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CASE STUDY

NUMEROUS FACES OF THE MYSTERIOUS MONSTER! IMMATURE TERATOMA TONGUE- A RARE CASE REPORT AND A DISCUSSION ON ITS VARIED HISTOPATHOLOGY

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ABSTRACT

A teratoma is a true neoplasm, containing tissue from the ectodermal, mesodermal and endodermal layers. (Ward and April, 1989) Teratoma of the head and neck region are very rare, true neoplasms of unknown etiology, composed of an assemblage of tissue often alien to the site in which they arise. (Becker *et al*., 2007; Haghighi and Cleveland, 2004; Tolentino *et al*., 2005). The present article describes a case report of a lesion with an unusual location and a detailed description of the histopathology.

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INTRODUCTION

Teratomas may be located in the midline from the brain to the sacral area, but are most common in the sacrococcygeal area. 90% of head and neck teratomas occur in the nasopharyngeal area and neck, teratomas from other oral structures especially those occurring on the tongue are extremely rare. (Ward and April, 1989; WHO, 2005; Biglioli,1996; Okafor *et al*., 2004; Nasir *et al*., 2006)

Case report

A 3day old female baby was brought to the clinic with a growth from the oral cavity. The irregular mass measuring 4.5 X 3.5 X 2cms, reddish white in colour, variable in consistency was present on the dorsal and lateral surface of the tongue (Fig.1). The infant was experiencing difficulty feeding and breathing. After pre-operative evaluation the lesion was surgical excised. On macroscopic examination 3 tissue masses measuring approx 4.5X4X2.5cms, 1X1X1cms, reddish white in colour, soft to firm in consistency and surrounding tongue tissue measuring 2.5X2X2cms (Fig.2). Histopathological examination revealed mature stratified squamous epithelium, keratinizing cysts, ciliated columnar epithelium, odontogenic epithelium, Neuroepithelium, developing serous and mucous salivary glands, mature fibrofatty tissue, bone, cartilage, skeletal muscle, loose myxoid tissue and nests of clear cells (Fig. 3-14). Since tissues from all three germ layers could be identified a diagnosis of teratoma was made. Post surgical examination of the patient revealed satisfactory healing and recurrence till date (Fig. 15).

DISCUSSION

The term 'teratoma', derived from the Greek word 'teraton' (meaning monster), was first used by Virchow in 1863. A teratoma is a true neoplasm, containing tissue from the ectodermal, mesodermal and endodermal layers. It is also described in the WHO blue book as a "Tumour composed of a variety of mature tissues that are foreign to the site of occurrence. There are typically tissues derived from two or three germ layers." (WHO, 2005) Teratomas may arise from different sites of the body; the most common site in the newborn is the sacrococcygeal region, (accounting for nearly 40% of the total cases). 2-9% occur in the head and neck. (Biglioli *et al*., 1996; Okafor *et al*., 2004; Nasir *et al*., 2006) The present case occurred on the tongue which is a very rare site for the rare oral teratomas.

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Fig. 1. Clinical photograph demonstrating the a lesion on the tongue of a neonate

Fig. 2. Macroscopic appearance of the excised tissue

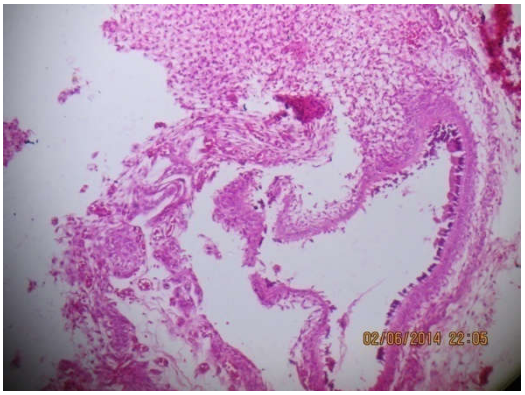

Fig. 3. Odontogenic epithelium

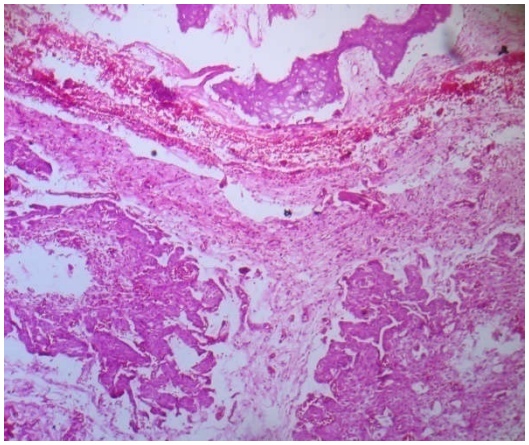

Fig. 4. Neuroepithelium

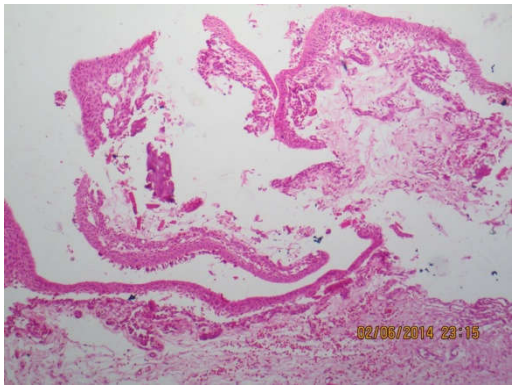

Fig. 5. Respiratory epithelium

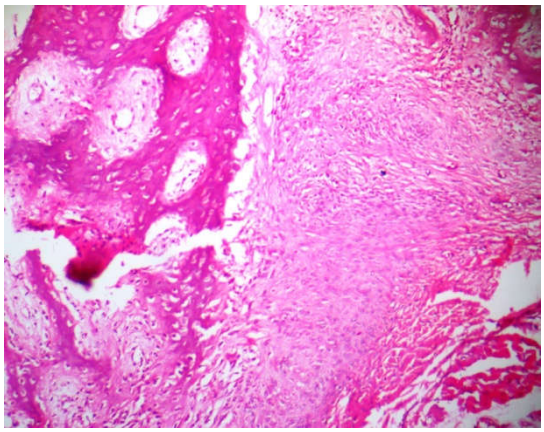

Fig. 6. Bone

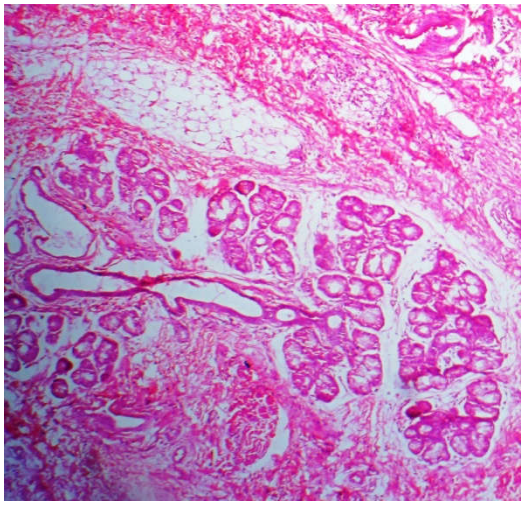

Fig. 7. Seromucous glands

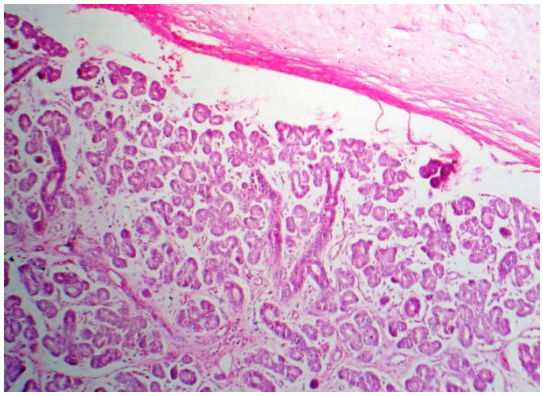

Fig. 8. Serous glands

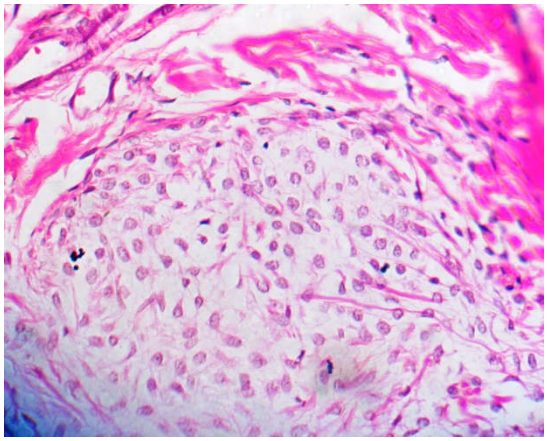

Fig. 9. Nests of clear cells

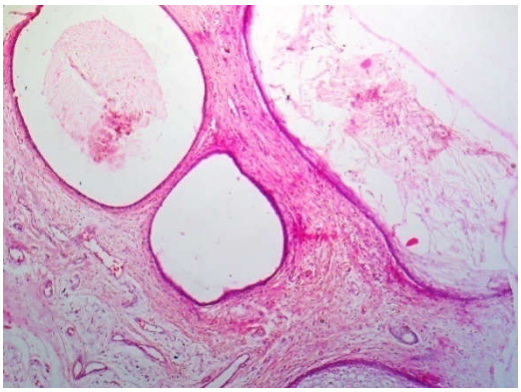

Fig. 10. Epithelial cysts

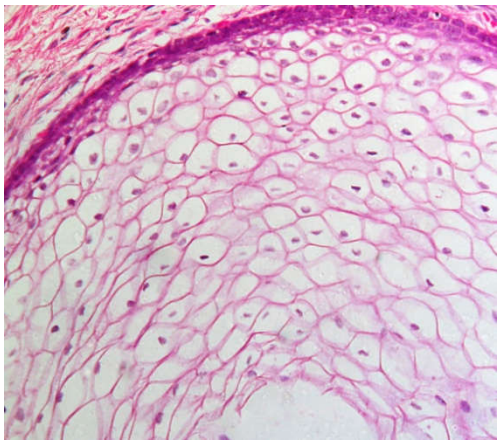

Fig. 11. Stratified squamous epithelium

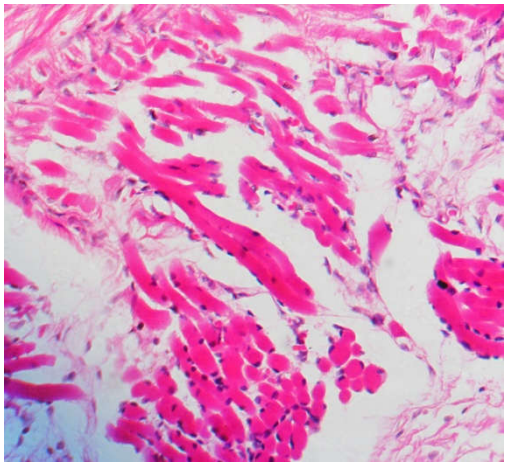

Fig. 12. Muscle

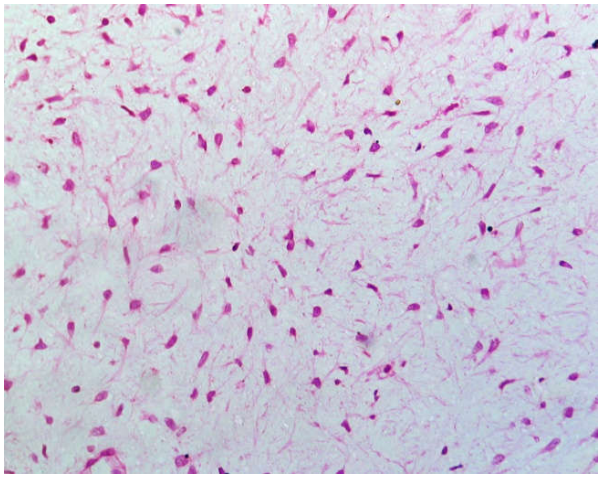

Fig. 13. Myxoid tissue

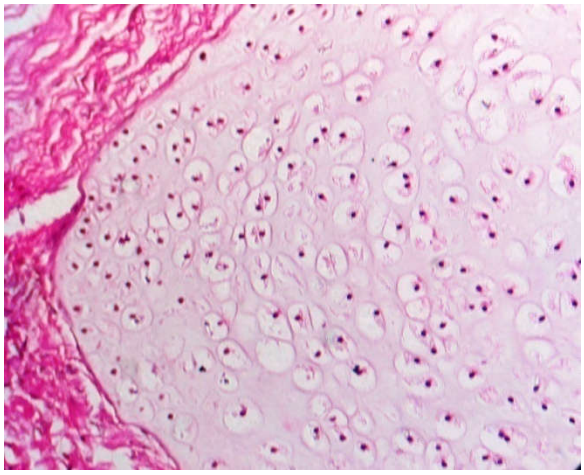

Fig. 14. Chondroid tissue

Fig. 15. Post operative follow showing satisfactory healing

Etiology of teratoma is unknown, but various theories have been proposed to explain their etiology like

1. Acquired traumatic implantation of skin or mucous membrane within deeper tissues.
2. Congenital inclusion of germ layers into deeper tissue at point where developing somatic regions fails to completely fuse during embryogenesis.
3. Implantation of pluripotential cells, which subsequently grow in a disorganized manner (Haghighi and Cleveland, 2004).

The term teratoma is often used to describe any of the four types of tumor that constitute tissue foreign to the area in which it is located. These include dermoids (hairy polyps), teratoid, true teratomas, and epignathi. In 1992, Tharrington and Bussen proposed following histologic classification for the congenital tumors (Tolentino *et al*., 2005).

1. Dermoids are neoplasm composed of only of ectodermal and mesodermal elements.
2. Teratoids contain ectodermal, mesodermal, and endodermal elements. These elements are however not well differentiated and lack clear organization.
3. True teratomas contain tissue derivatives from 3 germ layers. Cell layers demonstrate more organization.
4. Epignathi are the least frequent but most striking of teratomas in that well-formed organs and limbs may be found occurring in abnormal locations (e.g., an arm coming out of the oral cavity) (Tolentino *et al*., 2005).

Generally, an epignathus is a highly developed oral tumor; however, the term specifically refers to a jaw or alveolar point of origin. (El-Musa *et al*., 2006) Ozolek *et al* in their article state that whenever they have inspected sections from teratomas under the microscope, they have found something, some structure that was not present on previous viewing and this is part of what makes them special and fascinating lesions to study. In fact, the process of histopathological reporting gets difficult when considering the teratoma since tissues from all three primordial germ layers are not only present but are present in a developmentally heterogeneous fashion. That means that a single tissue type can be represented as it might appear histologically anyway it would from embryogenesis to adulthood. Teratomas are often large, well-circumscribed masses firmly attached to adjacent parenchymal structures. The tumors are generally multicystic and composed of a mixture of recognizable mature elements including keratin balls, hair, cartilage, and bone. Immature elements are less obvious but are commonly associated with areas of necrosis and hemorrhage. The histologic appearance of teratomas varies according to the presence of these immature elements and their degree of differentiation. As in the gonadal and extragonadal examples, three variants are identified: mature, immature and teratomas with malignant transformation. Histologically mature teratomas are composed of fully differentiated, “adult-type” ectodermal, mesodermal, and endodermal elements. These are often organized in an orderly pattern resembling adult tissues, for example, skin with adnexae, cartilage and bone, adipose tissue, bundles of striated and smooth muscle, glioneuronal tissue with choroid plexus, and retina with pigmented ocular epithelium. Immature teratomas are composed of incompletely differentiated or immature elements derived from one or more of the three germinal layers. Teratomas with malignant transformation show a malignant component of one or more of the three adult tissues as typically encountered in other organs and tissues for example, carcinomas and sarcomas, admixed with the regular mature or immature teratomatous elements. The present case on histopathological examination revealed structures from all the three germ layers as described above in the various photomicrographs and more than 10% and less than 50% tissue was made up with immature tissues and hence the reported case represents an immature teratoma. (Gnepp, 2013) The tumor was surgically excised and the patient is under regular follow up. Though operative management is the treatment in most cases especially in the developing countries, prenatal ultrasound diagnosis can be made early in pregnancy (15–16 weeks); 3D ultrasound and MRI may enhance the accuracy of the antenatal diagnosis and the delivery should involve elective cesarean section with the exit procedure or resection of the tumor mass, which may be performed to increase the chances of postnatal survival. (Tonni *et al*., 2010)

Conclusion

Teratomas are rare tumors of the oral cavity. Though often benign may cause respiratory distress and need to be treated at the earliest. With the advances in antenatal diagnosis and its availability it is time we start treating them earlier. On the other hand more research needs to be done into the development of these tumors and various cell lines that can be obtained from them because it may answer certain questions regarding development and tumorigenesis.

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