Mature teratoma in a neonate presenting as an intraoral cystic lesion: A case report

Srijan Singh

From Senior Registrar, Department of Neonatology, Seth G S Medical College and KEM Hospital, Mumbai, Maharashtra, India

Correspondence to: Srijan Singh, Department of neonatology, Seth G S Medical College and KEM Hospital, Parel, Mumbai, Maharashtra, India. E-mail: srijanstar89@gmail.com

Received - 22 April 2020 Initial Review - 11 May 2020 Accepted - 08 June 2020

ABSTRACT

Teratomas are germ cell neoplasms that can occur in the cervical and craniofacial regions. A male neonate, antenatally diagnosed with intraoral cystic lesion, presented with a large 5 cm × 5 cm cystic swelling at birth arising from the palate. Alpha-fetoprotein levels of the patient were >1000 IU/L and beta-human chorionic gonadotropin levels were <0.9 mIU/ml. Computed tomography neck showed a cystic hypodense lesion of palatal origin measuring 4.9 cm × 4.3 cm × 4.6 cm. The baby developed respiratory distress at around 10 min of life and underwent coablation-assisted cyst excision of the lesion. Histopathology revealed mature cystic teratoma. Postoperatively, the baby was mechanically ventilated for 2 days and subsequently weaned off respiratory support.

Key words: *Cystic lesion, Mature teratoma, Neonate, Oropharyngeal*

The word "teratoma" has been derived from "teraton," a Greek word meaning "a monster" [1]. The incidence of teratomas has been estimated to be 1:4000 births [2]. Teratomas are germ cell neoplasms that can occur in the cervical and craniofacial regions.

They consist of all three embryonic germ layers. Teratomas affecting the head and neck account for 2–5% of all germ cell neoplasms [2-4]. Teratomas in utero can lead to pulmonary hypoplasia, feeding issues, along with respiratory distress at birth caused by obstruction of the aerodigestive tract [3]. The resultant respiratory distress may necessitate the need for an ex-utero intrapartum treatment (EXIT) procedure. Obstruction is overcome through intubation or tracheostomy with complete surgical excision later.

On histology, teratomas are classified as immature and mature. Mature teratomas commonly affect the pediatric population and consist of mature skin, hair, fat, tissue, cartilage, bone, and glands. Immature teratomas contain immature elements. There is a 5% risk of malignancy with increasing age at resection. Surgical resection is often difficult because teratomas frequently infiltrate adjacent tissues. The surgical risk of mortality may be as high as 15% [4].

CASE REPORT

A male neonate was born to a 30-year-old mother at a government hospital. She had developed acute viral hepatitis with disseminated intravascular coagulation in her previous pregnancy and delivered a stillbirth child. Antenatal scan at 20 weeks showed cystic lesion along the inferior surface of palate suggestive of the nasopalatine cyst, epignathus, or evolving cystic teratoma. The scan at 35 weeks suggested an enlargement of the lesion to 4.6 cm × 4 cm with right renal hydronephrosis and hydroureter. There was no history of similar swellings in other family members.

The baby was born at 37 weeks gestation by vaginal delivery with a birth weight of 3.102 kg. A large 5 cm ×5 cm pink-colored well-circumscribed cystic swelling was arising from the palate. The swelling was non-tender and non-pulsatile (Fig. 1).

Alpha-fetoprotein (AFP) levels of the patient were >1000 IU/L and beta human chorionic gonadotropin (beta HCG) levels were <0.9 mIU/ml. The histopathology of the mass revealed cystic lesion lined by stratified squamous epithelium. The wall of the cyst showed mature neuroglia tissue, adipose tissue, skeletal muscle, blood vessels, nerve bundles, few tiny cystic spaces lined by cuboidal epithelium, and goblet cells were seen. No immature tissues were seen. Histopathology was, therefore, suggestive of mature cystic teratoma.

Computed tomography neck showed cystic hypodense lesion measuring 4.9 cm × 4.3 cm × 4.6 cm within the oral cavity in its anterior aspect with near-complete bony cleft palate (Fig. 2). No fat or calcifications were seen within the cyst. Luminal compromise of the oropharynx was seen by mass effect in the form of effacement of sulcal spaces, ipsilateral lateral ventricle, and midline shift of the posteriorly displaced tongue. The findings were suggestive of intraoral cystic lesion of palatal origin with mass effect with complete cleft palate.

The baby developed respiratory distress at around 10 min of life and underwent coablation-assisted palatal cyst excision on day one of life (Fig. 3). An incomplete cleft palate was seen. Around 45 cc serous fluid was aspirated from the cyst. The

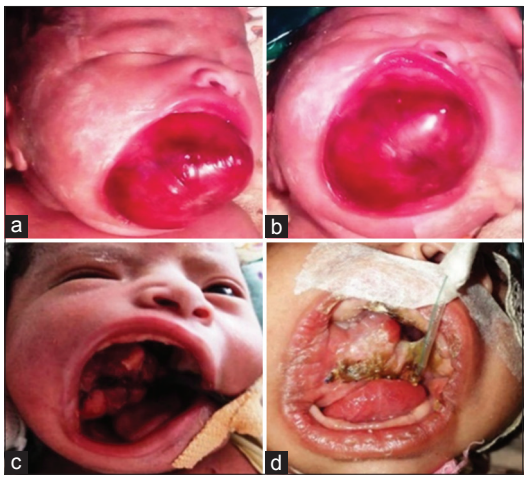

Figure 1: (a) and (b) Presentation of the child soon after birth. (c) and (d) After surgical resection

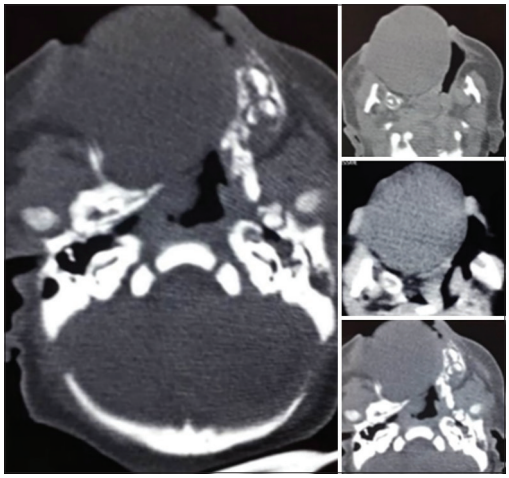

Figure 2: Computed tomography neck showed cystic hypodense lesion measuring 4.9 cm ×4.3 cm × 4.6 cm within the oral cavity in its anterior aspect

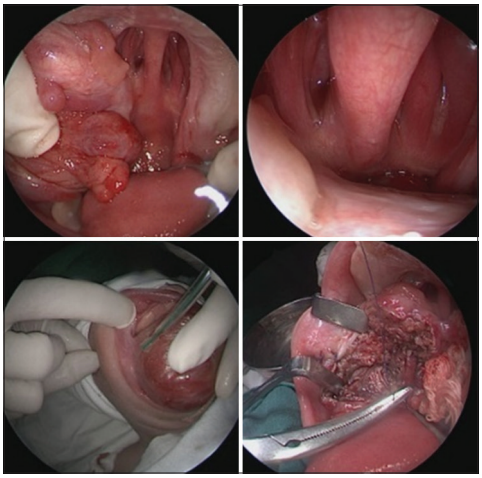

Figure 3: Video laryngoscopic images of the lesion during resection

cavity of the cyst was obliterated and closed with purse string sutures.

Postoperatively, the baby was mechanically ventilated for 2 days and subsequently weaned off respiratory support. On day 9 of life, the baby developed aspiration pneumonia requiring mechanical ventilation for 6 days. Simultaneously, the baby was diagnosed to have late-onset sepsis and meningitis and required antibiotics for the same. The baby was weaned off respiratory support on day 15 of life and started on orogastric tube feeds. There was pooling of secretions from the oral lesion which required frequent suctioning. At present, the baby is 68 days old, and complete excision of the lesion has been done at the maxillofacial surgery department of another hospital. Postoperatively, the baby is doing well on follow-up. The child can tolerate oral feedings without complaints concerning respiration or feeding and has started to gain weight.

DISCUSSION

Teratomas affecting the palate, termed as epignathus, are the most common craniofacial teratoma in newborns. They are attached to the palate through a stalk and, once resected, imaging is paramount to determine the persistence of disease at the skull base and parapharyngeal areas. They are variable in consistency, with both cystic and solid areas, and are also made up of cartilage, bone, and pigmented areas. The primary initial goal is to secure the airway and provide respiratory support. Early surgical excision, when feasible, can aid in the prevention of infectious sequelae, coagulopathies, and bleeding.

Carvalho *et al*. reported the case of a giant epignathus teratoma discovered at birth which was successfully managed and followed up for 7 years. Surgery was performed at 14 days of age, but due to residual tumor and high AFP levels, the patient was submitted to chemotherapy, resulting in complete regression of the lesion, and normalization of AFP levels. The surgical repair of cleft palate was performed at 5 years of age. At 7 years of age, the patient was in good general health and showed no clinical signs of recurrence [5]. In our case, the teratoma was diagnosed antenatally and excised at birth.

Bahgat *et al*. reported a 2-month-old girl who presented with repeated vomiting, failure to thrive and recurrent bouts of stridor, and cyanosis since birth. On examination, there was a mass originating from the oropharynx. The mass was excised under general anesthesia by CO_2 laser. A pathological diagnosis of mature solid teratoma of the oropharynx was made. Stridor and cyanosis attacks were no longer observed, and the child was able to tolerate oral feedings. After 6 months, she was doing well [6]. In contrast, teratoma, in our case, was a cystic lesion.

Brodsky *et al*. described a retrospective case series of 14 patients with teratoma of the head and neck in the neonatal population. There were seven patients who were diagnosed prenatally, while the remaining seven patients were diagnosed at birth or shortly afterwards. The tumor emanated from the neck in nine patients, the nasopharynx/oropharynx in three patients, the external nose in one patient, and the face in one patient. There were nine patients with associated upper airway obstruction. An EXIT procedure was performed in four patients, with three requiring intubation and one requiring tracheostomy. All patients underwent surgical

resection. There was one patient who demonstrated recurrence at follow-up [7].

Mc-Kenney *et al*. reported detailed histologic studies of 22 congenital teratomas. Among 15 survivors with follow-up, five patients had malignant mixed germ cell tumors, and five had immature teratomas. No patient developed recurrent or metastatic disease after treatment by complete surgical excision alone (mean follow-up 37.6 months). They concluded that the clinical behavior of congenital teratomas is determined predominantly by whether the tumor can be resected completely [8].

Elmasalme *et al*. reported nine cases of neonatal cervical teratoma, of which one patient died before surgical intervention. All others underwent resection. There was one intraoperative death and one post-operative death. The remaining six patients did well postoperatively with no significant sequelae with 3–14 years of follow-up [9].

In our case, the patient was a neonate, and oropharyngeal teratoma is unusual in this age group and at this site. The prognosis of neonatal teratoma is favorable with an 80–100% survival reported after surgical excision of the tumor and treatment of any recurrence. Gundry *et al*. reported 80% mortality in cervical teratomas which was brought down to 15% by prompt surgical intervention [10].

Tumor recurrence is high if resection is incomplete, although complete resection with clear margins may be challenging due to the involvement of critical structures. Tumor recurrence may be monitored by AFP levels, although it is unclear how long these patients should be followed clinically and radiographically [1,3,4].

CONCLUSION

Germ cell tumors of the head and neck presenting in the neonatal period are typically benign teratomas. Most fetal teratomas can be detected on routine prenatal ultrasound. The primary treatment for teratoma of the head and neck is surgical resection. Recurrence is rare and long-term prognosis is excellent. A multidisciplinary team approach is paramount to the successful management of teratoma.

REFERENCES

1. Lakhoo K. Neonatal teratomas. Early Hum Dev 2010;86:643-7.
2. Coppit GL 3rd, Perkins JA, Manning SC. Nasopharyngeal teratomas and dermoids: A review of the literature and case series. Int J Pediatr Otorhinolaryngol 2000;52:219-27.
3. Rosenfeld CR, Coln CD, Duenhoelter JH. Fetal cervical teratoma as a cause of polyhydramnios. Pediatrics 1979;64:176-9.
4. Paradis J, Koltai PJ. Pediatric teratoma and dermoid cysts. Otolaryngol Clin North Am 2015;48:121-36.
5. Carvalho CH, Nonaka CF, Elias CT, Matheus RD, Dias RM, Souza LB, *et al*. Giant epignathus teratoma discovered at birth: A case report and 7-year follow-up. Braz Dent J 2017;28:256-61.
6. Bahgat M, Bahgat Y, Bahgat A. Oropharyngeal teratoma, a rare cause of airway obstruction in neonates. BMJ Case Rep 2012;2012:859104.
7. Brodsky JR, Irace AL, Didas A, Watters K, Estroff JA, Barnewolt CE, *et al*. Teratoma of the neonatal head and neck: A 41-year experience. Int J Pediatr Otorhinolaryngol 2017;97:66-71.
8. Heerema-McKenney A, Harrison MR, Bratton B, Farrell J, Zaloudek C. Congenital teratoma: A clinicopathologic study of 22 fetal and neonatal tumors. Am J Surg Pathol 2005;29:29-38.
9. Elmasalme F, Giacomantonio M, Clarke KD, Othman E, Matbouli S. Congenital cervical teratoma in neonates case report and review. Eur J Pediatr Surg 2000;10:252-7.
10. Gundry SR, Wesley JR, Klein MD, Barr M, Coran AG. Cervical teratomas in the newborn. J Pediatr Surg 1983;18:382-6.

Funding: None; Conflicts of Interest: None Stated.

How to cite this article: Singh S. Mature teratoma in a neonate presenting as an intraoral cystic lesion: A case report. Indian J Child Health. 2020; 7(6):274-276.

Doi: 10.32677/IJCH.2020.v07.i06.008