the mouth and peri-stomal tightness. He also noticed symmetrical swellings in the submandibular region on both sides. Clinical examination revealed enlargement of the submandibular glands bilaterally. An intraoral examination revealed induration and scarring in the floor of mouth with mild ankyloglossia. The lower bucco-gingival sulcus was obliterated due to scarring. He had drooling of saliva, though the orbicularis oris function was satisfactory. Diminished sensation in the scarred area of the lower lip was documented pre-operatively. Ultra-sonographic examination revealed submandibular salivary gland enlargement without any calculi in either the gland or the duct. The cause of the ductal obstruction was attributed to be due to the scarring around the ductal orifice and the terminal part of the submandibular salivary ducts. A review of his previous upper G.I. endoscopy revealed that he did not sustain any caustic injury of the hypopharynx, oesophagus, or stomach.

The surgical procedure involved release of the scar in the lower bucco-gingival sulcus (Fig. 1) and excision of both the enlarged submandibular glands (Fig. 2). A free radial forearm flap was used to resurface the resulting mucosal defect. The lateral ante-brachial cutaneous nerve was anastomosed to the end of the mental nerve on the right side. Though initially the lip was bulky (Fig. 3), he underwent debulking of the flap 2 months later. The scars of the excision of the mandibular glands on both sides had settled significantly (Fig. 4). Following the secondary procedure the final result was satisfactory (Fig. 5), with restoration of lip contour and creation of a deeper vestibule. On follow-up, he was relieved of the peri-stomal tightness and the symptoms of salivary drooling gradually subsided. He was able to eat

Figure 2 Enlarged submandibular salivary gland.

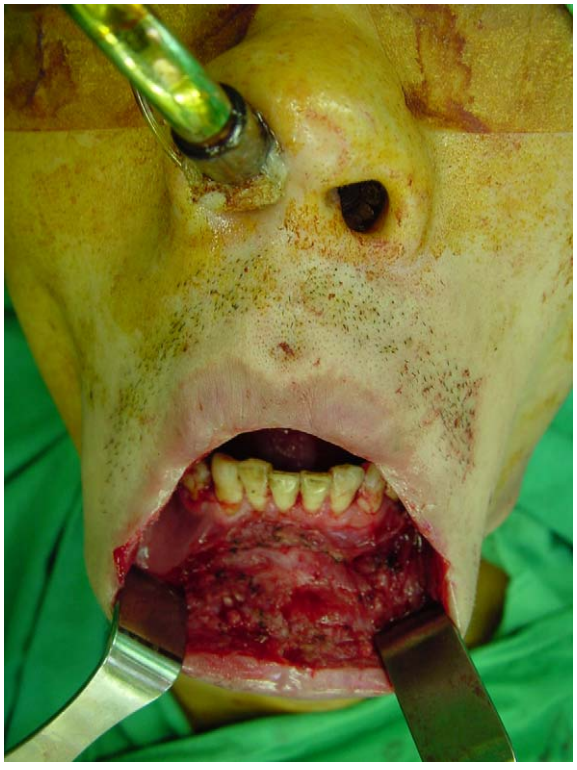

Figure 1 Defect following release of cicatricial tissue in the lower bucco-gingival sulcus.

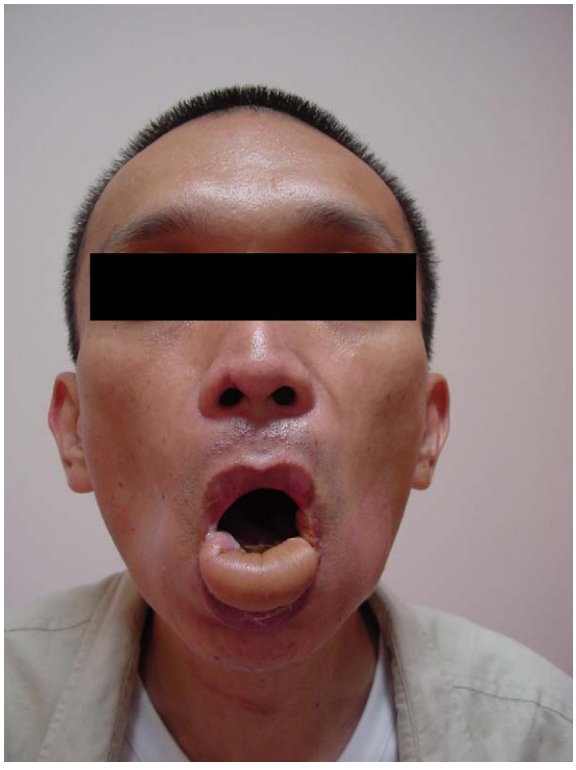

Figure 3 Early post-operative appearance after the radial forearm flap.

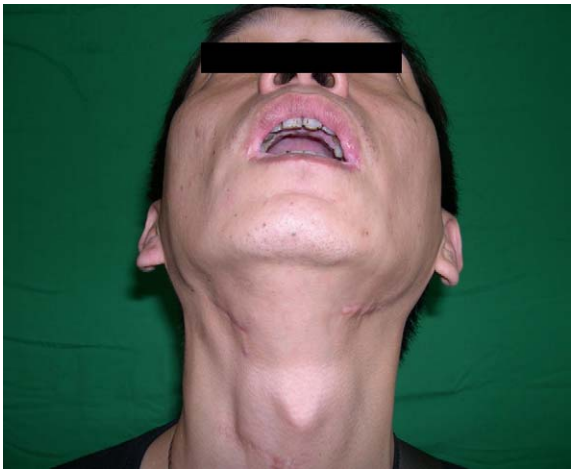

Figure 4 Scars of excision of submandibular salivary glands.

better, as his mouth opening had improved. He has been on regular follow-up for a period of 6 months and has been satisfied with the result.

Discussion

The commonly ingested alkalis are bleaching agents which are compounds with ammonia, sodium hydroxide, or potassium hydroxide. Alkalis are tasteless and odourless and often can be swallowed without much difficulty, unlike acids which are irritant and stimulate protective reflexes which prevent them from being swallowed easily. Acids have been observed to be less implicated in corrosive burns.[1] Following liquid caustic ingestion, most patients have some form of oro-pharyngeal burn, although some cases have severe upper gastrointestinal burns and yet no oral burns.[2] This may be related not to the chemical nature of the agent, but to the way it was swallowed. In this case the ingestion was accidental and a timely intervention resulted in him spitting out what he had taken, and hence the injury was restricted to the oral cavity. Other factors determining the severity of the burn are related to the concentration, quantity and the duration of contact.

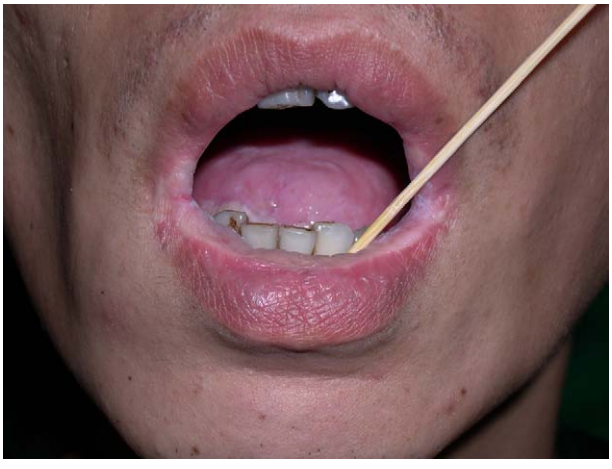

Figure 5 At 5 months after the first surgery: satisfactory colour match of the lip, and restoration of depth of bucco-gingival sulcus.

Most oral burn contractures have been addressed with release and skin grafting. Local flaps such as the facial artery musculo-mucosal flap (FAMM)[3] or the caudally based nasolabial flap can also be used. The FAMM flap however needs normal unscarred mucosa. An alternate option is the facial artery perforator flap as described by Hofer et al.[4] Scattered reports of using micro-vascular free flaps to bring in pliable tissue have been published. The flaps to be used for this purpose have to be thin and pliable and attempts with radial forearm flap and also jejunal patch have been done.[5] The present case report highlights two issues, namely the rare presentation of the oral burn and the use of free tissue transfer. The clinical presentation of caustic oral burn injury as salivary gland enlargement due to stricture of submandibular salivary ducts is unusual and has not been reported earlier. The stricture caused ductal obstruction and enlargement of the submandibular salivary glands. As the cicatricial tissue in the anterior part of the floor mouth had to be excised, the strictured segment of the ducts and the enlarged submandibular glands were excised. Strictures of salivary ducts have been treated with advancement ductoplasty and balloon dialation.[6] However, in the present patient, neither of these was possible as the ductal papillae could not be identified and excessive scarring around the area made identification of the ducts impossible. This necessitated the excision of the both the submandibular glands along with the scar tissue. Most reports on management of submandibular duct strictures have been associated with stones in the ducts and short segment strictures. Hence, these cannot be extrapolated to the treatment of large segment strictures.

Oral burns are often difficult to treat as the contractures are in multiple planes. Surgical release and skin grafting combined with splinting seem to be the common mode of treatment. This modality is frequently associated with secondary contracture. Hence, attempts to restore pliable lining using flaps like radial forearm flap, jejunal patch and adipo-fascial flaps have been done.

Not much study on oral burns has been done due to the fact that there is a variability of the type of involvement and the array of agents that produce a varied spectrum of injury making it difficult to formulate treatment guidelines. Hashem and Al Khayal[7] proposed a treatment guideline based on their classification of oral burns in children. According to their classification, Type I contractures included those involving the anterior part of oral cavity. Type II had contracture involved only the posterior part of oral cavity and Type III had involvement of the whole oral cavity. Although the classification serves as a useful guideline, the accompanying case series do not include the use of flaps as an option for resurfacing such a defect. This may be due to the fact that the case series included only paediatric population. The use of free tissue transfer is technically more difficult in this group. Free tissue transfer to bring pliable tissue for mucosal resurfacing has been often using the radial forearm flap.[5,8] In this case the scar tissue between the lower lip and the gingivo-buccal sulcus was released and the lining restored with a radial forearm free flap. Satisfactory contour was achieved after one debulking surgery, and restoration of the depth of the vestibule contributed to the decrease in drooling subsequently. The need for restoring the depth of vestibule also assumes importance in caustic injuries which have produced severe damage to teeth, and hence need major prosthetic treatment. Often this is not possible as there is diffuse scarring of the mucosa. The use of a flap also has the benefit that it does not need frequent follow-up, as against skin grafting which will need prolonged use of splints to prevent secondary contractures. Complex wire splints have been described to be of use in prevention of secondary contracture.[9] Martins et al. used local flaps to achieve depth of the vestibule.[10] The nasolabial flap was shown to be beneficial in this regard as shown by Hofstra et al.[11] They also evaluated the aesthetic outcome and the sensory recovery following the use of this flap and suggest that satisfactory results can be achieved with local pedicled flaps also.

The rare presentation of corrosive oral burns causing salivary duct obstruction has not been reported earlier. Treatment of oral burns can be satisfactorily achieved by selection of a pliable flap that can provide a stable lining and adequate vestibule depth. This will obviate the chance of secondary contractures as compared to skin grafts. Revision surgery may often be needed to achieve better aesthetic results.

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