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Retrospective analysis of ten cases of congenital sublingual teratoid cyst

ABSTRACT

Aim The aims of this study were to perform clinicopathological analyses of teratoid cysts of the floor of the mouth and to assess the possibility of surgical treatment.

Materials and methods The incidence, manifestations, histogenesis, treatment and prognosis of 10 patients admitted to our department between April 2000 and May 2008 were reviewed.

Results Teratoid cysts occurred almost exclusively in neonates and children, and appeared as mobile, cystic solid and well-defined lesions. Because of the special location, consequences such as difficulty in feeding, breathing and even impairment in growth and development frequently occur. Results of histological examination indicated that squamous, respiratory and gastrointestinal epithelium composed the basic structure of teratoid cyst. Transonogram, CT and MRI examination were conducted, and complete surgical excision was the only treatment of choice. Usually, immediate treatment is not necessary unless the cyst obstructs the upper airway. There has been no recurrence of the lesion after surgery.

Conclusion Although teratoid cyst of the floor of the mouth is a cystic disease, it affects patients to a great extent because of its special location. Therefore, early and complete excision is mandatory for good prognosis.

Keywords Sublingual; Teratoid cyst; Clinicopathological analysis.

Introduction

Congenital teratoid cyst, also known as heterotopic oral gastrointestinal cyst, is a very rare developmental cyst. It is a type of dermoid cyst that probably arises from ectodermal differentiation of multipotential cells along the lines of embryonic fusion points. They occur primarily in the testes and ovaries that are trapped during the midline fusion of the first and second branchial arches [Meyer, 1955; Gold et al., 1974; Ettinger and Manderson, 1973]. Of all dermoids, nearly 7% are found in the head and neck, most commonly at the lateral third of the eyebrow and 25% of them is seen in the floor of the mouth [New and Erich, 1937; Howell, 1985; Taylor et al., 1966]. According to Meyer’s classification and complexity, teratoid cyst are oral dermoid cysts: a squamous-cell or respiratory-tract epithelial-lined cavity with skin appendages, distinct mesodermal components (such as fat, muscle, bone, and blood vessel), and endoderm components (such as respiratory and gastrointestinal tissues) [Faerber et al., 1988; Gol et al., 2005]. Rarely, they may show malignant transformation.

We analysed the clinical data of ten congenital sublingual teratoid cysts on patients admitted to our department between April 2000 and May 2008, and discussed the histogenesis, clinical feature, clinical pathology, diagnosis, treatment timing and prognosis.

Clinical data

The study was approved by the Ethics Committee of our university. We have followed the guidelines outlined in the Helsinki Declaration during this study. Ten children with congenital sublingual teratoid cyst were admitted to our department between April 2000 and May 2008, six males and four females. At the time of the preliminary visit age ranged from one day to 15 months with an average age of two months, and the age of the subjects at the time of surgery ranged from 28 days to 48 months, with an average age of ten months.

The average measurements of the cysts was 2.55 cm x 3.20 cm. All of them were sublingual, soft, well-defined and smooth.

General health status of patients

The patients had no apparent symptoms except snoring, in two cases, and insomnia. The cysts included ivory and creamy materials. The preoperative laboratory examination results for the ten cases are shown in Table 1. The average weight of the patients was 7.8 kg (the normal average weight of 10-month babies is 8.0 kg); the average HGB was 115.4 g/L (normal value is 115-175 g/L); and the average HCT was 0.345 L/L (normal value is 0.37-0.52 L/L).
Treatment
The patients underwent a transpolar operation under general anaesthesia delivered by oral tracheal intubation. A transverse mucosal incision was made over the sublingual mass. Submucosal dissection was made and a well-encapsulated mass from the sublingual area was delivered. No dense attachment to neighboring tissues was detected. During the surgery, bilateral lingual nerves, sublingual glands and submaxillary ducts were protected. After the surgery, a drainage system was set up in the sublingual area. A retention suture was placed on the tongue. The patients were intubated for 1-2 days after surgery because of the concerns of tongue swelling, and then sent back to the sickroom from the SICU.

Pathological diagnosis
Histopathological examination revealed the presence of endodermic (gastrointestinal tract mucosa and respiratory mucosa), mesodermic (smooth muscles, cartilage, skeletal, fat, bone, skeletal muscle and blood vessels) and ectodermic (hair, skin, sebaceous glands, and fair follicles) tissues in all of the teratoid cysts (Fig. 1, 2). A histopathologic diagnosis of teratoid cyst was made.

Classic case
A 63-day-old infant was referred to our department with a sublingual mass pushing the tongue upward and causing difficulty in feeding and breathing, and prostration. The baby also snored in the lateral decubitus position. The mother stated that the cyst had been identified at birth but progressed rapidly during the 63 days before the visit. There was no history of infection, systemic disease, or other congenital diseases. The baby exhibited little appetite, poor sleep, normal urine and stool, and normal weight (birth weight was 3.05 kg and weight upon admission was 4.0 kg). Clinical examination revealed a 4 cm × 4 cm mass of soft consistency that was firm, high tension, and fluctuant.

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Gender</th>
<th>Age at preliminary visit</th>
<th>Age at surgery</th>
<th>Measurements (CM × CM)</th>
<th>Preoperative weight (KG)</th>
<th>Follow-up (years)</th>
<th>Respiratory or feeding issues</th>
<th>Malignant degeneration</th>
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**TABLE 1** Basic information of the ten cases.

**FIG. 1** (A) Gastrointestinal tract mucosa (endoderm tissue); (B) Stratified pavement epithelium (ectoderm tissue); (C) Smooth muscles (mesoderm tissue) [HE stain, magnification 100×].

**FIG. 2** (B) Stratified pavement epithelium (ectoderm tissue); (C) Smooth muscles (mesoderm tissue); (D) Sebaceous glands; (E) Sweat glands [HE stain, magnification 100×].
which pushed the tongue upward against the palate and filled the floor of the mouth. It had well-defined borders, a smooth surface, normothermia, reddish in color and was covered by normal mucosa. The results of the differential diagnosis tests of position moving and volume compressing were all negative, as it is for most cysts. There was no regional lymph node enlargement. Laboratory examination results were as follows: WBC 12.4 × 10⁹ /L, RBC 4.29 × 10¹² /L, HGB 124 g/L, and HCT 0.373 L/L. Computed tomography (CT) scan revealed a midline, hypodense, 24.5 × 26.8 × 23.9 mm sublingual mass resembling a cyst with no augmentation in enhancement CT (Fig. 3, 4).

Results

All surgical incisions healed uneventfully. No wound infection or obstruction of the upper respiratory tract occurred after the surgery and all related symptoms disappeared. No recurrence of the lesion was observed at the follow-ups (1.5-9.5 years with 4.9 years average).

Discussion

Congenital dermoid cysts occur primarily in the testes and ovaries [Faerber et al., 1988] and can be divided into three histological varieties: dermoid (compound), epidermoid (simple), and teratoid (complex). Dermoid cyst is an epithelial-lined cavity that shows skin appendages in the wall of the cyst. Epidermoid cyst has no skin appendages, whereas the lining of the teratoid cyst ranges from simple squamous to ciliated respiratory epithelium and contains derivatives of ectoderm, mesoderm, and endoderm [Goel et al., 2005]. Teratoid cysts may contain embryonic layer elements such as muscle, bones, teeth, and mucous membranes. Of all dermoids, only 7% occur in the head and neck region [Faerber et al., 1988] and teratoid cysts of the oral cavity are rare [Bonilla et al, 1996; Harada et al., 1995]. New and Erich [New and Erich, 1937] have found that only 1.6% of congenital dermoid cysts involving the oral cavity arise from the floor of the mouth. Taylor et al. [1966] have reported that 6.5% of 541 congenital dermoid cysts of the head and neck are located intraorally. They are painless lesions that can displace the tongue. Patients may present with dysphagia, dysphonia and dyspnoea [Howell, 1985; Longo et al., 2003].

Our data show that teratoid cyst lacks pathognomonic symptoms, which makes a definite diagnosis difficult. Ranula or sialolithiasis is usually the first hypothesis of lesions in the floor of the mouth. However, these conditions hardly ever affect the infants. Moreover, in these cases, the clinical and radiographic aspects were not compatible with these two conditions because of the solid appearance and the diffuse radiodensity of the mass. And we should also diagnosis whether the cyst is an epidermoid cyst, dermoid cyst, ectopic thyroid gland, thyroglossal tract cyst, hemangioma/lymphangioma, sialadenitis, oral alimentary tract cyst, or cystic teratoma [New and Erich, 1937]. They appear in two different ways depending on their relationships with the mylohyoid muscle. Teratoid cysts superior to this muscle are considered intraoral swelling, whereas those inferior to it manifest as swelling in the midline of the neck. Fine-needle aspiration biopsy may be helpful in the differential diagnosis of lesions located in the floor of the mouth.

We can diagnose the lesions by radioisotope scan, cyst content smear, ultrasonic testing, CT and MRI. CT and MRI can delineate the internal architecture and reveal the relationship between the cyst and

FIG. 3 CT examination (anteroposterior view).

FIG. 4 Preoperative image.

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the mylohyoid muscle. CT typically shows a midline unilocular mass with a well-defined capsule, which may be enhanced with contrast material. However, MRI characteristics are variable. Teratoid cysts are usually isointense or hypointense to muscle on T1-weighted images and hyperintense or heterogeneous on T2-weighted images, depending on their internal content [Som and Curtin, 1996]. MRI would show excellent contrast and definition due to the high signal intensity of the mass on T2 weighted image. We can only make a definite diagnosis by histopathology.

Because of the special position of the congenital teratoid cyst, it can stress the upper respiratory tract and pharynx in newborns, or even block the respiratory tract and endanger the patients if not handled in a timely manner. So it is important to prevent and take emergency therapeutic measures including early discovery of the mass by antenatal imageology in ultrasound. In order to do that, parturition must be carefully assisted in order to avoid tearing of the mass (or cyst) and also massive haemorrhage; moreover endotracheal intubation is suggested before the placental circulation is cut off. If the respiratory tract is narrow or tortuous, tracheotomy should be performed immediately. Then the patient can be sent to the neonatal intensive care unit, where further examinations should be performed to understand the extent of the disease and eliminate intracranial lesions as soon as the respiratory tract is stable. On this occasion, surgical resection should be performed as soon as possible. After the surgery, good care should be taken of the respiratory tract, especially the mouth floor, pharyngeal edema and secondary hemorrhage [Gold et al., 1974; Bonilla et al, 1996]. But because of the high-risk of general anaesthesia in newborns, if the mass does not seriously stress the respiratory tract, we suggest postponing the operation. Aspiration is useful as a temporary relief, which help to postpone the surgical excisions.

In the classic case reported here, however, it was not possible to postpone the surgery because the lesion was rapidly growing, interfering with breastfeeding, and causing concerns on the respiratory tract. Surgical excision is the treatment of choice. Modest oedema is the most common postoperative problem encountered, with little alteration in tongue movement, swallowing, and breathing.

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References