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CASE REPORT

SUCCESSFUL MANAGEMENT OF INTRAORAL EPIDERMAL CYST IN A NEWBORN: A RARE CASE REPORT

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ABSTRACT: Epidermal cysts involving head and neck are uncommon representing around 7% of all epidermal cysts in the body. Sublingual cysts are still uncommon (1.5-2%) usually presenting in young adults in 2nd or 3rd decade of life. An epidermal cyst presenting at birth in sublingual position is extremely rare. A large sublingual epidermal cyst in a newborn baby girl, which is causing breathing and swallowing difficulties leading to hypoxia in supine position and continuous drooling of saliva with inability to close the mouth, is reported. Successful excision of the cyst without rupture by transoral route under general anesthesia is performed at 24 hours of life.

KEYWORDS: Neonate, Sublingual Epidermal Cyst, Transoral approach, Difficult intubation, Sharing of airway.

INTRODUCTION: Epidermal and dermoid cysts are benign lesions in the body. These cysts are defined as epidermoid when the cyst lining presents only epithelium, dermoid when skin adnexa are present and teratoid when other tissues like muscle, cartilage or bone are present.[1,2,3]

These cysts can occur anywhere in the body, with incidence of about 1.6% to 6.9% around head and neck area and 1.6% in oral cavity.[4] They are very rare in oral cavity and represent less than 0.01% of all intraoral cysts like ranula, lymphatic malformation, dermoid/teratoid cysts, heterotopic gastrointestinal cyst and duplication foregut cysts.[2,5]

CASE REPORT: A Newborn female neonate born after caesarean section was referred to neonatal intensive care unit of Basant Sahney Pediatric hospital, Secunderabad immediately after birth with a large intraoral, sublingual swelling of about 4.0cmx3.5 cms size. This swelling is pushing the tongue on to the palate, with inability to close the mouth causing continuous drooling of saliva. Baby was becoming hypoxic in supine position, as the sublingual mass was causing fallback of elevated and pushed tongue. Hence, the neonate was nursed in prone or lateral positions to prevent hypoxia. A nasogastric infant feeding tube was placed in the stomach to institute gavage feeds and to rule out Oesophageal atresia.

On examination, baby was healthy, pink with a birth weight of 2.2kgs. Antenatal scans did not reveal any foetal anomaly. Intraoral examination revealed a midline, large, cystic sublingual mass which was fluctuant and nontender (Fig. 1). The mucosa over the swelling appeared normal, stretched out without any signs of inflammation. Submental bulge noticed which was bimanually palpable. There were no associated cervical lymph nodes.

Ultrasound examination with colour Doppler study revealed a cystic mass without any vascular malformations or abnormal blood vessels. Haematological reports were within normal range and chest radiograph was normal.

As the neonate was not able to maintain O2 saturation levels satisfactorily, emergency surgical excision under G. A. was performed at 24 hours of life. A multichannel monitor with ECG,

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NIBP, SpO2, HR was connected. The baby was premedicated with 0.1mg Atropine & 0.25mg Ondansetron. Intubation was done under Inhalation anesthesia. (Fig. 2). 100% oxygen and Sevoflurane 0-6% was administered gradually and intubation was attempted with straight blade Laryngoscope, but intubation could not be done in the 1st attempt, and as the child was recovering from the anesthesia, again, the depth of anesthesia was increased with O2 and Sevoflurane 0-6%. With finger pressure on the trachea, intubation could be done with 3mm Portex tube & throat packed with gauze and ETT fixed at 9cm. After confirming the tube placement, 2mcg Fentanyl and 1.5mg Atracurium were given. Anesthesia was maintained with O2/N2O/0.8% Sevoflurane. 10% Dextrose 8 ml per hour IV drip as maintenance fluid was given.

Baby was placed in supine position with a bolster under the chest and a roll under the neck for support. A traction stitch was placed at the tip of the tongue which was pulled upwards and forwards. A curved Chevron incision was made over the stretched out sublingual mucosa, above the submandibular ducts, exposing the cyst. A good access was achieved and the entire cyst was excised in toto, by gentle dissection using needle point diathermy (Fig. 3). Good care was taken to avoid injury to Whartons submandibular ducts. There was no blood loss during the procedure and the sublingual mucosa was reconstructed under the tongue achieving normal shape (Fig. 4, 5). Vitals were stable throughout the procedure and reversal was done with 0.15mg Neostigmine and 0.1mg Atropine. Throat pack was removed and after suctioning of the oral cavity, extubation was done. The baby was comfortable and active. Vitals and saturation were normal and the baby was shifted to NICU for further management.

Postoperative period was uneventful. Gavage feeds were started 5 hours postoperatively and normal breast feeds 12 hours afterwards. Baby had normal recovery and was discharged from the hospital after 5 days. The baby was reviewed after 3 and 6 months showing excellent healing. No evidence of any recurrence was found.

Pathology report revealed a cyst with a lining of stratified squamous epithelium with granular layer with luminal content of laminated keratin material suggestive of an epidermal cyst.

DISCUSSION: Congenital cysts are dysembryogenetic lesions that can occur due to ectodermal elements getting entrapped during midline fusion of mesodermal elements of first and second branchial arches, between 3rd and 4th weeks of intrauterine life.[6] Common age of presentation is in young adults in the second or third decade of life due to the slowly progressive growth of the lesion.[7] The differential diagnosis includes ranula, lymphangioma, teratoma and enteric duplication cysts.[2,5]

Large intraoral cysts in the newborn can cause airway obstruction and swallowing difficulties.[7] In this patient airway management was achieved by nursing the baby in lateral and prone positions. Dysphagia occurred due to the inability to close the mouth causing continuous drooling of saliva, which led to a suspicion of Oesophageal atresia, which was promptly ruled out by passing a nasogastric tube through which gavage feeds were instituted.

Preoperative evaluation include clinical examination, transillumination, high resolution ultrasonogram including Color Doppler studies, and C.T Scan or M.R.I. Clinical examination with palpation and transillumination revealed the cystic nature of the swelling, and ultrasound examination confirmed these findings. Color Doppler study ruled out the possibility of a vascular malformation. C.T. Scan or M.R.I reveal more precise anatomical details, but these investigations were not planned in this neonate due to the high risk of hypoxia in supine position under sedation.

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Large intraoral swellings in newborn is challenging for both the Surgeon and the Anesthesiologist. In this patient initial plan is to intubate with Sevoflurane anesthesia. The plan 'B' is to aspirate the swelling partially or totally so as to facilitate intubation.[7] A plan 'c' is also kept ready, that is to perform an emergency tracheostomy. An ENT surgeons team is informed and kept ready as backup. Intubation under Sevoflurane anesthesia is the first choice as this procedure does not disturb the local anatomical planes at all, and this swelling can be excised in toto. Aspiration of the swelling alters the anatomy and tracheostomy adds to the morbidity.

In most of the published case reports the surgical excision was performed at 2 months, at 8 months and at 2 years respectively.[7,8,9] In these babies aspiration of the cyst followed by nasotracheal intubation was performed, whereas in our case report cyst aspiration was avoided, orotracheal intubation was achieved and surgical excision was performed at 24 hours of life.

Surgical excision is very effective and curative. Prognosis is very good with a very low incidence of relapse. Planning the surgical excision and achieving it requires a well considered team approach with the involvement of Radiologists, Neonatologists, Anaesthesiologists and Pediatric Surgeons.

CONCLUSION: These swellings are very rare in neonates and if not treated expeditiously in a tertiary neonatal care centre, they may cause upper airway or GIT impairment and can potentially be fatal.

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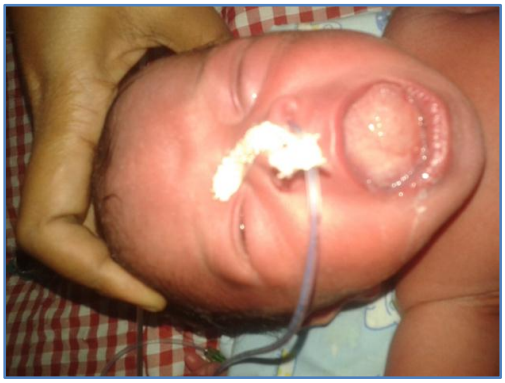

Fig. 1: A large intraoral swelling with drooling of saliva in a neonate

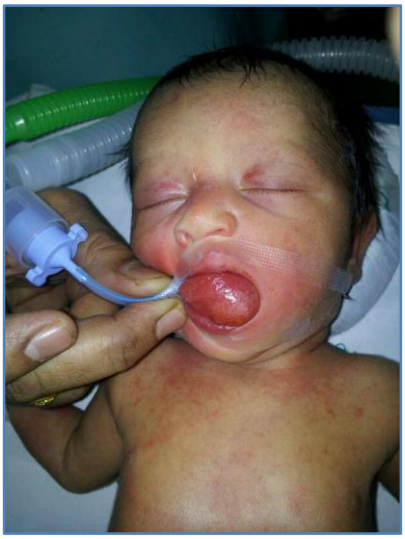

Fig. 2: Orotracheal intubation in place

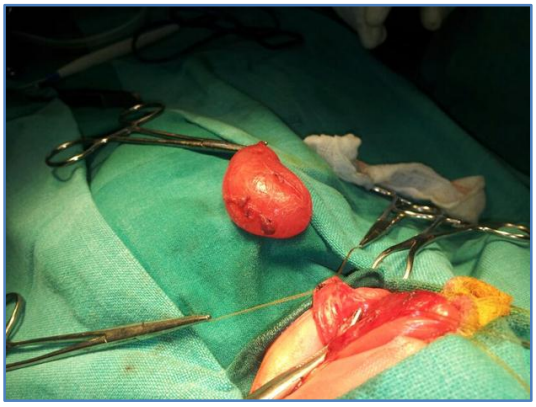

Fig. 3: Surgical excision in toto with intact specimen of excised cyst

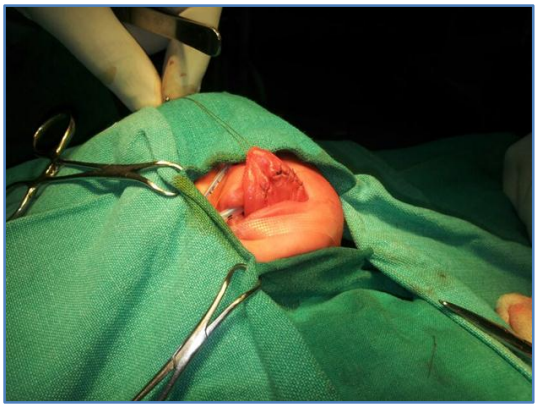

Fig. 4: Reconstructed tongue showing closure of Chevron incision

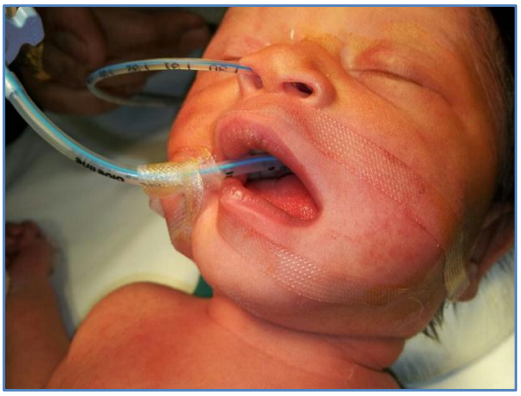

Fig.5: Post-surgical picture showing normal oral cavity

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