

Oral Teratoma-An Unusual Presentation

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Abstract

An oropharyngeal teratoma (epignathus) is a rare malformation which is composed of cells from all the three germ layers. Epignathus may arise from the palate or pharynx and then protrudes out from the mouth. We intend to present a case of oropharyngeal teratoma originating from the hard palate in a newborn. The lesion was excised which on histopathological examination showed features of mature teratoma without malignant transformation. Recurrence was not observed after 12 months of follow-up.

Introduction

A teratoma is a tumour consisting of multiple tissues that are not indigenous to their site of origin [1]. Teratomas originate from the pluripotent cells and are composed of tissues from all the three germ layers. They are usually benign in nature. The most common sites of origin are the sacrococcyx, anterior mediastinum, testicle, ovary and the retroperitoneum [1,2]. Teratomas are exceedingly rare in the head and neck region. Most of the 10% of teratomas found in this area are seen in the nasopharynx and the cervical region [3,4].

Respiratory compromise is the most important complication of oropharyngeal teratomas, leading to death in most neonates [5,6]. Surgical resection is the treatment of choice for oropharyngeal teratomas, with few cases of recurrence [6,7]. We report a case of a hard palate teratoma in a neonate, who had no evidence of recurrence at 1 year of follow up.

Case Report

A full term male infant was born by normal vaginal delivery with a birth weight of 2.8 kg with a congenital tumour protruding from the mouth, which prevented oral feeding. Oral clinical examination revealed a well-defined mass measuring 4.0×3.0×2 cm³, originating from the hard palate. The prenatal and perinatal periods were uncomplicated, with no family history of similar lesion.

The infant was admitted to the neonatal intensive care unit (NICU) because of respiratory distress. Shortness of breath and cyanosis of the lip was observed, especially when the infant was being fed. Computed tomography was performed which showed an oropharyngeal mass, with no intracranial extension. The mass was excised under general anaesthesia from the hard palate and the mucosal defect was repaired by a local palatal flap, after a week of NICU admission. The oblong mass was variegated on cut section with a short peduncle. (Figure 1) No cerebrospinal fluid leakage occurred from the excision site. Post operatively, the wound healed well and the baby tolerated

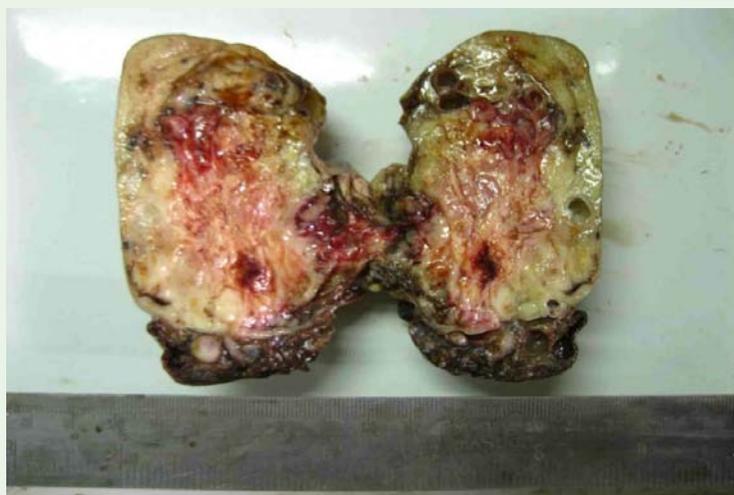


Figure 1: The oblong mass was variegated on cut section with a short peduncle with solid and cystic areas.

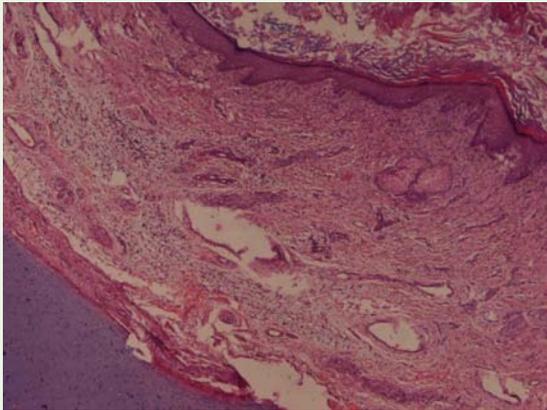


Figure 2: Histopathological examination revealed a mature teratoma composed of mature keratinized squamous epithelium, sebaceous glands, fibrous connective tissue and foci of mature chondrocytes. Haematoxylin and Eosin x 10X.

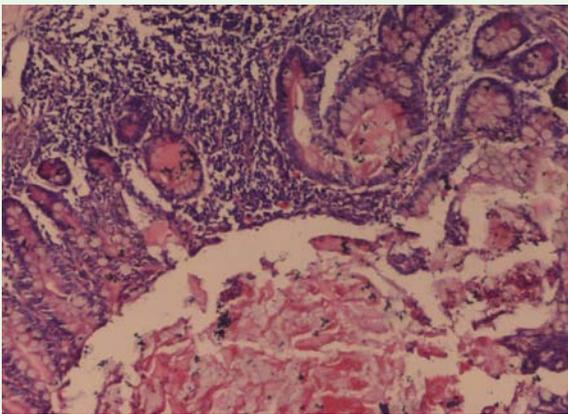


Figure 3: Microscopically tissue section shows mature glands and tubules. Haematoxylin and Eosin x 40X.

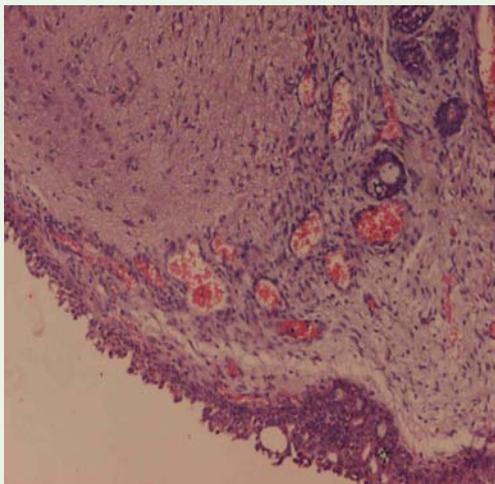


Figure 4: Tissue section shows pseudo-stratified squamous epithelium, mature neuroglial tissue and congested blood vessels. Haematoxylin and Eosin x 40X.

oral feeds with no further respiratory discomfort. Histopathological examination revealed a mature teratoma composed of mature respiratory epithelium, glandular tissues, keratinized squamous epithelium, neuroglial tissue, mature chondrocytes and blood vessels (Figures 2-4). Masseteric function and swallowing slowly improved over several weeks. The infant was discharged after a month and is well and symptom-free at 12 months follow-up.

Discussion

Teratomas are rare malformations containing cells from ectodermal, mesodermal and endodermal germ cell layers.⁸ They have an incidence of 1:4000 live births with less than 2% originating from the oropharynx [1,7].

Nasopharynx is one of the most frequent sites for head and neck teratomas with a 6:1 female predominance. In contrast, oral teratomas do not present a clear gender predilection and may arise from anywhere in the oronasal cavity [8,9]. Teratomas may be diagnosed ante-natally by imaging, which permits early multidisciplinary management.

The main therapy of teratomas is complete surgical excision, which minimizes the chances of malignant transformation [10]. The neonate's prognosis worsens as the size of the tumour increases. Unless the teratoma has an intracranial extension, resection of tumour may be attempted. Operation is difficult in lesions with intracranial involvement with poor prognosis [11]. Exclusion of intracranial extension is an important part of preoperative management [12]. Mortality rate associated with large teratomas in the head and neck are generally high in the absence of a proper anaesthetic work-up or meticulous delivery planning to secure the airway, as the majority of these teratomas are associated with obstruction of airway and difficulty in intubation. So, the initial treatment should be directed towards airway management and feeding problems.¹⁰ Stabilization of the airway should be the first priority in a neonate with respiratory discomfort [11]. Feeding was hampered in our patient due to the obstructive mass with fatal respiratory compromise and emergency airway management was advocated.

Grossly, these tumours have a variegated appearance with cystic and solid areas. Histologically, teratomas represent tissues of all the germ cell layers. The most common tissues observed are the nerves, cartilages, muscles, skin and respiratory epithelia [9]. Our case showed mature respiratory epithelia, glandular tissues, keratinized squamous epithelium, cartilage and blood vessels. Teratomas are associated with concomitant malformations in 6% of all cases, with cleft palate being the most commonly associated anomaly [5]. Our case also presented with cleft palate, which was corrected surgically by local palatal flap repair.

Teratomas in the neonates are mostly benign and consist of mature tissue components [13]. While teratomas with malignant potential occur in adults and contain immature tissues, with a high incidence of malignancy. Incomplete resection and presence of primitive neural tissue are associated with a malignant relapse [13,14].

Conclusion

Congenital epignathus is prone to recurrence and fatal outcome. Hence significance of strict follow-up screening should be emphasized.

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