A CASE REPORT ON PALATAL DERMOID WITH BIFID TONGUE

Md Akbar Husain Bhuiyan [1] Md Ashraf ul Huq Kazal [2] Shahanara Chowdhury [3] Shafquat Haider Chowdhury [4]

Abstract

Congenital tumors of the oral cavity are extremely uncommon. The teratoid tumors (epiganthi, dermoid and hairy polyps) account for almost all of the reported cases. They might be associated with cleft of the soft palate and other midline defect yet its association with bifid tongue have not yet been reported. We present here a baby of 2 days old with dermoid over hard palate associated with bifid tongue, first of its kind as far as our reach. The review of the literature, the description of the lesion, the diagnosis, and the management of this finding are outlined.

Kew Words: dermoid; hard palate; bifid tongae

Introduction

During the development of head and neck as the process of fusion completes, a small piece of epidermis may get entrapped deep in the mid line, just behind the fusion of hard palate and later forms a palatal dermoid cyst. (1) The term dermoid cyst clinically refers to describe three different lesions and includes epidermoid cyst, dermoid cyst and teratoid cyst. Though considered to be developmental anomaly, these cysts can also occur following some surgical procedure causing minor trauma to the oral cavity in early period of life. (2) These cysts are rarely reported but still should be differentially diagnosed from common pathologies lipoma and leiomyoma. The occurrence of dermoid just over the bifid tongue makes its pathology crystal clear (pressure over developing tongue bud) and may come under certain types of midline craniofacial defect, but literature review failed to find any similar case being reported. There are a few reports on associated lesion of dermoid with cleft palate but not bifid tongue. Bifid tongue by itself has been reported with babies of diabetic mother which is not the case in our case. Infants born to diabetic mothers (IDDM) are well documented to have a higher rate of congenital malformations. Sacral agenesis/hypogenesis and caudal dysgenesis are classically linked to maternal diabetes, but many other types of anomalies are more frequent. In this case report, we rather describe a male infant born to a nondiabetic mother who in addition to other typical congenital abnormalities was born with an impressive bifid tongue. Review of the literature reveals an additional case of an infant with a bifid tongue born to a diabetic mother (Comess et al, 1969). In conclusion, bifid tongue with oral dermoid, a rare congenital anomaly, can be an associated finding of midline defect.

1. Assistant Professor of Pediatric Surgery Chittagong Medical College, Chittagong
2. Associate Professor of Pediatric Surgery Chittagong Medical College, Chittagong
3. Associate Professor of Gynae & Obstetrics Chittagong Medical College, Chittagong
4. Post graduate student of MS (Pediatric Surgery) Part I Chittagong Medical College, Chittagong

Correspondence : Dr Md Akbar Husain Bhuiyan

Clinical case

we present here a first sibling of couple named Mr. Nasir & Mrs. Shirin who were known to be healthy without any family history of hereditary disease. He was referred to our ward from neonatal ward following normal uneventful delivery on 21.12.2008 with normal birth weigh, no antecedent antenatal problem and without any family history as he developed difficulty in breathing associated with bifid tongue and palpable mass on hard palate. We compiled case history, and physical examination reveled bluish mass of approximately 3cm in the midline of hard palate with bifid tongue (snake tongue). No other abnormalities were detected and it was diagnosed as palatal dermoid with bifid tongue. Excisional biopsy of dermoid and reconstruction of bifid tongue was planned and operated on 5th day after completing base line investigation. At the time of surgery, endotracheal intubation was used with no reported difficulty. Examination of the cleft and palate revealed a moderate size mass on the hard palate with bifid tongue. The mass just overlies the fissure implicating its pathogenesis as. Adenoids looked normal. No bony gap in the basal bony structure of the nasopharynx was detected. At this point, the surgical repair was attempted on 23.12.2008. Excisional biopsy of the mass and repair of bifid tongue using Vicryl 5/0 with utmost importance in saving functional integrity of tongue was done and mass sent for histopathological examination. Histopathological study of the specimens demonstrated a stratified, squamous, keratinizing epithelium, hair follicles, sweat glands and sebaceous glands typical of epithelial dermoid

with no evidence of malignancy. These findings were consistent with our clinical diagnosis of dermoid cyst. The post operative perods was unevenful though there was a minor stitch line infection of the tongue which was removed and reconstruction in a later life planned and parents consoled. The patient was discharged on 02.01.2009 with an advice to come back for follow up after 15days. On follow up proper clinical examination of the repaired tongue with the oral cavity was done and no abnormality was detected. Differential diagnosis of the lesions included leiomyoma, lipoma, angioma. associated with bifid tongue.

Discussion

Tumors of the newborn oral cavity are very uncommon (Cohen, 1978). Because teratoid tumors account for most of the reported cases associated with a cleft palate, the number of lipomas associated with cleft palates must be low. In fact, a literature search failed to find any reported case with a similar finding. Congenital lipomas of the fornix of the vestibule (De Carvalho et al, 1987) and of the tongue (Dimitrakopoulos et al, 1990) were reported in a 7-month-old boy and a 20-day-old girl respectively. Neither case was associated with a cleft deformity of the lip or palate. Congenital hairy polyps of the nasopharynx in association with cleft palate were diagnosed in two newborns (Haddad et al, 1990). In 1991 Lowry and Yon published a report of two brothers, both born with a cleft palate and sacral lipoma. A lipoma of the corpus callosum with a median cleft of the upper lip and cutaneous facial polyps was also documented (Rudnik-Schoneborn and Zerres, 1994).

The position of the dermoid overlying the bifid tongue proves beyond any doubt that the dermoid is the cause for bifid tongue.

Fig 1 : Preoperative picture of dermoid with bifid tongue

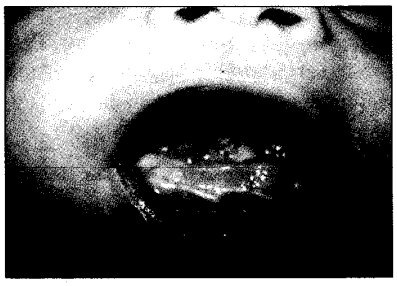

Fig 2 : Postoperative picture after removal of dermoid and repair of bifid tongue

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