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Cervical ranula

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THE intra-oral ranula is a readily recognizable condition with characteristic localization and appearance. Although a variety of causes may produce a cyst-like lesion of the oral mucosa (Morf, 1905; Fitzwilliams, 1927; Catone *et al.*, 1969), the presence of a bluish, fluctuant, translucent swelling in the floor of the mouth, frequently with a history of spontaneous rupture and release of viscid mucus into the oral cavity, is practically synonymous with this diagnosis (Bailey, 1931; Mandel and Baurmash, 1957; Quick and Lowell, 1977). The intra-oral ranula is a benign condition, but may occasionally attain a considerable size, and cause displacement of the tongue with speech and swallowing difficulties (Morf, 1905; Caspersen, 1968; Catone *et al.*, 1969).

A less frequently recognized form is the plunging or burrowing ranula, which primarily presents as a swelling in the upper part of the neck, but may extend to the skull base or the supraclavicular region and upper mediastinum (Fitzwilliams, 1927; Bailey, 1931; Keen *et al.*, 1954; Crile, 1957; Catone *et al.*, 1969). The differential diagnostic possibilities in such cases are numerous, and the correct preoperative diagnosis is seldom made. Clinical awareness is probably at a low level since the ear, nose and throat literature contains only one report of ranulas presenting in the cervical region (Quick and Lowell, 1977). The present communication details our experiences with four cases of cervical ranula seen during a six-year period.

Report of cases

Case 1. A 10-year-old girl was admitted to the Department of Otorhinolaryngology in 1973 for treatment of a tumour in the right submandibular region which had gradually increased in size over a 6-week period. Both oral examination and submandibular sialography were unremarkable. The submandibular gland was exposed through a classical skin-crease incision, and a 2 cm diameter cyst identified lateral to Wharton's duct. The cyst extended deep to the mylohyoid muscle, contained tenacious mucus, and was removed together with the submandibular and sublingual salivary glands. It was not possible to decide clinically whether the cystic lesion was derived from the deep lobe of the submandibular or from the sublingual gland. There has been no evidence of recurrence in the subsequent six years.

Case 2. A 46-year-old female was first seen in 1973. She had been treated by irradiation in 1946 for a presumed salivary mass in the right submandibular area, and in 1953 the left submandibular gland had been removed, although she was unable to recall the previous symptomatology. She was apparently well until 1968 when she noticed a new swelling in the right submandibular region, which had subsequently persisted unchanged in size. On examination, a soft, 5×3 cm tumour was found in the anterior triangle of the neck, the lower pole having a more solid consistency. The mass extended down to the level of the thyroid

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cartilage, with the long axis in the vertical plane. There were no abnormalities in the floor of the mouth, nor evidence of systemic disease. Submandibular sialography was unsuccessful due to failure to cannulate the duct orifice. At surgery, a neck incision was made, and the upper part of the lesion was found to consist of a multilobulated, cystic mass which had displaced the submandibular gland downwards in the neck. The cystic mass was followed deep to the mylohyoid muscle, where it almost enveloped the lingual nerve and Wharton's duct, and was continuous either with the deep lobe of the submandibular gland or the sublingual salivary gland. Both glands were excised along with the cystic mass, and there has been no recurrence six years later.

Case 3. A 26-year-old female presented with a 2 week history of a gradually increasing swelling in the right submandibular region. She had been medically treated for a neck tumour 5 years previously, but was unable to recall the localization. A 4×5 cm cystic tumour was found in the right submandibular and submental triangles, but oral examination was negative. Sialography was not performed. At surgery, a large, cystic tumour was exposed inferior to the body of the mandible and deep to the investing fascia, which had displaced the submandibular salivary gland posteriorly. The main portion of the cystic mass was situated lateral to the plane of the mylohyoid muscle, but further dissection revealed that the mass had perforated the muscle approximately 2 cm anterior to its free posterior border. Following retraction of the muscle fibres, the neck of the cystic swelling was found to be continuous with the subjacent salivary tissue. Both salivary glands and the cystic mass were removed in continuity. There has been no evidence of recurrence, although it was considered unlikely at the time of operation that all prolongations of the cystic tumour had been removed.

Case 4. A 12-year-old girl was admitted for removal of a left submandibular tumour of ten weeks' duration. Oral examination was unremarkable. A swelling of approximately 2 cm diameter was palpated anteriorly in the left submandibular triangle, approximately equidistant from the symphysis menti and the angle of the mandible. Submandibular sialography indicated slight posterior displacement of the salivary gland but was otherwise unremarkable. The operative findings were practically identical to those described in case 3, with extension of the cystic lesion through the mylohyoid muscle. The submandibular gland displacement was less pronounced due to the smaller size of the lesion, which was removed in continuity with both salivary glands. There has been no recurrence since operation.

Histological examination revealed a pseudocyst in each case, the lumen being lined with granulation tissue. Only in case 3 were some areas covered with an epithelial layer. All submandibular glands were completely normal, the cystic lesions originating on the sublingual aspect of the junction between the two glands (Fig. 1).

Discussion

The lesion described in this presentation has been previously termed either the plunging (Bailey, 1931; Keen *et al.*, 1954; Crile, 1957) or burrowing (Fitzwilliams, 1927; Catone *et al.*, 1969) ranula. We have preferred the term cervical, thus emphasizing the localization of the mass lateral to the plane of the mylohyoid muscle, and the clinical presentation as a lump in the neck, with the

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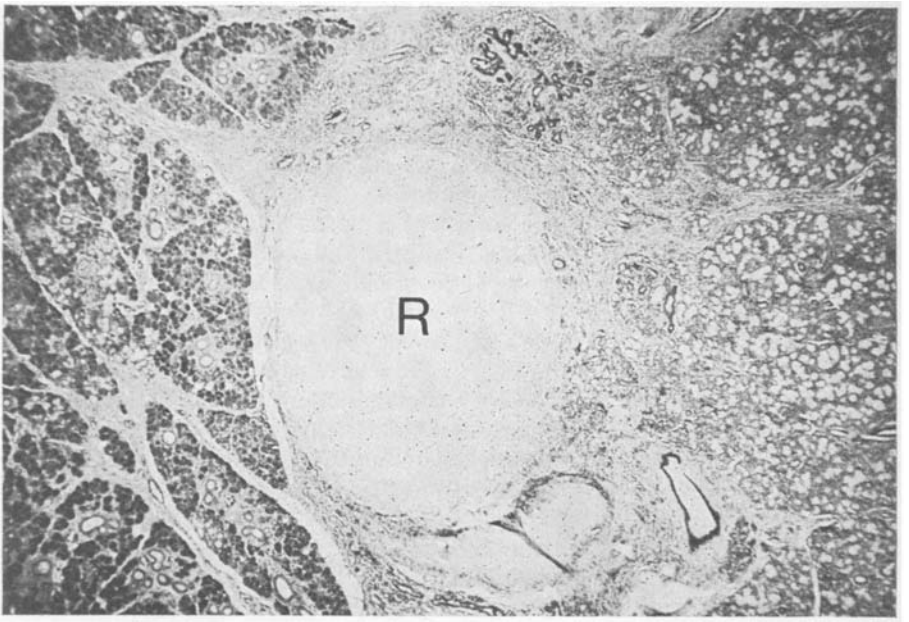


FIG. 1

Section from case 3 showing glandular tissue from the submandibular (left) and sublingual (right) salivary glands with intervening cystic loculus of the ranula (R). Haematoxylin and eosin, original magnification ×24.

differential diagnostic implications of the latter condition. The establishment of a correct diagnosis preoperatively is dependent both on an awareness of a condition's existence, and the results of specific investigational procedures. Unfortunately, there are no diagnostic tests for sublingual gland pathology, and none of the standard otorhinolaryngological texts contains any mention of this condition, the majority of references being found in the odontological and stomatological literature.

Clinical suspicion will be more readily aroused in those instances in which the cervical tumour is either accompanied by a typical ranula in the floor of the mouth, or there is a history of earlier intra-oral treatment for a ranula (Mandel and Baurmash, 1957; Whitlock and Summersgill, 1962; Rayne, 1973; Khafif *et al.*, 1975; Roediger and Kay, 1977). Preoperative diagnosis is, however, exceedingly unlikely when the cervical ranula is the sole anomaly (Quick and Lowell, 1977). Although none of our patients presented with intra-oral involvement, the correct diagnosis was suspected preoperatively in cases 3 and 4, our index of suspicion having been heightened by previous experience. A cervical approach has been used routinely in our four patients, since there is at present no investigation which would provide preoperative diagnostic confirmation.

Cervical ranulas occur comparatively commonly, but not exclusively as stated by Pattee (1969), in the South African Bantu (Keen *et al.*, 1954), and have been frequently reported in the canine veterinary literature (Glen, 1972). An interesting aspect of cervical ranulas is the preponderance of female patients. Perusal of the literature has produced twenty-two cases in which the patients'

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sex was specified, which, together with the four female patients from the present series gives a total of twenty-six documented cases. Only 23 per cent of these cervical ranulas have occurred in males, which is in marked contrast to the equal sex distribution of oral mucoceles derived from four larger series reported in the literature (Standish and Shafer, 1959; Cohen, 1965; Caspersen, 1968; Cataldo and Mosadomi, 1970). The reason for this difference in incidence between the two related conditions is at present unknown.

Although a branchial origin has earlier been proposed for the cervical ranula (Thompson, 1920; Bailey, 1931), it is now generally accepted that both the more common intra-oral and the cervical forms usually originate from the sublingual salivary gland (Fitzwilliams, 1927; Catone *et al.*, 1969; Roediger *et al.*, 1973; Roediger and Kay, 1977; Quick and Lowell, 1977). Further evidence has been provided by the ease with which Wharton's duct can be cannulated (Fitzwilliams, 1927), and by earlier reports of normal submandibular sialograms (Rayne, 1973). Both of our patients in whom submandibular sialography was performed had normal ductal architecture, and the submandibular glands from all four patients were unremarkable on histological examination. Biochemical analysis of the fluid content of cervical ranulas has also indicated similarities with the salivary secretion from the sublingual glands (Roediger *et al.*, 1973).

The sublingual salivary gland is situated below the mucosa of the floor of the mouth, and is related inferiorly to the mylohyoid muscle (Castelli *et al.*, 1969). In two of our patients (cases 3 and 4) the ranula had attained the neck by passing through the muscle, a finding which has been previously reported by other authors (Fitzwilliams, 1927; Mandel and Baurmash, 1957; Rayne, 1973). A possible explanation for this phenomenon is provided by earlier descriptions of dehiscences in the mylohyoid muscle (Malpas, 1927; Gaughran, 1963) and by the finding that portions of the sublingual gland may occasionally penetrate the muscle (Fitzwilliams, 1927; Gaughran, 1963; Castelli *et al.*, 1969). Cervical ranulas may therefore invade the neck by two routes (Fig. 2); either posteriorly, where they lie lateral to the lingual nerve and Wharton's duct, and will produce displacement of the submandibular gland if treatment is not instituted at an early stage (Whitlock and Summersgill, 1962; Khafif *et al.*, 1975; Quick and

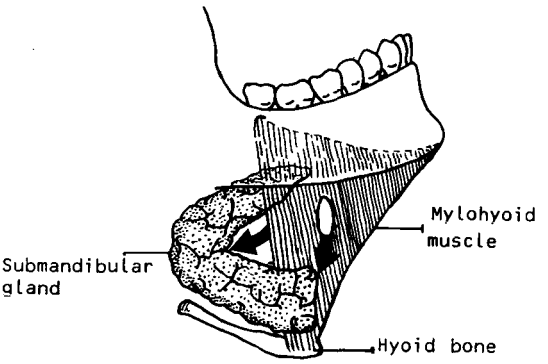


FIG. 2

The two pathways by which a ranula may invade the neck; either posterior to the free border of the mylohyoid muscle with displacement of the submandibular salivary gland, or through a dehiscence in the muscle.

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Lowell, 1977) or, less frequently, the ranula may perforate the mylohyoid muscle (Fitzwilliams, 1927; Mandel and Baurmash, 1957; Rayne, 1973). Local anatomical features probably determine the clinical development in each case.

The results of treatment of ranulas, both the intra-oral and the cervical forms, have frequently been discouraging, with a high incidence of multiple recurrences (Crile, 1957; Whitlock and Summersgill, 1962; Caspersen, 1968; Quick and Lowell, 1977). A wide variety of treatment modalities has been attempted, including simple incision, radiation, marsupialization, insertion of grommets, and attempts at total extirpation (Pattee, 1969; Keen *et al.*, 1954; Catone *et al.*, 1969). Meticulous dissection and removal of the entire 'cyst' lining is still regarded as an essential prerequisite of successful management (Khafif *et al.*, 1975; Quick and Lowell, 1977).

Although a ranula contains considerable quantities of tenacious mucus, the cavity is lined by non-secretory tissue elements. In similarity with oral mucous membrane mucoceles, the majority of ranulas are not true cysts since an epithelial lining can seldom be identified (Standish and Shafer, 1959; Cohen, 1965; Caspersen, 1968; Cataldo and Mosadomi, 1970). Most ranulas are therefore pseudocysts, and arise secondarily to mucous extravasation from the sublingual salivary gland (Caspersen, 1968; Roediger *et al.*, 1973; Roediger and Kay, 1977). The aim of treatment should therefore be removal of the secretory tissue from which the ranula originates, which, in the majority of cases, may be achieved by simple excision of the sublingual gland (Catone *et al.*, 1969; Roediger *et al.*, 1973; Roediger and Kay, 1977). This is successful both in the case of the intra-oral (Catone *et al.*, 1969; Caspersen, 1968) and cervical (Crile, 1957; Whitlock and Summersgill, 1962; Roediger *et al.*, 1973) ranulas, and time-consuming, and possibly destructive, dissection of the neck tumour ramifications is therefore unnecessary (Roediger and Kay, 1977). Simultaneous excision of the submandibular gland may be indicated either when the precise glandular origin of the cervical ranula cannot be determined, or in order to facilitate surgical access to the sublingual gland (Thompson, 1920; Patey, 1970). The problem in the management of cervical ranulas is not a question of treatment modality, but rather one of recognition and pre-operative suspicion.

Summary

Ranulas are mucous extravasation cysts, and usually originate from the sublingual salivary gland. They may occasionally infiltrate the tissue planes of the neck and present as a cervical tumour. The literature is reviewed, and four personal cases of cervical ranula presented. Successful treatment may be achieved in the vast majority of cases by removal of the sublingual salivary gland.

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