The recurrence of a soft palate teratoma in a neonate: a case report

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Abstract
Teratomas are rare malformations containing cells from ectodermal, mesodermal and endodermal layers, seldom seen in the soft palate. A 2-day-old girl presenting a teratoma arising from the soft palate was surgically treated. Unfortunately, recurrence was observed during follow-up.

Introduction
Teratomas are rarely observed in the soft palate. A teratoma, as defined by Weaver et al., is a tumour consisting of multiple tissues that are not indigenous to their site of origin. The most important complication of oral teratomas is respiratory compromise, which is the main cause of death in neonates. Surgical resection is the treatment of choice for oral teratomas. Although the follow-up is short in most cases, there are no reports of recurrence of oral teratomas. A case of a soft palate teratoma in a neonate is reported herein; recurrence was observed in 4 years follow-up.

Case report
A female infant was born to a 20-year-old mother by vaginal delivery after 37+3 weeks gestation with birth weight of 3.05 kg. A congenital tumour was found in the mouth, which prevented oral feeding. Oral clinical examination revealed an asymptomatic mass measuring 3.0 × 4.0 × 2.5 cm, originating from the soft palate near the midline (Figure 1). The mother did not undergo a three-dimensional (3D)-ultrasonography examination during her pregnancy. The prenatal and perinatal courses were uncomplicated, and the family history was negative. The father was 27 years old and in good health. The infant was admitted to our department on the second day after birth because of irregular respiratory cycle. Shortness of breath and lip cyanosis was observed, especially when the infant was being fed. Due to repeated episodes of apnea, tracheal intubation ventilator support was provided and the infant was transferred to a neonatal intensive care unit. Computed tomography showed an oral cavity mass without intracranial extension. On the seventh day after birth, under general anaesthesia with intubation, the mass was excised from the soft palate finding a short peduncle 1 cm in diameter and the mucosal defect was repaired by suturing a transferred local palatal flap (Figure 2). No cerebrospinal fluid leakage occurred from the excision site. After surgery, the wound healed well and the baby tolerated oral feeds. No respiratory compromise occurred anymore. Histopathological examination revealed a mature teratoma composed of mature respiratory epithelium, glandular tissues, neuroglial tissue, choroid plexus, muscle and blood vessels (Figure 3). Some margins showed nerve and muscle. Signs of recurrence were found in the second year of follow-up. Magnetic resonance imaging (MRI) indicated maxillary tumour with cystic and solid areas with fat density (Figure 4). The parents wanted to "wait and watch", while hoping for a better prognosis, instead of opting for immediate reoperation.

Discussion
Teratomas are rare malformations containing cells from ectodermal, mesodermal and endodermal layers with a variable degree of differentiation. They have an incidence of 1:4000 live births; less than 2% of these arise from the oropharyngeal cavity. Nasopharynx is one of the most frequent sites for head and neck teratomas and has a 6:1 female predominance. In contrast, oral teratomas do not present a clear gender predilection. Oral teratomas arise anywhere in the oronasal cavity and are regarded as expanding, cavity-filling lesions, especially in the neonatal period.

The main therapy of teratomas is complete surgical excision, which depends on the site of the tumour. Unless the teratoma is expanding massively into the cranial area, resection of tumour may be attempted. Initial treatment should be directed toward airway management and feeding problems. When a neonate is experiencing respiratory difficulty, the first priority should be stabilization of the airway. Our case presented with an obstructive mass causing respiratory embarrassment and immediate threat to life, which demanded establishment of an airway with tracheostomy.

Histologically, teratomas may present different characteristics. In oral teratomas, the most common tissues observed are nerves and cartilages. Other tissues commonly seen are muscles, bones and respiratory epithelia. Our case showed mature respiratory epithelia, glandular tissues, neuroglial tissues, choroid plexuses, muscles and blood vessels. Teratomas are associated with concomitant malformations in 6% of all cases, with cleft palate being the most commonly associated anomaly. In the present case, congenital cardiac septal
defects and patency of ductus arteriosus were detected. Richieri-Costa et al. have also reported the observation of cardiac abnormalities.

Teratomas are mostly benign in the neonatal period 5% of the cases present malignity criteria on histopathology. Usually, benign teratomas consist of mature tissue components, while those with malignant potential contain immature tissues; there is a higher incidence of malignancy in adults. Incomplete resection and presence of primitive neural tissue entail the risk of a malignant relapse. Becker et al. reported the recurrence of a congenital epignathus post-operation; this did not necessarily imply malignancy, although the clinician should continue follow-up screening.

It has been 3 years since the recurrence of oral teratoma; the girl is still in good condition without any drugs or surgery. Because the risk of malignant change is evident, long-term follow-up is mandatory.

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References