Introduction

Teratoma is the most common germ cell tumour in childhood.1 The most common origin sites of teratomas in children are the sacrococcygeal region, gonads and mediastinum.2 Teratomas involving the head and neck are rare, occurring once in every 40,000 births.2 In addition, teratomas of the neck region represent one of the most unusual causes of respiratory distress during the neonatal period.3,4 In this paper, we present a case of an immature teratoma in the parapharyngeal space presenting with airway obstruction in an infant.

Case Report

A 12-day-old male infant was referred to our hospital with inspiratory stridor and respiratory difficulty. The patient was the second child of a 27-year-old Korean woman. The infant was born at 40 weeks estimated gestational age in a normal vaginal delivery. Weight at birth was 3560 g. There was no history of teratogens or polyhydroamnios during gestation. Family history was unremarkable.

A flexible laryngoscopy was performed to rule out congenital anomalies of the larynx. The nasal cavity, larynx and external appearance were non-specific. Flexible laryngoscopic examination revealed a protruding soft palate mass covered by a normal mucous membrane and causing airway obstruction (Figure 1). Computed tomography (CT) revealed a well-defined mass with a fatty component and calcification in the left parapharyngeal space measuring 3 cm × 4.5 cm × 3 cm, and severe occlusion of the airway (Figure 2). Magnetic resonance imaging (MRI) identified a soft tissue mass in the left parapharyngeal space with low intensity on T1-weighted images, high intensity on T2-weighted images and heterogeneous enhancement on enhanced T2-weighted images (Figure 3). MRI also revealed a heterogeneous enhancing lesion in the left anterior suprasellar area adjacent to the supraclinoid portion of the left internal carotid artery with internal calcification and a fat component (about 1.2 cm).
There has been no recurrence in the oropharynx and there has been a quiescent lesion (i.e. with no progression) in the parapharynx without any respiratory problem for 4 years. There has also been no change in the enhancing lesion in the left suprasellar area, which is suggestive of a teratoma.

**Discussion**

Teratomas are derived from the three germinal layers and show varying degrees of differentiation. Pathologically, teratomas can be broken down into three groups: mature teratoma, immature teratoma and malignant teratoma. Immature teratomas are characterised by the presence of immature neural tissue and a greater propensity for recurrence.

Teratomas of the head and neck are usually present at birth, and patients present with signs and symptoms of airway obstruction. Eighty percent of teratomas found in the first two months of life occur in the sacrococcygeal region, and 5-14% occur in the head and neck region. The most common sites of occurrence in the head and neck region are the cervical region and nasopharynx. Large teratomas in the head and neck cause severe respiratory distress. In this case, the infant presented with symptoms of airway obstruction after birth without anaesthesia. There has been no recurrence in the oropharynx and there has been a quiescent lesion (i.e. with no progression) in the parapharynx without any respiratory problem for 4 years. There has also been no change in the enhancing lesion in the left suprasellar area, which is suggestive of a teratoma.

These findings led to a pre-operative suspicion of teratoma. An intra-oral left parapharyngeal mass excision with a longitudinal palatal incision was performed under general anaesthesia. A tumour in the left parapharyngeal space was identified and removed (Figure 4). However, it was impossible to distinguish from normal tissue because it lacked a capsule and there was a considerable adhesion. The histopathological examination identified an immature teratoma, grade 1. The postoperative course was uneventful. Five months after the operation, recurrent masses were found in the left oropharyngeal and parapharyngeal regions (both measuring approximately 1.5 × 1.5 cm). The oropharyngeal mass was removed under general anaesthesia.

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**Figure 2**
CT shows a well-defined mass with fatty component and calcification (arrow) in the left parapharyngeal space measuring 3 cm × 4.5 cm × 3 cm, and severe occlusion of the airway.

**Figure 3**
MRI shows a mass (arrow) with low intensity on T1-weighted images (A), high intensity on T2-weighted images (B), and heterogeneous enhancement on enhanced T2-weighted images (C).

**Figure 4**
Intra-operative picture shows a tumour in the left parapharyngeal space after longitudinal palatal incision.
Immature teratoma of the parapharynx in an infant

any external deformity of the head and neck region. It was therefore difficult to diagnose teratoma of the head and neck initially before radiological examination.

Teratomas are frequently diagnosed during the prenatal sonogram. The reported CT and MRI features of teratoma are cystic and solid areas, and areas of fat density. CT has also demonstrated regions of calcification in most teratomas, and the high incidence of calcification on CT scan suggests the possibility of teratoma. The diagnosis suggested by CT was confirmed histopathologically in this case. MRI may be subsequently performed for better pre-operative mapping, although MRI is insensitive in terms of detecting calcifications.

The treatment of choice for head and neck teratomas is the complete excision of the lesion. However, teratomas involving the deeper soft tissue of the oropharynx are less amenable to surgical excision, as in our patient. Incomplete resection and female gender are important risk factors for relapse or death. After total excision, the survival rate with no recurrence for 3 years is reported to be 93%. In recurrent disease, surgery combined with chemotherapy results in 98% to 100% survival rates for 3 years. Untreated teratomas have mortality rates of 80% to 100%.

Conclusion

In conclusion, although teratomas of the head and neck are uncommon, they typically emerge during the neonatal period and are associated with airway obstruction and high infant mortality rate. Surgeons should consider the possibility of head and neck immature teratoma in the differential diagnosis of respiratory distress in an infant, even if the infant does not have an external deformity. In addition, the surgeons should also recognise the possibility of other accompanying congenital anomalies.

References