

Thoracic ranula: an extremely rare case

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Abstract

We present the first case of a thoracic ranula which originated from the left submandibular area extending into the subcutaneous tissue planes of the anterior chest wall. The patient had a history of surgery for a previous benign left salivary gland cyst, and presented with an enlarging mass in the anterior chest wall. This was a recurrence of a ranula, with an extension into the anterior thoracic wall. The thoracic ranula was excised, together with ipsilateral sublingual and submandibular glands, via a transcervical approach. No recurrence was detected over a 3-year post-operative follow up.

Key words: Ranula; Thorax; Sublingual Gland; Submandibular Gland

Introduction

Ranulas commonly occur in the floor of the mouth, confined to the sublingual space, but occasionally can 'plunge' through the mylohyoid muscle and present in the upper neck. They may also spill out into one or more adjacent areas. There have been earlier reports of plunging ranulas localized in the parapharyngeal space.¹ We present here the first ever described case, to the best of our knowledge, of a thoracic ranula found in the subcutaneous planes of the anterior chest wall.

Case report

A 19-year-old boy presented complaining of a gradually increasing mass in the anterior chest wall. He gave a history of an operation done for a small left neck lump which he was informed to be of 'benign origin', about a year ago. The swelling in the anterior chest wall had been increasing in

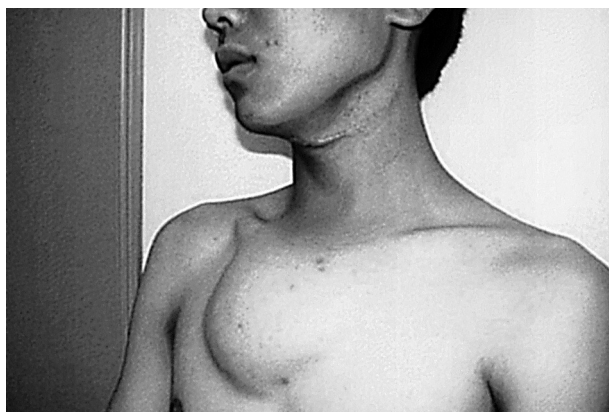


FIG. 1

Clinical photo showing the patient with the thoracic ranula in the anterior chest wall.

size over the past 6 months, and had gradually given him marked discomfort and embarrassment. There was no associated fever, pain or tenderness.

Clinical examination revealed an essentially normal otorhinolaryngological examination; the oral cavity, anterior and posterior nasal spaces were normal. The larynx and vocal cords were normal. There was a well healed horizontal cervical scar over the left submandibular area. All branches of the facial nerve were intact bilaterally. There was a soft, fluid-filled, fluctuant mass, 32 cm in diameter, within the anterior thoracic wall. It appeared to be in the subcutaneous plane, and was in continuity with the left cervical scar (Figure 1). There were no overlying skin changes or cutaneous fistulation.

An aspiration of the thoracic swelling was done under local anaesthesia. Two hundred millilitres of clear yellow fluid was obtained. This was sent for microbiological examination and biochemical analysis. There were no microorganisms cultured, and biochemical analysis revealed a large quantity of salivary amylase in the fluid.

A computed tomogram (CT) of the oral cavity, neck and thorax showed a collection of fluid in the subcutaneous plane, continuous with the left cervical submandibular region. There were no other masses or cervical lymphadenopathy noted on the scan. Haematological investigations, including a full blood panel and electrolytes, were all within the normal range. Treatment options were explained to the patient, but he preferred surgery.

Surgery was performed under general anaesthesia utilizing and extending from the previous surgical incision, two finger-breadths below the left jaw line. Subplatysmal flaps were raised to reveal both left sublingual and left submandibular glands intact. Both glands were carefully isolated and excised. The left lingual nerve, left mandibular branch of the facial nerve and left hypoglossal nerve were all preserved. All remnant fluid from the subcutaneous thoracic collection was removed and the thoracic ranula was excised in its entirety. The pathological report of the pseudocyst wall confirmed the histological diagnosis of a

mucocele. The patient recovered well post-operatively, and there was no recurrence after a 3-year follow up.

- **This is the first report of a thoracic ranula originating from the submandibular area**
- **The lesion was excised together with the ipsilateral sublingual and submandibular glands, and the patient was free of recurrence at 3 years**

Discussion

Ranulas are mucous extravasation cysts that usually originate from the sublingual gland.² They reflect a mucus escape reaction that develops after disruption of sublingual gland elements. As such, they comprise an accumulation of mucus within connective tissue and lack an epithelial lining.³ Two varieties are described: a simple or oral ranula found in the floor of the mouth, and a plunging or cervical ranula presenting as a neck lump. The precise aetiology of plunging ranulas is unknown, although local trauma, obstruction of its ducts and inherent mylohyoid dehiscence may play important roles.⁴ Bridger *et al.* have reported that, in many cases, a plunging ranula is iatrogenic and follows surgery to an oral ranula.⁵

Here, we present the first ever described case, to the best of our knowledge, of a thoracic ranula in a young adult with a history of surgery for pre-existing benign salivary gland disease. We postulate that this young man presented with an oral ranula a year ago and was treated surgically without complete removal of the sublingual gland, as evident by the finding of an intact gland intra-operatively. Hence, he presented with a recurrent cyst that had extended beyond the mylohyoid muscle into the anterior neck and further into the subcutaneous planes of the anterior thoracic wall.

The diagnosis of a ranula was made based on a combination of the elevated salivary amylase in the mucus-filled aspirate and the histological report of a lack of epithelial lining in the pseudocyst wall. We agree with various authors⁶ on the need for aspiration of the cyst to obtain a proper pre-operative diagnosis to guide appropriate therapy, especially in atypical presentations in which the clinical diagnosis is uncertain.

A CT scan further supported the diagnosis by showing accumulated secretions dissecting through the cervical and thoracic subcutaneous tissue planes, originating from the submandibular region. However, the so-called 'tail' sign, which is pathognomonic for plunging ranula,⁷ was not present.

Treatment options for plunging ranulas include excision of the cyst, incision and drainage of the cyst, marsupialization, and excision of the sublingual gland. Most authors agree that excision of the sublingual gland is the treatment of choice, with the lowest recurrence rate.^{2,4,8-13} Parekh *et al.* reviewed English literature from 1910 to 1987, covering 139 procedures in 89 patients with plunging ranulas, and reported that the recurrence rate was 85 per cent after excision of cyst in the neck, 70 per cent after incision and drainage, 53 per cent after marsupialization, and 2 per cent after excision of the sublingual gland.⁸ Another comparison, of three methods used to treat ranulas in 27 patients, made by Yoshimura *et al.* in 1995, showed a recurrence rate of 36.4 per cent after marsupialization, 25.0 per cent after excision of the cyst and 0 per cent after removal of the sublingual gland combined with excision of the cyst.⁹ Davison *et al.* reported

in 1998 that removal of the sublingual gland was necessary for the management of plunging ranula, while excision of the pseudocyst was probably unnecessary and placed surrounding structures at risk.⁴

Although complete removal of the mucocele lining appears to be unnecessary, removal of the mucus contents is essential. Thus, authors have advocated that removal of the sublingual gland should be followed by drainage and evacuation of the cyst contents.^{10,14} Mucoceles originating from the submandibular gland are extremely rare.⁷ However, Voerpahl and Schauss suggested that, in extensive cases, the additional removal of the submandibular gland is advisable to further lower the recurrence rate, even if the ranula originates from the sublingual gland.¹⁵

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