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Plunging ranula following bilateral submandibular duct transposition

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Abstract
Submandibular duct transposition is now a standard surgical procedure for the treatment of severe drooling. However, this is our first experience of a plunging ranula arising as a complication of the technique. In the surgical management of this complication, the single most important step is excision of the sublingual gland to prevent recurrence.

Introduction

Bilateral submandibular duct transposition (BSDT) is routinely employed as the method of choice in our institution for the management of severe drooling in children (Bailey and Wadsworth, 1985). As 70 per cent of resting salivary flow is from the submandibular gland, BSDT is a physiologically acceptable and logical procedure. Success rates vary between 80–100 per cent with good long term results (Cotton and Richardson, 1981; Bruton *et al.*, 1990). Complications are few and include submandibular gland swelling and sialadenitis. There is an 8–12 per cent risk of ranula formation, but these have all been simple intra-oral ranulas. We describe here a case of a plunging ranula following BSDT and its surgical management.

Case report

A 10-year-old autistic child with poor oromotor control was first referred in 1987 for severe drooling. BSDT was carried out in November 1987 with good initial results. In June 1988, the girl presented with a right submandibular swelling which gradually increased in size but remained soft and without inflammation. Exploration of the right side of the neck was carried out in November 1988. A large, thin walled cyst was seen to extend from the hyoid backwards to the carotid sheath and up beneath the ascending ramus of the mandible to the base of the skull. The submandibular gland which was lying within the cyst was removed and the cyst excised.

Three months later, a further soft, transilluminable swelling developed in the submandibular triangle and this was explored again. A thin walled cyst surrounding normal structures was found. The cyst was removed as completely as was possible and the skin was tagged down to the underlying structures.

In September 1989, the swelling recurred below the previous incision and she was referred to our institution in November 1989. At that stage, it was noted that although the floor of the mouth appeared normal, there was a very large transilluminable, soft cyst filling both anterior triangles of the neck (Fig. 1). An MRI scan confirmed the cystic nature of the swelling; however, although the mass was very superficial inferiorly, being subcutaneous and superficial to the strap muscles, superiorly there was extension into the parapharyngeal space around the mandible (Figs. 2 & 3) in close approximation to the carotid sheath.

The question of further surgery was discussed with the child's family and with the proviso that there was no definite guarantee of success, consent for re-exploration was obtained.

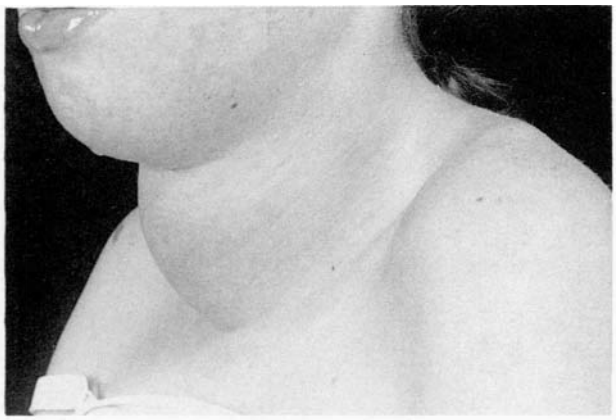

Fig. 1
Pre-operative picture showing large cyst involving both anterior triangles.

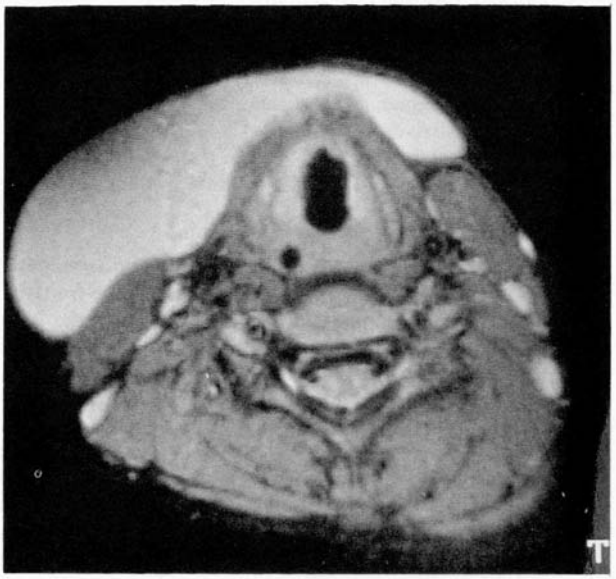

Fig. 2
MRI scan shows cyst lying superficial to strap muscles.

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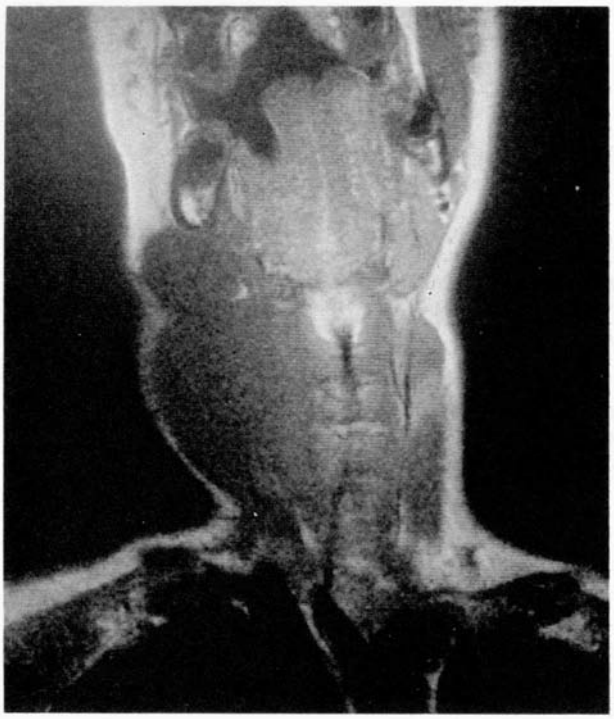

FIG. 3

Extension of plunging ranula into parapharyngeal space seen on MRI scan.

At the end of October 1990, the neck was explored via a suphrahyoid neck incision. A huge lobulated salivary pseudocyst was found immediately deep to the platysma taking origin from the right sublingual region. The cyst burst midway through the procedure but the flimsy lining was excised as completely as possible. Identification of the hypoglossal and lingual nerves was achieved with difficulty owing to dense adhesions from the previous surgery. A hugely hypertrophic right sublingual salivary gland was completely excised by the external route and the floor of mouth mucosa was repaired intra-orally. Histology of the specimens showed a normal salivary gland and a pseudocyst. Post-operatively, there was copious drainage from the operation site for the first two days but this decreased subsequently and the drains were removed on the fifth post-operative day. After suture removal on the seventh day, oral feeds were commenced, and the patient was discharged on the ninth post-operative day.

At Out-patient review six weeks post-operatively, the patient was symptom-free with no residual swelling (Fig. 4).

Discussion

Ranulas have been known since the time of Hippocrates and Celsius (Quick and Lowell, 1977). Taking their name from the Latin word Rana, meaning a frog and alluding to the blue translucent underbelly of these amphibians, they are cysts of the sublingual gland. Biochemical analysis of their mucinous contents reveals high protein and moderate amylase levels consistent with secretions from the mucinous acini of the sublingual gland (Roediger *et al.*, 1973). They are classified as either simple ranulas, which are intra-oral epithelial-lined cysts, or plunging ranulas that are in reality pseudocysts with a connective tissue lining only. Although their pathogenesis remains controversial (Van Den Akker *et al.*, 1978), there is general agreement that the simple ranula arises from partial obstruction of the distal end of the sublingual gland duct, whilst the pseudocyst forms from disruption of the duct with extravasation of saliva first into the floor of the mouth and then via a hiatus in the mylohyoid muscle into the fascial planes of the neck (Roediger *et al.*, 1973; Van Den Akker *et al.*, 1978).

With these basic rules of pathogenesis in mind, it is possible to postulate how a plunging ranula occurs after BDST, and indeed in our patient. Each sublingual gland drains via about 15 ducts, half of which open separately on the sublingual papillae and the other half into the submandibular duct (Last, 1972). These ducts are divided during surgery but usually re-establish drainage into the oral cavity as the floor of the mouth heals. Some of the ducts may become blocked by scar tissue during healing, in which case either atrophy of part of the gland may occur or else the duct will become dilated by continuing salivary secretion to form a simple ranula. In the case of the patient reported here, however, duct disruption must have occurred allowing saliva to leak into the surrounding tissues with consequent formation of a pseudocyst.

Several treatment modalities have been advocated for the plunging ranula. In one series an average of three operations was necessary before a ranula was cured (Van Den Akker *et al.*, 1978). Misconceptions regarding pathogenesis result in inappropriate treatment (Van Den Akker *et al.*, 1978; Black and Croft, 1982). Most authors agree that to prevent recurrence, the secretory source of the plunging ranula, the sublingual gland, must be removed (Roediger *et al.*, 1973; Khafif *et al.*, 1975; Harrison *et al.*, 1976; Van Den Akker *et al.*, 1978; Black and Croft, 1982). It is inevitable that the ranula will burst intra-operatively due to the thin false lining, but total removal of the cyst wall does not seem necessary and is sometimes technically difficult (Quick and Lowell, 1977; Van Den Akker *et al.*, 1978; Black and Croft, 1982).

Plunging ranulas are rare, but the possibility of occurrence after BSDT must be borne in mind. In view of its origin from the sublingual gland, it is mandatory that the gland be removed

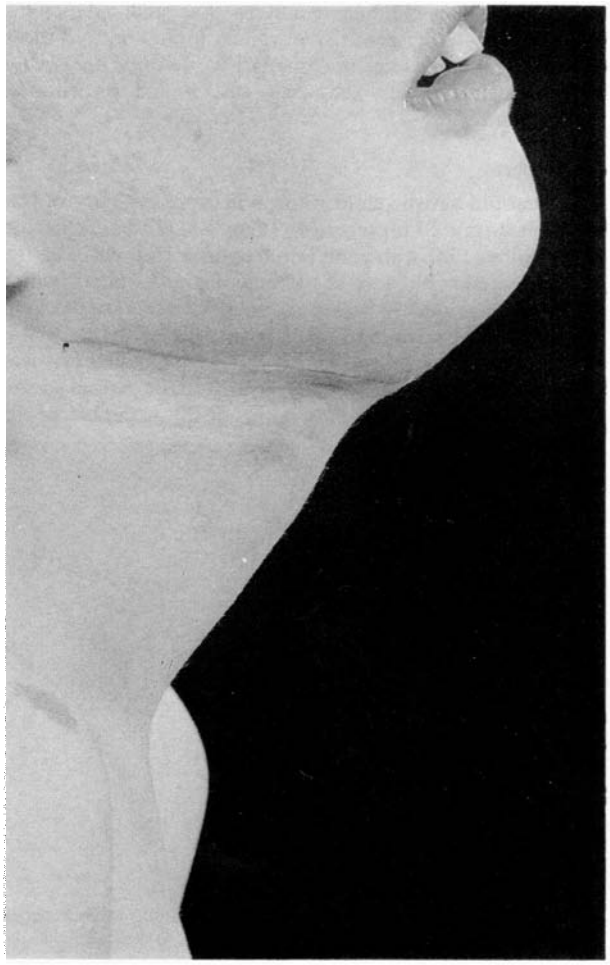

FIG. 4

Six weeks post operatively, child's neck shows no residual swelling.

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if recurrence is to be prevented. It has been suggested that in order to prevent ranula formation after BSDT, the sublingual glands should be routinely excised intra-orally as part of the procedure (Bruton *et al.*, 1991). However, in the authors' view this can hardly be justified in view of the low incidence of ranula formation, and the ease with which the usual, simple ranulas may be treated by intra-oral marsupialisation.

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References

Bailey, C. M., Wadsworth, P. V. (1985) Treatment of the drooling child by submandibular duct transposition. *Journal of Laryngology and Otology,* **99:** 1111–1117.

Black, R. J., Croft, C. B. (1982) Ranula; pathogenesis and management. *Clinical Otolaryngology,* **7:** 299–303.

Burton, M. J., Leighton, S. E. J., Lund, W. S. (1991) Long term results of submandibular duct transposition for drooling. *Journal of Laryngology and Otology*, **105:** 101–103.

Cotton, R. T., Richardson, M. A. (1981) The effect of submandibular duct re-routing in the treatment of sialorrhoea in children. *Otolaryngology-Head and Neck Surgery,* **89:** 535–541.

Harrison, J. D., Sowray, J. H., Smith, N. J. D. (1981) Recurrent ranula: A case report. *British Dental Journal,* **40:** 180–182.

Khafif, R. A., Schwartz, A., Friedman, E. (1975) The plunging ranula. *Journal of Oral Surgery,* **33:** 537–541.

Last, R. J. (1972) Anatomy, Regional and Applied. Fifth Edition. Churchill Livingstone: London. p 643.

Quick, C. A., Lowell, S. H. (1977) Ranula and sublingual salivary glands. *Archives of Otolaryngology,* **103:** 397–400.

Roediger, W. E. W., Lloyd, P., Lawson, H. H. (1973) Mucous extravasation theory as a cause of plunging ranulas. *British Journal of Surgery,* **60:** 720–722.

Van den Akker, H. P., Bays, R. A., Becker, A. E. (1978) Plunging or cervical ranula: Review of the literature and report of four cases. *Journal of Maxillofacial Surgery,* **6:** 286–293.

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