

## Submandibular salivary duct cyst mimicking an external laryngocele

G. W. BACK, F.R.C.S., F. FAHMY, F.R.C.S., A. HOSNI, F.R.C.S.

### Abstract

The clinical and radiological differential diagnosis of cystic lesions of the submandibular region can be difficult. We report an unusual case of a submandibular salivary duct cyst mimicking an external laryngocele on presentation by appearing to expand on Valsalva manoeuvre, and where computed tomography (CT) scanning was unhelpful in reaching a diagnosis. We present the case, discuss the theories of pathogenesis, and review the literature on the differential diagnosis of cystic lesions in the submandibular region.

**Key words:** Salivary glands; Laryngocele; Diagnosis, differential

### Case report

A 30-year-old military policeman was referred to the Otolaryngology clinic with a six-week history of a painless swelling in the left submandibular region which was only noticeable on swallowing and nose blowing. He denied any history of regional trauma, dental infection or viral illness. He was otherwise healthy, had no other symptoms, was a non-smoker and did not play a reed instrument.

On examination there was a non-tender, ill-defined, soft, 3 × 2 cm mass in the left submandibular region which expanded significantly on Valsalva manoeuvre. Examination of the floor of the mouth, including bimanual palpation, was normal, as was flexible fibre-optic pharyngolaryngoscopy. There were no other palpable masses and the remainder of the head and neck examination findings were normal.

Fine needle aspiration was subsequently performed. A small amount of hazy fluid with particles was aspirated and microscopic evaluation showed erythrocytes only.

The clinical findings suggested a fluid-filled external laryngocele and on advice of the radiologists a CT scan of the neck without contrast was performed to confirm the diagnosis and define its extent. Surprisingly, this was reported as showing a well-defined mass with relatively low attenuation arising within the left submandibular salivary gland (Figure 1). It had an average CT number of 33 HU (Hounsfield units), suggestive of a soft tissue mass rather than cyst fluid.

In view of the CT findings the patient was listed for elective submandibular gland excision. After dissecting the superficial portion of the gland free from the surrounding tissues and ligating the facial artery and vein however, a golf ball-sized, thin-walled cyst filled with tenacious, straw-coloured fluid arising deep to the mylohyoid muscle and in continuity with the submandibular duct was revealed (Figure 2). The submandibular gland itself had a normal appearance macroscopically. The cyst was resected together with the gland and the patient had an uneventful post-operative course. Histological evaluation of the

specimen confirmed a normal submandibular salivary gland and showed a salivary duct cyst lined by stratified epithelium with lymphohistiocytic infiltrate present in the cyst wall, and intimately associated with salivary gland tissue with a moderate infiltrate of lymphocytes, plasma cells and occasional lymphocytic aggregates. The smaller ducts were mildly dilated and there was some periductal

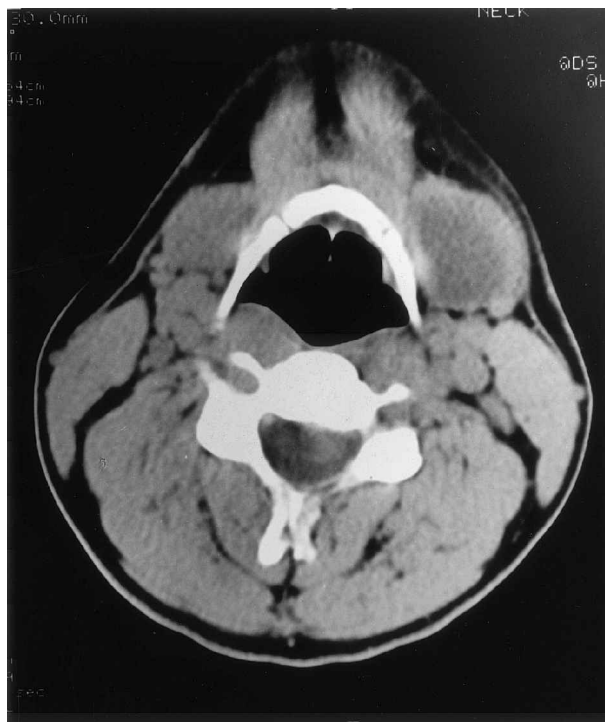


FIG. 1

Axial CT scan without intravenous contrast showing well-defined mass within left submandibular gland.

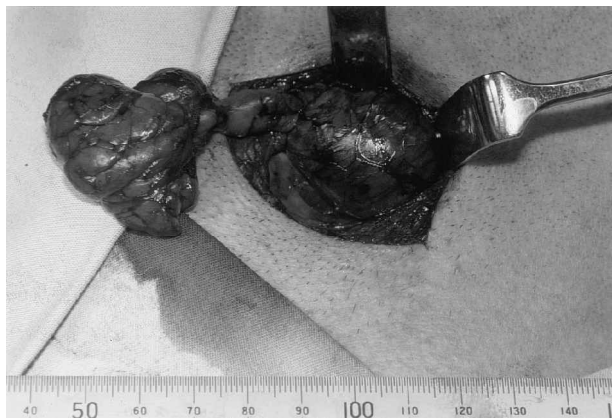


FIG. 2

Large submandibular duct cyst presenting at posterior border of mylohyoid muscle after superficial and deep portions of left submandibular gland have been dissected free from surrounding tissues.

inflammation (Figure 3). There was no evidence of malignancy.

### Discussion

Salivary gland cysts are not uncommon. It is the most common lesion of the minor salivary glands and comprises five per cent of all parotid lesions. Submandibular gland cysts, however, are rare.<sup>1</sup> The differential diagnosis of cystic masses in the submandibular region includes branchial cyst, dermoid/epidermoid cyst, plunging ranula, mucocele, external laryngocele and abscess. Rare cases of hydatid cysts and cystic hygromas of the submandibular gland have also been reported.<sup>2,3</sup> A single case of submandibular gland pneumatocele in a 15-year-old boy in whom dilatation of the submandibular duct was involved, has also been described. Here recurrent soft-tissue swelling was produced by the passage and retention of air in the gland parenchyma secondary to increased intrabuccal pressure associated with glass-blowing.<sup>4</sup>

In our case the clinical findings of a soft, lateral neck mass expanding on Valsalva manoeuvre initially suggested an external laryngocele.<sup>5</sup> Partially or completely fluid-filled laryngoceles have also been reported in the literature.<sup>6</sup> Subsequent surgical exploration however, revealed a large



FIG. 3

Photomicrograph demonstrating epithelium-lined cystic cavity (centre), intimately associated with salivary gland tissue (lower right) (H & E;  $\times 4$ ).

submandibular duct cyst and normal submandibular gland. The prominent submandibular swelling on Valsalva manoeuvre was most likely due to the cyst being pushed out under the posterior border of the mylohyoid muscle by contraction of the floor of mouth muscles, especially the hyoglossus, that forms the floor of the deep portion of the gland and proximal submandibular duct.

The CT scan, reporting a solid mass within the submandibular gland, was not helpful in reaching a diagnosis, but the radiological differential diagnosis of cysts in the submandibular region can be difficult and the possible origin of such a cystic mass from the submandibular gland is not always obvious.<sup>7</sup> In our case the CT showed the mass to be homogeneous and to have a CT number of 33 HU. The CT number is a measure of CT density, which reflects the contents of the lesion. An analysis of 35 submandibular cystic lesions consisting of 20 ranulas, seven dermoids and eight lateral cervical cysts undertaken by Kurubayashi *et al.*<sup>8</sup> to assess the usefulness of CT in their differential diagnosis showed that all the lesions were homogeneous on CT and that the CT density in all ranulas, that have the same content as submandibular salivary gland cysts, ranged from +10 to 39 HU. This was the same for dermoid cysts. They concluded that CT density was not very useful for the differential diagnosis of submandibular cystic lesions.<sup>8</sup> An epithelium-lined true cyst arising from submandibular salivary duct with a normal gland, in an adult, has to our knowledge not been previously reported in the English literature.

A plunging ranula – a cystic lesion of sublingual salivary gland origin, attributed to duct obstruction leading to dilatation and rupture of the duct, with fluid dissecting between the fascial layers of the floor of the mouth into the submandibular region, where it forms a pseudocyst – seldom presents as a submandibular mass with no visible intra-oral connection.<sup>9</sup>

Eleven cases of congenital dilatation of the submandibular duct have been reported in the literature, but they were all secondary to atresia of the duct ostium, all occurred in infants and all presented with a cystic lesion in the anterior floor of mouth without submandibular swelling.<sup>10,11</sup> In our case histological evaluation of the specimen showed inflammatory changes in and around the submandibular duct cyst. Inflammation has been shown to cause epithelium-lined retention in mucocoeles resulting from partial obstruction of the duct, and this may well have been the cause of the cyst in our case, although unusually the cyst was in continuity with the submandibular duct itself and the gland was unaffected, in contrast to what has been described in the literature.

### References

- 1 Surkin M, Remsen K, Lawson W, Som P, Biller F. A mucocele of the submandibular gland. *Arch Otolaryngol Head Neck Surg* 1985;**111**:623–5
- 2 Simons JN, Beahrs OH, Woolner LB. Tumors of the submaxillary gland. *Am J Surg* 1964;**108**:485–94
- 3 Singh S. Submandibular hydatid cyst. *J Laryngol Otol* 1972;**86**:647–50
- 4 Capova L. Pneumatocele glandulae submandibularis. *Cesko-Slov Otolaryngol* 1975;**24**:116–7
- 5 Ojala K. External, asymptomatic laryngocele without known predisposing factors in middle-aged man. *J Laryngol Otol* 1983;**97**:767–9
- 6 Johnson TH Jr, Fioranelli RJ. Laryngeal cyst (filled laryngocele): laryngographic diagnosis. *Radiol* 1969;**93**:875–7
- 7 Van der Goten A, Hermans R, Smet MH, Baert AL. Submandibular gland mucocele of the extravasation type: report of two cases. *Pediatr Radiol* 1994;**25**:366–8

- 8 Kurabayashi T, Ida M, Sasaki T. Differential diagnosis of submandibular cystic lesions by computed tomography. *Dentomaxillofac Radiol* 1991;**20**:30–4
- 9 Quick CA, Lowell SH. Ranula and the sublingual salivary glands. *Arch Otolaryngol Head Neck Surg* 1977;**103**:397–400
- 10 Pownell PH, Brown OE, Pransky SM, Manning SC. Congenital abnormalities of the submandibular duct. *Int J Pediatr Otorhinolaryngol* 1992;**24**:161–9
- 11 Addante RR. Congenital cystic dilatation of the submandibular duct. *Oral Surg* 1984;**58**:656–8

Address for correspondence:

Mr G. W. Back,  
37 Greenways,  
Thorpe Bay,  
Southend-on-Sea,  
Essex SS1 3BS, UK.

---

Mr G. W. Back takes responsibility for integrity of the content of the paper.

Competing interests: None declared.

---