

GIANT ORAL TERATOMA IN A NEWBORN

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INTRODUCTION

Teratomas are true neoplasms composed of tissues of all three germinal layers. They have an unknown origin and eccentric microscopic appearance. Teratomas arising from the oral cavity are rare in the newborn. It is a benign tumor, although malignancy has been described in adults. In utero it can cause polyhydramnios or fetal death. In the newborn it can cause respiratory distress due to tracheal obstruction [1].

CASE REPORT

A 3.5 Kg female neonate born at 37 weeks gestation was brought at birth with a large, bony, fleshy, cylindrical mass 17 × 09 × 08cm in diameter protruding from her oral cavity. The obstetric history was unremarkable. Due to the oral mass, feeding was not possible and respiratory distress noted. She was referred from her hospital of birth (Allama Iqbal Hospital Sialkot) to our tertiary care unit, CMH Sialkot.

At the initial examination, a large bony fleshy cylindrical mass was noted protruding from her oral cavity. The mass was attached to a stalk that originated from the hard palate (cruciform suture) (fig. 1). As there was no associated anomaly and the mass was localized, an excisional biopsy was planned. Under general anesthesia, the mass was ligated and totally excised. After excision of the mass, hemostasis achieved (fig. 2). After removal of the tumour the mouth remained open due to maldevelopment of the jaw, the symphysis was inferiorly and posteriorly positioned. On next post operative day the patient developed respiratory symptoms and

was referred to the pediatrician but in spite of all efforts the child expired on 5th post operative day.

Histopathological examination of the mass revealed mature teratoma with mature keratinising squamous epithelium, skin adnexae, adipose tissue, neurological tissue and bone formation. Smooth muscle fibers were scattered throughout.

DISCUSSION

Teratoma occurs in 1 out of 4000 live births [1,2]. Head and neck occurrence is usually localized to the neck and nasopharynx and comprise 1- 10% of cases. Other extra cervical presentation of teratoma are very rare and only 13 cases of teratoma (seven from the tongue and six from the hard palate) have been reported in the literature [2-5].

The histogenesis of teratoma remains debatable. The most popular theory suggests that presumably due to alteration of cellular membrane chemistry, teratoma arises from totipotential embryonic tissue which somehow displaced during ontogeny. This leads to an assemblage of tissue often alien to the site in which they arise [6]. The synchronous presence of embryonal, fetal and adult tissue elements is possible and identifies the level and type of differentiation. Immaturity should be equated with malignancy and usually correlates with the immaturity of the host [7, 8].

Pathologically teratomas are designed as an unusual tumor composed of multiple heterotopic tissues foreign to the site in which they arise and classified into four types [9].

Dermoids are the most commonly found type of teratomas and contain tissue of ectodermal and mesodermal origin. Most tissues are present in the neonatal periods and are extremely rare in adulthood.

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Teratoids contain tissues from the three primary germ layers but are poorly differentiated.

True Teratoma is similar to teratoids since they contain tissues from the three primary germ layers but the layers are differentiated into recognizable tissue histology. There have been reports of malignant alteration of these tumors in the head and neck region and elsewhere.

Epignathi are also tridermal in origin but differentiated into recognizable organs, sometimes with limbs or even a second fetus visible. The condition is also known as "fetus in fetus". They are very rare and in the early literature, epignathy and teratoma of the skull base has been considered incompatible with life. Epignathus is a misnomer and its etymological meaning is "upon the jaw".

Head and neck tumours are uncommon but can be associated with airway obstruction and high rate of mortality in untreated patients. Teratomas of the oral cavity and pharynx are not infrequently pedunculated tumors, covered by hairy skin or mucous membrane and composed of tissues from three germ layers with varying degree of differentiation [10]. Sometimes within the tumor calcification is visible. Hairy polyps are less rare than non hairy polyps they are found more frequently in female infants, mainly of the left side.

Failing to diagnose and treat head and neck teratomas until late adolescence or adulthood causes a risk of malignant degeneration of up to 90% [11]. Alpha fetoproteins (AFP) have been shown to be a reliable indicator of disease activity [12,13].

Teratomatous polyposis is usually recognized in the first day or week of life. Most nasopharyngeal teratomas are a cause of various degree of airway obstruction. Large teratomas obstructing the airway can be a cause of asphyxiation just after birth. The majority of neonatal births with nasopharyngeal teratomas can be intubated without problem via the oral and nasal



Fig. 1: Pre-operative presentation of giant oral teratoma.



Fig. 2: Post operative appearance.

routes. The dislocated pedunculated polyp can be a cause of feeding difficulties.

CONCLUSION

Oropharyngeal tumors appear most frequently in the first week of neonatal life, giving symptoms connected with mechanical obstruction of airway. Surgical resection is the only management and may involve staged operation.

Favourable location and small size of the tumour can favour persistence of the tumour to later childhood or to the moment of its enlargement.

REFERENCES

1. Rybak LP. Obstructing oropharyngeal teratoma in neonates, a report of two cases. *Arch Otolaryngol Head Neck Surg.* 1991; 117: 1411-5.
2. April MM, Ward RF, Garelick JM. Diagnosis, management and follow-up of congenital head and neck teratoma. *Laryngoscope.* 1998; 108: 1398-1401.

3. Ashley JV, Shefer AD. Teratoma of tongue in a newborn. *Cleave Clin Q*. 1983; 50:34-7
4. Bras G, butts D, Hoyte DA. Gliomatosis teratoma of tongue. Report of a case. *Cancer*. 1969; 24: 1045-50.
5. Dudgeon DL, Isaac H Jr, Hayas DM. Multiple teratoma of the head and neck. *J Paediatr*. 1974; 85:139-40.
6. Batsakis JG. Pathology consultation. Nomenclature of developmental tumours. *Ann Otol Rhinol Laryngol*. 1984; 93: 98-9.
7. Green JS, Dickinson FL, Rickets A, Moir A. MRI in the assessment of a newborn with cervical teratoma. *Pediatr Radiol*. 2004; 28: 709-10.
8. Rothschild MA, Catalano P, Urken M, Brandwein M, Som P, Norton K, et al. Evaluation and Management of congenital cervical teratoma. Case report and review. *Arch Otolaryngol Head Neck Surg*. 1994; 20(4): 444-8.
9. Sexton M. Hairy polyp of the oropharynx, a case report with speculation on nosology. *Am J Dermatopathol*. 1990; 12: 294-8.
10. Hirabayashi S, Ueda K. Nasopharyngeal teratoma attached to the lower jaw. *Plast Reconstr Surg*. 1985; 76: 939-41.
11. Bucklay NJ, Burch WM, Leight GS. Malignant teratoma in the thyroid gland of an adult, a case report and a review of literature. *Surgery*. 1986; 100: 932-7.
12. Billmire DF, Grosfeld JL. Teratoma in childhood, analysis of 142 cases. *J Pediatr Surg*. 1986; 21: 548-51.
13. Marras T, Poenaru D, Kamal I. Perinatal management of nasopharyngeal teratoma *J Otolaryngol*. 1995; 24: 310-4.