A massive sublingual—cervical dermoid

By N. K. Soni and P. Chatterji

Sublingual dermoids are of special interest to the oral surgeon and the oto-laryngologist. These cysts are comparatively rare. New and Erich (1937) recorded only 24 in the floor of the mouth out of 1495 dermoid cysts diagnosed at the Mayo Clinic over a period of 25 years; of these, only 6·9 per cent were in the head and neck, and of these sublingual dermoids constituted approximately 23 per cent. A similar distribution was found in a smaller series of 50 cases, reported by Brownstein (1973). McAvory and Zuckerbraun (1976) recorded only one case of sublingual dermoid in their series of 33 dermoid cysts in the head and neck.

Sublingual dermoids are of major clinical importance because they may attain a large size when hidden in the floor of the mouth, and can thus impair normal deglutition or speech, or even manifest themselves as lesions interfering with the airway. Resouly (1976) reported a single case of a very large sublingual dermoid. Another case, measuring $17 \cdot 5 \times 8$ cms, was reported by Mukerjee *et al.* (1977). Recently we have seen a case of an extensive sublingual dermoid which was confined above by the floor of the mouth but also had a very marked extension into the neck. Such a large sublingual–cervical dermoid does not appear to have been reported hitherto.

Case report

A 60-year-old female was admitted with a huge swelling of the neck, which had been present for about 7 years. An operation for the same swelling had been carried out in one of the district hospitals six years earlier. About two months after the initial operation, she had again noticed some swelling at the same site which had gradually increased in size, but did not cause her much trouble. Suddenly the swelling began to increase rapidly in size and it extended more into the floor of the mouth 6 months prior to her hospitalization. At the same time she developed some change in her speech and also felt pain in the swelling. There was no history of fever, nor any suggestion of malignancy.

On examination, the patient was of average height, but poorly nourished and anaemic. The liver and spleen were not palpable. The heart and the lungs were normal on clinical examination. There was no cervical lymphadenopathy. Her speech was slow, and she pronounced many consonants with difficulty.

A huge external swelling was present in the submental area, i.e. above the larynx (Figs. 1 and 2). There was no pulsation, no venous engorgement, and no pigmentation over the swelling. A 5 cm-long transverse scar was seen over the swelling. The overlying skin was stretched and had shiny appearance. Palpation revealed the presence of a mass which was smooth, rounded, cystic, opaque and fluctuant, with a well-defined margin. It measured 35×20 cms in size.

Intraorally, the sublingual swelling occupied the whole of the floor of the mouth and the tongue was elevated and pushed posteriorly (Fig. 3). The lower

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FIG. 1

Photograph of the patient described in the text showing the size (lateral view).

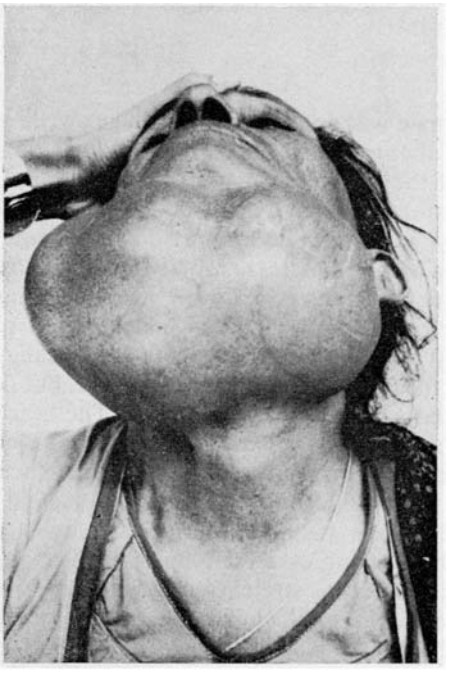


FIG. 2

Pre-operative photograph of the patient showing expansion of cyst in the neck.

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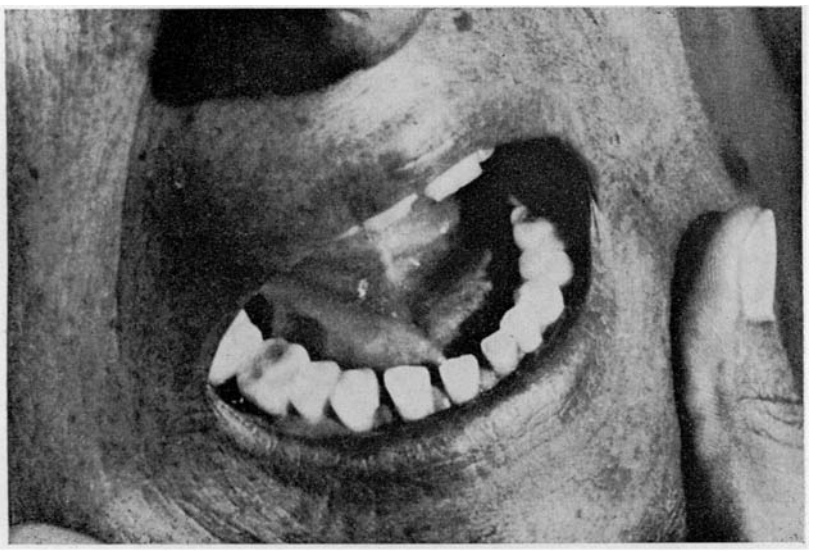


FIG. 3

Photograph of the patient: the sublingual swelling can be clearly seen.

incisors were slightly proclined. The mass was fluctuant, and the overlying mucosa was normal and freely movable over the swelling. All the teeth were present.

The blood examination showed that haemoglobin was 8 gm/per cent and the white cell count was 5600/cu mm, with 55 per cent polymorphs, 42 per cent lymphocytes and 3 per cent eosinophils. Urine analysis was normal. The results of blood urea, blood sugar, serum cholesterol and serum protein examinations were within normal limits. V.D.R.L. was non-reactive. The electrocardiogram was normal, as was the chest x-ray. The lateral soft-tissue film (Fig. 4) revealed a large, smooth, well defined, circumscribed soft-tissue shadow in front of the laryngopharynx and larynx. There was evidence of pressure atrophy of the mandible.

The cyst was aspirated; it contained grey-coloured, thick, foul-smelling fluid. Microscopic examination of the aspirated fluid showed only desquamated squamous epithelial cells. No cholesterol crystals or malignant cells were present, and bacteriological culture of the fluid was sterile.

The patient was operated upon under general anaesthesia and, despite the size of the swelling over the neck, the intubation was smooth. Anaesthesia was maintained by gas and oxygen with a relaxant. The swelling was removed through a transverse incision over the middle of the swelling. The subcutaneous tissue, platysma, investing layer of the deep facia and upper parts of the sternohyoid and thyrohyoid muscles were all atrophied and stuck together. There was no separate layer of cervical facia surrounding these structures, which were severed transversely. The cyst wall was then exposed, and it was seen to have expanded below the hyoid bone, deep to the sternhyoid muscle. Posteriorly, the sternothyroid and thyrohyoid muscles had become atrophied. Above the hyoid, the cyst expanded into the floor of the mouth between the myelohyoid muscles which were also atrophied. Postero-laterally, on either side, the cyst wall was

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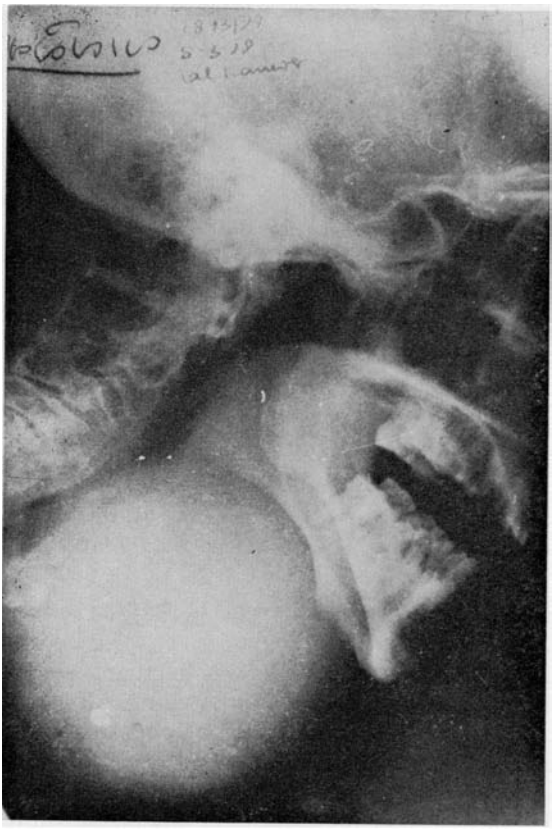


FIG. 4

Radiograph (Soft-tissue neck, lateral view) showing the extension of the sublingual dermoid cyst and pressure-atrophy of the mandible.

adherent to the carotid sheath, from which it was carefully separated. The cyst was so bulky that its separation and removal from its bed was cumbersome and so, to facilitate its dissection, it was aspirated, about 800 ml of fluid (which was ash-grey in colour and foul-smelling) were drained. Thereafter, the dissection became easy and the cyst was removed, along with its entire wall. Superiorly the cyst had caused pressure-atrophy of the lower border of the mandible, and the intraoral extension was removed with the main mass. The wound was closed in layers, and a tracheostomy was made through a separate transverse incision. During the operative procedure, a moderate amount of bleeding occurred and the patient was given 600 ml of blood by transfusion. She made an uneventful recovery and was discharged on the ninth post-operative day.

Gross examination of the specimen

The cyst contained a further 100 ml of foul-smelling, dirty, thick, ash-grey fluid. The specimen measured 25×15 cms, and it was cut open (Fig. 5). The inner surface of the cyst wall had a smooth line and contained soft, friable and greyish-white, small nodular areas at places. No hair or tooth was observed.

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Histopathological examination of the tissues showed a covering of stratified squamous epithelium (Fig. 5). There were areas of connective tissue, muscle fibres and mononuclear cell infiltration. In a few places, normal salivary gland tissue was present. A few glands were also seen which were lined by tall columnar epithelium with pseudostratification (Fig. 7). These glands seemed to be of endodermal origin. No hair cells or teeth were visible. These findings are compatible with the diagnosis of a dermoid cyst.

Discussion

Dermoids in the sublingual area are rather unusual and are amongst the clinical variants of the mid-line dermoid tumours of the neck. They may be present in any age group, and have even been reported in the newborn (Hitchin, 1970), the commonest incidence being in the second and third decades of life. There is no sex predilection (Resouly, 1976).

Sublingual dermoid cysts are developmental in origin and are believed to arise from epithelial debris or rests, during the mid-line closure of the bilateral first branchial arches and also probably the second (Taylor *et al.*, 1966; Katz, 1974; Akinosi, 1974). Yoshimura (1970) carried out a histochemical study of sublingual dermoids and showed a distribution of enzymes similar to those found in normal skin This finding supports the above theory. Colyp (1925) and Johnson (1942) support the theory that some may develop from remnants of the tuberculum impar of His which, together with the lateral processes from the inner surface of each first arch, form the body of the tongue and floor of the mouth. These cells of either remnant multiply to form a dermoid, later in life, through an unknown stimulus. Some do not present themselves until the patient is in his forties, while others have been reported in the newborn. Hitchin (1970)

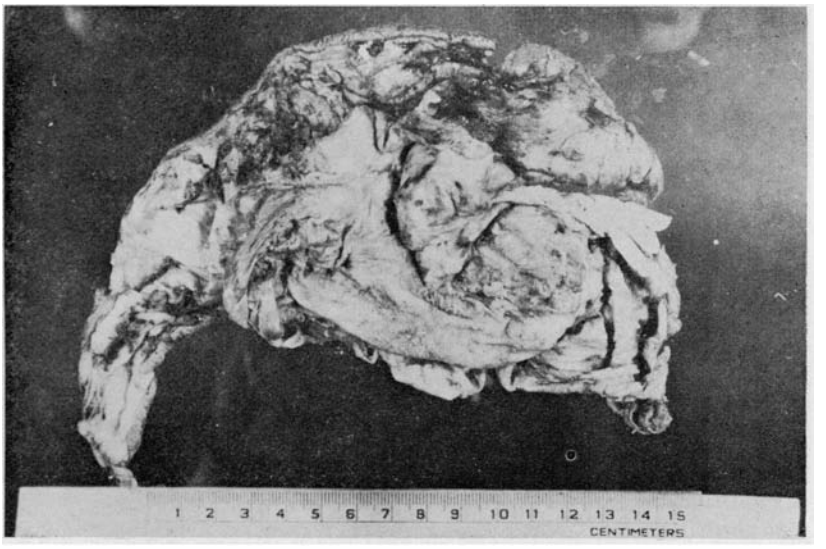


FIG. 5
The cut specimen showing the inner surface of the cyst wall.

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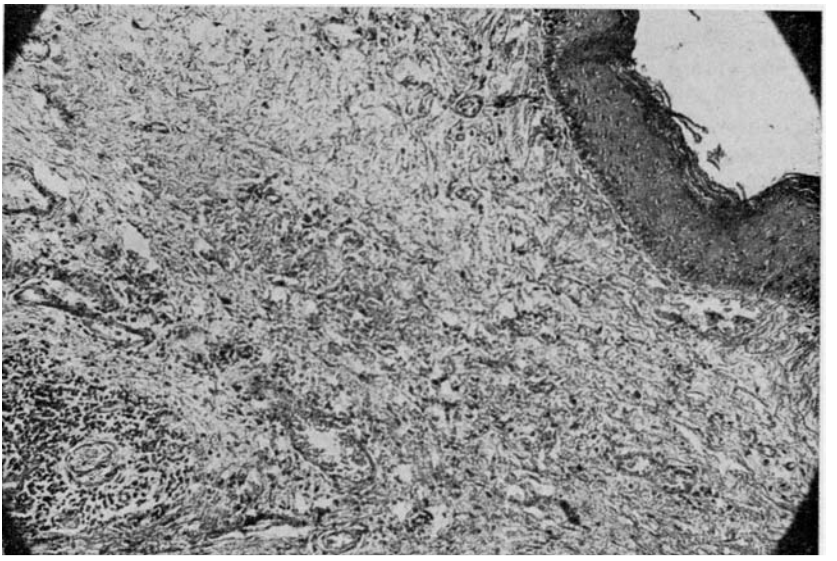


FIG. 6

Photomicrograph demonstrating stratified squamous epithelium, with areas of connective tissue, muscle fibres and mononuclear cell infiltration. At places normal salivary glands are seen (H.E. ×80).

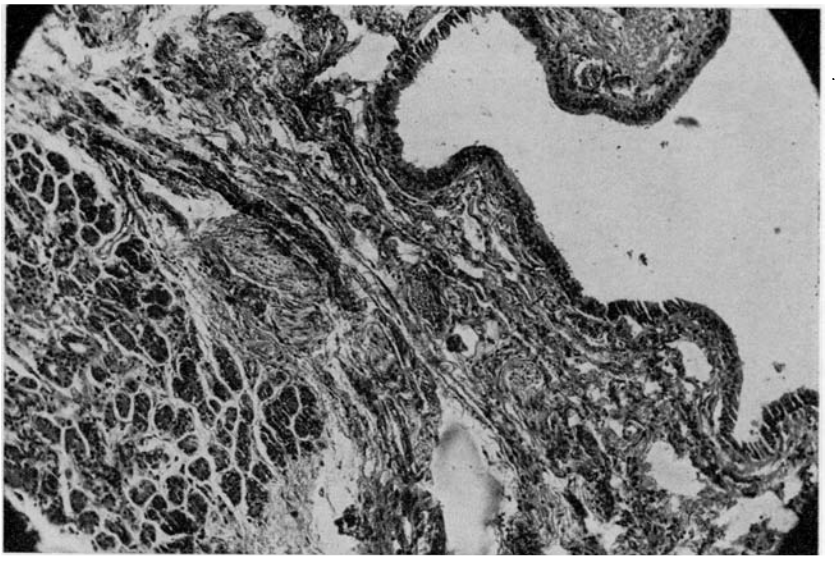


FIG. 7

Photomicrograph showing glands which are lined by tall columner epithelium with pseudo-stratification and probably of endodermal origin (H.E. ×80).

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and Seward (1965) have reviewed the embryology of the floor of the mouth in relation to both midline and laterally-situated dermoids.

A dermoid cyst may be congenital or acquired. Thus it can be sequestral (or inclusion) and post-traumatic (or implantation) in type. The congenital or sequestral cysts are present along the lines of embryonic fusion, and common sites are the post-auricular region, outer angle of the eye, scalp and mid-line of the neck. The acquired or post-traumatic cysts are due to the implantation of a few cells, or perhaps to a hematoma which has been lined by epithelium from adjacent sweat glands which the hematoma has stimulated to proliferate. The common sites are the finger, palm and sole, but they may develop in the conjunctiva and sclera; they are rarely present in the floor of the mouth. Mayer *et al.* (1965) classified dermoid cysts of the cervico-facial region into three groups, according to their histological appearances, thus:

(i) The epidermoid type, which is lined by flattened stratified epithelium and contains no other cells apart from supporting connective tissues.
(ii) The dermoid type, which has a fibrous wall containing skin structures such as hair follicles, sweat and sebaceous glands.
(iii) The teratoid type, which contains tissue developed from all three germinal layers; thus cartilage, respiratory and gastrointestinal elements are seen.

The present case belongs to the second type. No dermal element were seen in the cyst wall, but the lining epithelium had a mixed epithelial-sebaceous nature. In the present case, it is possible that dermal elements, such as hairs and teeth, were removed at the previous operation. In addition to this, Fulcher and Green (1966) stated that these cysts may or may not contain dermoid elements. They reported a case of sublingual dermoid similar to the present case.

Dermoids in the sublingual area are rather unusual. They present as a swelling in the floor of the mouth, or the patient may notice the development of a double chin. These may extend into a large cyst covering the whole of the anterior part of the neck, as in the present case.

Such huge sublingual dermoids are clinical curiosities which may cause diagnostic problems. They must be differentiated from ranula, thyroglossal cyst, cystic hygroma, subhyoid bursitis, diffuse colloidal parenchymatous multinodular goitre, epidermal cyst, sebaceous cyst, branchial cyst, salivary gland tumours, ectopic or aberrant thyroid, pharyngeal diverticulum, laryngocele, hemangioma, lymphadenopathy, lipoma, neurofibroma, and so on.

A ranula is always bluish in colour intraorally and contains a thin but slightly viscous, clear or straw-coloured fluid, whereas a sublingual dermoid is a firm, well-defined lesion, always covered intraorally with natural mucosa; it is pinkish in colour. Thyroglossal cysts are smooth, rounded, elastic, fluctuant swellings in, or almost in, the mid-line of the neck, immediately above the hyoid bone, and they move with protrusion of the tongue (Smith, 1965). Cystic hygroma is a thin-walled cyst, wh.ch trans-illuminates and often has a light blue colour when exposed at operation (McDowell *et al.*, 1963).

Subhyoid bursitis produces an oval cystic swelling immediately below the hyoid bone, and it moves with the tongue because of its attachment to the hyoid, but its long axis is always transverse (Srinivasan, 1974). Epidermal cysts are thin-walled, and their walls are attached to the skin; there is often a history of previous trauma. Thyroid swellings can assume a very large size but they always obliterate

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the area between the cricoid and the suprasternal notch, while large sublingual dermoids (like the present one) may extend inferiorly, but without obliterating the cricoid and tracheal rings. Sebaceous cysts are subcutaneous, but are always attached to the skin at some point, i.e., the punctum. Branchial cysts are invariably situated laterally and they have their own characteristic features. A laryngocele is a cystic swelling, which is reducible by pressure but reappears on Valsalva's manoeuvre. Cavernous haemangiomas are compressible and slowly refill upon release of pressure; the overlying skin is bluish in colour. Lipomas are soft and have a tendency to slip under the examining finger: the patient unusually has a long history.

A lateral soft-tissue x-ray of the neck is most important, in order to outline the size and extension of the cyst, and to detect compression over the upper air passages. If cysts are large, they may render endotracheal intubation impossible (Stewart, 1973). Forster (1968) described a technique of injecting a radio-opaque medium into the cyst, to outline its size and extension.

Treatment in these cases is usually by simple excision. If a sublingual dermoid is small, then a routine transoral approach is suggested. The mucosa of the floor of the mouth is divided in the mid-line and the tumour enucleated. Rise (1964) described such a case in which the tongue was divided to obtain good access.

Larger sublingual dermoids should be removed by an external approach, by making a curvilinear incision, with its convexity up-wards, between the lower border of the mandible and the hyoid bone. Erich (1940) pointed out that dermoid cysts are often fixed to the surrounding tissues, thus making their removal by blunt dissection impossible. Kinnman (1968) recorded that, if a dermoid is so large as to make intubation impossible, an unplanned pre-operative tracheostomy under local anaesthesia may be required. Even if intubation is possible in such a large cyst, a tracheostomy must be done post-operatively, because of the danger of subsequent laryngeal oedema.

The prognosis is usually good and recurrence is rare.

Summary

An unusual case of a massive sublingual–cervical dermoid is presented. The salient features of the condition in relation to its incidence, embryology, pathology, symptomatology and management are discussed.

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Department of E.N.T.,
P.B.M. Associated Group of Hospitals,
Bikaner, India.

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