

# CONGENITAL SUBLINGUAL DERMOID CYST IN A NEONATE: A CASE REPORT

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## SUMMARY

Dermoid cysts of the head and neck are uncommon and represent only 6.9% of all dermoid cysts occurring in the body.

Sublingual dermoid cysts are even rarer, accounting for 1.6%-1.8% of all dermoid cysts. They are generally regarded as slow-growing congenital malformations, which are rare in children and usually present in young adults, with maximum incidence in the second or third decade. Case reports in neonates are extremely rare.

A case of a large congenital sublingual dermoid cyst, in a 2-month old baby girl, which was first noticed at birth, is reported. An aspiration method was used to reduce the size of the cystic lesion to facilitate intubation during general anaesthesia and the cyst was excised entirely via a modified intraoral surgical approach. It is recommended that this aspiration method combined with the modified surgical approach be used for large sublingual dermoid cysts to avoid the external approach.

**KEYWORDS:** Congenital sublingual dermoid cyst; neonate; aspiration method; modified intraoral approach

## INTRODUCTION

Dermoid cysts are epithelial-lined cavities with variable types of skin appendages such as hair, hair follicles, sebaceous glands, and sweat glands<sup>1</sup>.

Histologically and histogenically, they are germ cell derived teratomas having an ectodermal dominance with mesodermal elements. They differ from the true teratomas, which are composed of three germinal layers<sup>2</sup>.

Dermoid cysts are most commonly located in the ovaries and the sacral region but are infrequently found in the head and neck region where the most common site is the lateral eyebrow, the so-called angular dermoid. They are rare in the oral cavity but when they do occur, they usually present as a swelling in the floor of mouth where they may be

found in sublingual, submental, or laterally in the gutter between the hyoglossus and mylohyoid muscles.

Butlin<sup>3</sup>, in 1885, first reported a congenital dermoid cyst of the floor of mouth. New & Erich<sup>4</sup>, reviewing 1495 cases of dermoids in 1937, found that, of the 103 (6.9%) cases involving the head and neck region, only 24(1.6%) involved the floor of the mouth. Similarly, a more recent retrospective survey of all available literature on dermoids showed a sublingual incidence of 1.8%<sup>5</sup>.

This paper is a report of a case of an unusually large midline sublingual dermoid cyst in a 2-month old baby girl, which was initially observed at birth. The method of intubation and the surgical approach to the lesion, are described.

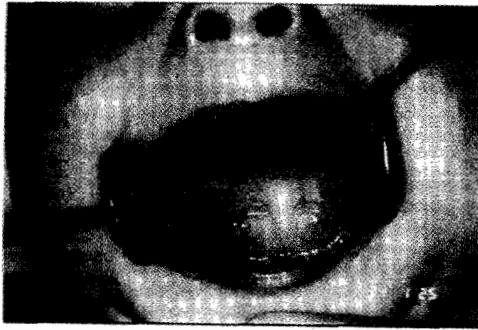
## CASE REPORT

A 2-month old girl was referred to the Department of Oral and Maxillofacial Surgery, King Fahd Central Hospital, Gizan, by the Paediatric Surgeon, in February 1994, with a large sublingual swelling, which had been present since birth and was getting bigger. The lesion had raised the tongue onto the palate thus causing difficulties with feeding and breathing.

The mother had a normal pregnancy and the baby was delivered at full term with no complications.

On examination the baby looked healthy and had no other abnormality. Intra-oral examination revealed a large midline sublingual swelling elevating the tongue into the palate and preventing patient from closing the mouth. She was breathing with great difficulty.

The swelling was fluctuant and non-tender, and the overlying mucosa had the same colour as the rest of the oral mucosa. It did not produce a submental bulge and there was no associated submandibular lymphadenopathy. Figure 1



**Figure 1** Clinical picture of the swelling

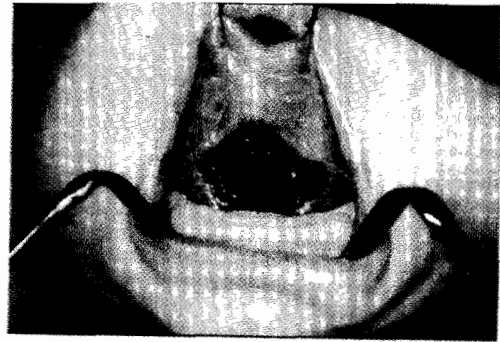
CT Scan showed a large non-enhancing cystic lesion occupying most of the substance of the tongue above the mylohyoid muscle (Figure 2).



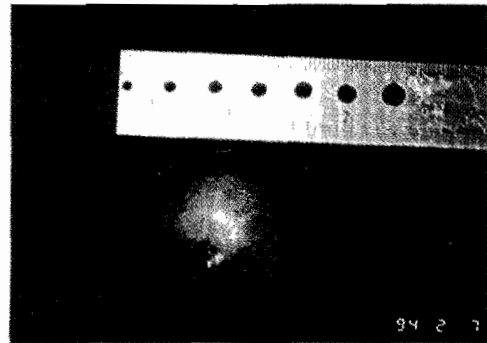
**Figure 2** CT Scan of the sublingual dermoid

Chest x-ray was normal. Aspiration biopsy produced only a cloudy white fluid with no trace of blood. Microscopic examination of the specimen was reported as containing lymphocytes and granular material. Haematological results were within normal range.

At surgery, the swelling was so large that about 20ml of the fluid had to be aspirated from the cyst before laryngoscopy and nasotracheal intubation could be done. Traction sutures were placed on either side of the tip of the tongue and pulled forward and upwards. A sagittal incision was made along the lingual frenum to the lingual sulcus, making an inverted 'T' junction with a horizontal incision below and parallel to the sub-mandibular ducts. This provided good access and allowed the cyst, measuring about 3cm in diameter, to be excised by blunt and sharp dissection without difficulty. The wound was closed in layers without the need for vacuum drainage. Care was taken to avoid ligation of the sub-mandibular duct during the suturing of the horizontal incision in the lingual sulcus (Figure 3a and 3b).

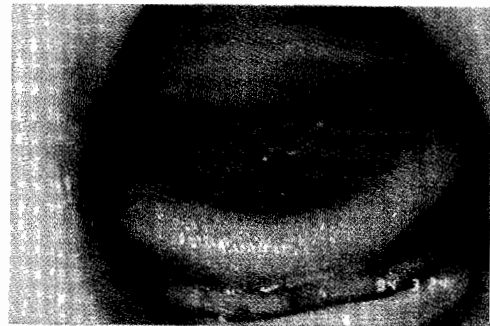


**Figure 3a** Surgical defect following excision



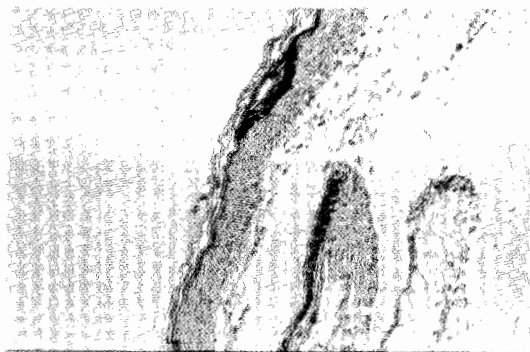
**Figure 3b** Excised specimen of sublingual dermoid

The patient started feeding after 12 hours. She was discharged on the 10<sup>th</sup> post-operative day. Post-operative healing was uneventful and the final result was excellent (Figure 4).



**Figure 4** Post operative picture of the tongue

The cyst wall was thick and the fluid content was made up of thick pultaceous material. Microscopically, the cyst was lined by stratified squamous epithelium with keratin layer. The wall was made up of fibrous tissue, sebaceous glands and hair follicles. The final diagnosis of sublingual dermoid cyst was made (Figure 5).



**Figure 5** Photomicrograph of the lesion H & E stain.

## DISCUSSION

Although dermoid cysts of the floor of mouth have been reported to present at any age from birth to 72 years, they are rare in children because they usually grow slowly and become apparent between the ages of 15 years to 35 years<sup>6</sup>. Case reports in neonates are extremely rare<sup>7,8</sup>. There is no sex predilection.

Dermoid cysts are usually asymptomatic until they become large enough to interfere with speech, deglutition or rarely respiration<sup>1</sup>. If left alone the large cyst can cause an increase of the size of the lower dental arch and cause proclination of the lower incisors in dentate patients<sup>9</sup> or sometimes cause obstructive submandibular sialadenitis<sup>10</sup>.

Sublingual dermoid cysts may occasionally present in the neonatal period and grow rapidly as reported by Bloom et al<sup>7</sup> as well as the present case, which was noticed at birth and reached a large size in 2 months. Tuz et al<sup>11</sup>, in 2003, reported a case of sublingual dermoid, which grew rapidly in a pregnant female resulting in deglutition and respiratory problems. They concluded that pregnancy accelerates the growth of sublingual dermoid cysts and recommended excision of such lesions before pregnancy.

Sublingual dermoid cysts may be found in association with thyroglossal duct cyst in the same patient<sup>8</sup>. This is, of course, a chance occurrence as there is no embryologic link between the two lesions.

The differential diagnosis of dermoid cysts of floor of mouth include ranula, thyroglossal duct cyst, cystic hygroma, sialolithiasis or infection of the sublingual and submandibular glands, acute infection or cellulitis of the floor of mouth, benign

Tumours (e.g. lipoma, fibroma, haemangioma), malignant tumours and normal fat mass in the submental region. Diagnostic aids include needle aspiration, ultrasound, CT scan, MRI and x-ray with injected contrast medium.

Treatment is by complete surgical excision. Cysts of the floor of mouth can be excised by intraoral approach, or Extraoral approach or a combination of both depending on site, size and extent of spread of the lesion.

When the cyst is small and cervical extension is negligible intra-oral approach alone is likely to be adequate. Those, which extend below the mylohyoid muscle, can be excised through a cervical approach. Where there is extension into the neck especially when the cyst has penetrated the mylohyoid muscle then the intra-oral approach alone is unlikely to be adequate. A combination of intra-oral and cervical approaches where complete removal is facilitated by direct visualization of important adjacent structures is preferable<sup>5,12</sup>.

An alternative to this combined approach has been recommended by Di Francesco et al<sup>13</sup> who avoided additional skin incision by aspirating the content of the cyst before excising it through the intraoral approach.

McGregor<sup>14</sup> noted that since some cysts are not truly sublingual but positioned in the midline almost directly behind the symphysis of the mandible, obscured by standing teeth, a combination of intra-oral and extra-oral approach might not be adequate. He therefore suggested that the most logical approach is to osteotomize the symphysis of mandible via a modified lip-splitting technique because the dermoid cysts always prove to be longer than the clinical examination would have suggested. This mode of treatment seems rather radical for such a benign lesion and should have a very limited application.

Some dermoid cysts are closely attached to the hyoid bone and such lesions should be excised using Sistrunk procedure<sup>8</sup>. This procedure involves excision of the dermoid cyst and the part of the hyoid bone to which it is attached.

The surgical procedure described in this report was facilitated in the patient when aspiration of the cystic content was done to reduce its size to facilitate intubation for general anaesthesia. The reduction of the size by aspiration also made it easier to excise the large cysts entirely via the intra-oral

route using an "inverted T" modification of the midline glossotomy approach.

Due to the fact that the transverse diameter of the lesion was far greater than the vertical diameter, making the distance from the tip of the tongue to the lingual sulcus very short, a modification of the midline sagittal glossotomy approach was used. The "inverted T" incision increased the access and allowed the cyst to be removed without undue tension on the submandibular ducts.

The precautions to take with this combined approach include avoiding aspirating the whole cystic content as this will cause total collapse of the lesion and make its identification during surgery difficult; ensuring that the horizontal incision is placed in the lingual sulcus well below the papillae of the submandibular ducts to avoid trauma to the ducts or the lingual nerves during the incision; and avoidance of accidental ligation of the submandibular ducts during suturing.

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