

## Case report: Sublingual epidermoid cyst in an elderly patient

The occurrence of epidermoid cysts in the floor of the mouth is uncommon, particularly in elderly patients. We present the case of a 77-year-old female, who presented with minimal symptoms, despite a large floor-of-mouth swelling, which obscured her tongue from vision. The mass was removed via an intraoral approach under local anaesthesia (LA) and intravenous (IV) sedation, keeping complications and recovery time to a minimum. The mass was revealed to be an epidermoid cyst and the patient made a swift recovery. The features and classification of these cysts are discussed.

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### Case report

A 77-year-old female was referred by her general dental practitioner (GDP) to the accident and emergency (A and E) department of Dublin Dental University Hospital (DDUH). The patient's dentist had noted an incidental finding of a swelling 'the size of an apple' in the patient's floor of mouth. The patient reported that the swelling had been present and unchanged for around three years. The patient presented with no dyspnoea, dysphagia, or pain, and seemed unconcerned by the swelling. The patient's family did report a slight change in speech over recent years – she had developed a slight lisp. There was no history of previous trauma or surgery to the region. Medically, the patient had hypertension and osteoarthritis, affecting the knees in particular. She took: indapamide; a thiazide-like diuretic; and, etoricoxib, a selective COX-2 inhibitor. She had no known drug allergies, never used tobacco, and drank around five units of alcohol per week. This placed her into class II of the American Society of Anesthesiologists physical status classification system. Examination revealed no cervical lymphadenopathy. The submandibular glands were unremarkable, with normal saliva drainage. The skin of the face and neck

appeared normal. The cranial nerves were intact. The intraoral swelling was displacing the floor of mouth tissues inferiorly, causing a double chin appearance. Intraoral examination revealed a large solitary midline swelling in the floor of the mouth occupying both sublingual spaces. Dimensions of 50mm by 50mm by 20mm were noted. The swelling had normal overlying mucosa and was slightly fluctuant, with a 'doughy' consistency. On initial inspection, the tongue was barely visible, being displaced posterosuperiorly by the swelling. The swelling moved minimally on swallowing or protrusion of the tongue. There were no signs of inflammation.

Imaging was not deemed necessary, as there was no sign that the swelling had herniated through the muscles of the floor of the mouth. If features of herniation had been present, magnetic resonance imaging (MRI) would have provided good visualisation of the swelling's relation to the surrounding structures.

Given the clinical appearance of the swelling, the two most likely diagnoses were felt to be a floor-of-mouth cyst, or a ranula. The swelling lacked the bluish hue typically seen with a ranula, and the consistency was more indicative of a floor-of-mouth cyst. A full list of differentials can be seen in **Table 1**.

**Stephen Anderson**

BDS MFDSRCSd

Lecturer in Emergency Dental Care

**Leo FA Stassen**

FRCS (Ed) FDS RCS MA FTCD

FFSEM(UK) FFD RCSI

Consultant Oral and Maxillofacial  
Surgery

Address for correspondence:

**Stephen Anderson**

Division three,

Dublin Dental University Hospital,

Lincoln Place,

Dublin 2.

T: (086) 446 5966

E: Steve85a@gmail.com



FIGURE 1: Swelling pre-operatively – patient sedated.



FIGURE 2: Mucosal incision.



FIGURE 3: Cyst enucleation.



FIGURE 4: Wound bed with surgical.

Options for anaesthesia included LA, LA with conscious sedation, or general anaesthesia (GA). Following discussion with the patient, the decision to operate under LA with conscious sedation was made (Figure 1).

Following administration of intravenous midazolam and dexamethasone, bilateral inferior alveolar nerve blocks were administered. This was supplemented with infiltrations around the periphery of the swelling. Aspiration of the swelling with a 21-gauge



FIGURE 5: Incision closure.

needle ruled out a vascular lesion. A midline mucosal incision was made over the swelling (Figure 2). Blunt dissection through the pericapsular tissue was performed, taking care in the regions of the submandibular ducts and lingual nerves bilaterally. Sharp and blunt dissection were used to expose the entirety of the cyst, and traction and counter-traction used to free and remove the cyst (Figure 3). No herniation through the mylohyoid was noted. The mass was delivered intact. The wound was closed in layers using resorbable suture materials (Figure 4 and 5). The specimen was sent for histopathological examination.

Table 1: Differentials

- Ranula
- Obstructive sialadenitis
- Infective sialadenitis
- Thyroglossal duct cyst
- Lymphatic malformation
- Arteriovenous malformation
- Cystic hygroma
- Branchial cleft cyst
- Sublingual/submandibular cellulitis/abscess
- Benign/malignant tumours of mucosa or salivary glands
- Lymphadenopathy/lymphadenitis



FIGURE 6: Specimen.

The specimen measured 80mm by 20mm by 25mm (Figure 6). The maximum thickness of the wall of the cyst was 2mm. The cyst contained a yellow, gritty, paste-like material. Histology revealed an epidermoid cyst with prominent multinucleated giant cell reaction to keratin.

At review two months later, the area had healed well (Figure 7). The lip had resolved. There was no residual-altered sensation and the submandibular glands continued to function normally. The patient had complete perioperative amnesia. She was very pleased with her overall experience.

## Discussion

The term 'dermoid cyst' causes some confusion, as it has historically been used differently by different specialties. Some authors use the term dermoid *in lieu* of teratoma. Others define a dermoid as a subcutaneous, congenital cyst. Meyer (1955) classified congenital cysts of the floor of the mouth as either epidermoid, dermoid, or teratoid<sup>1</sup>, according to their histological features:

**EPIDERMoids** - inclusion cysts of ectodermal origin, consisting of a thin connective tissue wall, lined by stratified squamous epithelium. Desquamated keratin may fill the cyst cavity. No dermal appendages are found within the underlying connective tissue.

**DERMOID CYSTS** - inclusion cysts of ectodermal origin, lined by stratified squamous epithelium. They may contain all the more complex tissues derived from ectoderm (Table 2).

**TERATOMOID CYSTS** - true neoplasms arising from misplaced embryologic cells. They are also lined with stratified squamous epithelium. They contain tissues that are histologically diverse and represent more than one of the embryonic germ layers, and sometimes all three.

However, usage of the terms epidermoid, dermoid and teratoid cysts today implies the following:

**EPIDERMoid CYST** – a common cutaneous cyst, frequently found in adults. They are derived from the ectoderm and are lined with squamous epithelium. They may be congenital or acquired. The



FIGURE 7: Two-month review.

acquired form is thought to result from traumatic or iatrogenic implantation of cells from the epidermis into the dermis. They tend to form slowly-expanding, unilocular masses, and are frequently identified during the third and fourth decades.

**DERMOID CYST** – an embryologically derived squamous epithelium-lined cyst. They are a congenital lesion of ectodermal origin, containing ectoderm derivatives including dermal appendages. They also tend to form slowly expanding, unilocular masses. Dermoid cysts tend to manifest earlier, usually in the second or third decade. However, cases have been described in a child of seven months<sup>2</sup> and in a 77-year-old female.<sup>13</sup>

**TERATOMOID CYST** – a germ cell tumour with a cystic component. They contain tissues that are foreign to the primary site of origin, or are histologically diverse and represent more than one of the embryonic germ layers.<sup>4</sup> They may present during infancy or during the second to third decade,<sup>5</sup> and tend to form multiloculated masses.

All three types of cyst contain a greasy, cheeselike, white/gray/tan material. Dermoid and teratoid cysts may also contain fragments of hair, nails, or dental enamel in their lumen.<sup>6</sup>

Epidermoid and dermoid cysts are encountered throughout the body. About 80% are found in the sacral region and ovaries. While only about 6.94% are in the head and neck, and only 1.6% in the floor of mouth. The most common oral site is the floor of the mouth, although they may rarely be encountered in other soft tissue locations, and rarely in the mandible or maxilla.<sup>7,8,9</sup>

Congenital floor of mouth cysts are thought to arise from entrapment of ectodermal tissue in the midline during fusion of the first and second brachial arches, during the third and fourth embryonic weeks<sup>10</sup>. Acquired forms are likely to be a result of either iatrogenic or traumatic implantation of epidermal fragments into the underlying tissues. Other hypothesis include the possibility that floor of mouth cysts may represent a variation of the thyroglossal duct cyst.<sup>11</sup>

Floor of mouth cysts generally present as slow growing, painless, doughy/fluctuant swellings. They are asymptomatic until large

enough to cause dysphonia, dysphagia or dyspnoea. The tongue will be displaced poster-superiorly, and the patient may present with a double chin, particularly if the mass sits superficial to the mylohyoid. Treatment is by surgical enucleation. Surgical approach is determined by the cyst's relationship to the muscles of the floor of the mouth, particularly the mylohyoid. The mylohyoid muscle separates the sublingual space from the submental and submandibular spaces. If the mass is in the sublingual spaces deep to mylohyoid, an intraoral incision is preferred. Care must be taken to preserve the lingual nerve and the submandibular ducts. The mass may herniate through the muscle, causing a 'plunging dermoid'<sup>12</sup> occupying both the sublingual and submental/submandibular spaces. If the mass lies superficial to mylohyoid, a submental/submandibular extraoral incision is required. The incision is made in a natural skin crease, but some degree of scarring is inevitable. There is a risk of damage to the marginal mandibular branch of the facial nerve. Care is taken to avoid rupture of the cyst, as the contents may be irritant, causing post-operative inflammation. Once removed, recurrence is unlikely. There are sporadic case reports of malignant transformation arising in the lining of dermoid, epidermoid and teratoid cysts.<sup>13</sup> One case of a squamous cell carcinoma in the lining of an epidermoid cyst in the sublingual gland has been reported.<sup>14</sup>

## Conclusion

There are a number of case reports of floor of mouth cysts presenting in the fifth, sixth, and seventh decades of life. This is, however, an uncommon presentation, as the majority of floor of mouth cysts are congenital. The aetiology in this case is unclear, with no history of trauma to the face or intraoral operative procedures.

During clinical examination, the posterior extent of the lesion was palpable, and the submandibular spaces appeared unaffected. Imaging was therefore deemed unnecessary. Despite the large size of this lesion (70mm by 20mm by 25mm), an intraoral approach was adopted to avoid the risks of scarring and facial nerve damage.

Conscious sedation offers several advantages over general anaesthesia. General anaesthesia carries the risk of pulmonary embolus and deep vein thrombosis, which often necessitates anticoagulation with a low molecular weight heparin. There is a fairly high risk of post-operative nausea, vomiting, and sore throat. Conscious sedation generally uses one drug to cause a central nervous system depression, whilst the patient remains conscious, is able to understand and respond to verbal commands, and the protective pharyngeal and laryngeal reflexes are preserved. Lower extremity muscle tone is at an adequate level to greatly reduce the risk of deep vein thrombosis and pulmonary embolism. Orotracheal/nasotracheal intubation is not required, lowering the risk of sore throat. Lower overall doses of narcotics are used and inhalation anaesthetics are avoided, resulting in less post-operative nausea and vomiting. All these factors allow a faster recovery and earlier discharge.<sup>15</sup>

The combination of conscious sedation and an intraoral approach resulted in a fast recovery time, earlier discharge and a high-perceived level of patient satisfaction.

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