Infancy congenital tongue base cyst: clinical analysis of 37 cases

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Abstract

Objective: To investigate the clinical characteristics of infant congenital tongue base cysts. Methods: This retrospective study involved 37 infants with congenital tongue base cysts in our hospital. Results: The median age at admission was 95 (15-360) d for these babies. The main clinical manifestations were laryngeal stridor (33/37 89.2%), dyspnea (8/37 21.6%), inspiratory trisomy (6/37 16.2%), and choking and vomiting milk (14/37 37.8%). Eight cases (21.6%) were misdiagnosed as laryngomalacia in another hospital, while 4 cases (10.8%) were misdiagnosed as pneumonia. One case required emergency intubation for rescue due to dyspnea before surgery. The root of the tongue was found to occupy space during intubation. Six cases (16.2%) had cysts found through CT examination. Three cases (8%) had cysts found during bronchoscopy due to pneumonia. The remaining cysts were found in the root of the tongue through laryngoscopy. Laryngoscopy was performed in all children before the operation, and CT examination in 28 patients before the operation showed that the median diameter of the cyst was 10 mm (5-20 mm), All children underwent radiofrequency ablation of glossal root cysts through laryngoscopy. Eight patients were transferred to the intensive care unit (ICU) for treatment post-operatively. The median intubation time was 4.5 days (2-5 days). Conclusion: Infancy congenital tongue base cysts have an early onset and atypical clinical manifestations. Electronic laryngoscopy combined with neck CT should be performed promptly in patients with laryngeal stridor and feeding difficulty to facilitate accurate diagnosis, and timely surgery is needed.

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Succinct key points:

1、Congenital tongue base cyst in infants is located at the bottom of the tongue root, with a low incidence.

2、This retrospective study involved 37 infants with congenital tongue base cysts in our hospital.

3、Infancy congenital tongue base cysts have an early onset and atypical clinical manifestations.

4、Electronic laryngoscopy combined with neck CT should be performed promptly in patients with laryngeal stridor and feeding difficulty.

5、It is vital for these patients to be diagnosed and treated as soon as possible.

Key words: Airway obstruction; infant; thyroglossal cyst; laryngoscopy

Congenital tongue base cyst in infants is located at the bottom of the tongue root, with a low incidence, which can cause respiratory and swallowing disorders[1]. The clinical manifestations of congenital tongue root cysts are similar to those of congenital laryngeal chondromalacia and congenital laryngeal web, which are easily misdiagnosed. This paper analyzes the clinical characteristics of 37 cases of congenital tongue root cysts diagnosed and treated in our hospital.

Methods:

This study is a retrospective study. From January 2014 to January 2022, 37 cases of congenital tongue base cysts confirmed by surgery and pathology in the Department of Otolaryngology of"Blinded for review" were included in this study for analysis.

The diagnosis mainly depends on the patient's medical history, physical examination and auxiliary examination, as follows: (1) The age of onset and visit ranged from 0-1 years old. (2) Main symptoms and signs: laryngeal stridor and feeding difficulty were found at admission. Inspiratory trisomy was found on physical examination. (3) Auxiliary examination: neck CT showed round or oval cysts at the root of the tongue, clearly demarcated from the surrounding tissues, with regular contours. Electronic laryngoscopy revealed a hemispherical gray-white cystic neoplasm at the root of the tongue, with a smooth surface, squeezing the epiglottis and blocking the upper airway. (4) Pathological examination after the operation mainly showed squamous epithelium, interstitial edema and congestion. Combined with the history, signs, auxiliary examinations and pathological results, congenital cysts of the root of the tongue can be diagnosed. We retrospectively analyzed the children's sex, symptom onset time, visit time, main manifestations, complications such as pneumonia and laryngomalacia, intubation before and after surgery, admission to the ICU, neck CT, treatment and postoperative outcomes.

The patients were followed up 6 months to 7 years, with an average of 56.6 months.

The descriptive analysis method was adopted. Count data are expressed as the number of cases and percentage. Measurement data with a normal distribution are expressed as ?±s, and measurement data with a nonnormal distribution are expressed as the median (min-max).

The clinical protocol used for these cases was established according to the ethical guidelines of the Helsinki Declaration and was approved by the Human Ethics Committee of "Blinded for review".

Ethical considerations

This study was approved by the Institutional Ethics and Research Committee of Our hospital

Results:

Among the 37 children with congenital tongue base cysts, 23 were male, 14 were female, 17 were born via cesarean section, and 20 were spontaneous deliveries. All of them were full-term infants, with a birth weight of 3331.6 ± 389.87 g.

The median admission age of the 37 children was 95 days (15-360 days). Twenty-six (70.3%) children had laryngeal stridor and inspiratory dyspnea after birth. Two (5%) patients were less than one month old. Eight (21.6%) patients were misdiagnosed with laryngomalacia in other hospitals, and 4 (10.8%) patients were misdiagnosed with pneumonia in other hospitals. One patient required emergency intubation for rescue because of dyspnea before the operation. The root of the tongue was found to occupy space during intubation, and 6 (16.2%) patients were found to have cysts of the root of the tongue through CT examination. Three cases (8%) were found to have tongue root cysts by bronchoscopy due to pneumonia, and the rest were found to have tongue root cysts by laryngoscopy.

A total of 33 cases (89.2%) had laryngeal stridor, 8 cases (21.6%) had dyspnea, 6 cases (16.2%) had inspiratory trisomy, 14 cases (37.8%) had cough and vomiting, 14 cases (37.8%) had cough and stridor at the same time, 13 cases (35.1%) had pneumonia, 10 cases (27.1%) had laryngomalacia, and 9 cases (24.3%) had pneumonia and laryngomalacia at the same time when they came to our hospital.

All children were examined with electronic laryngoscopy (Figure 1-A), 28 patients were examined with neck CT (Figure 1-B), and the median tumor diameter was 10 mm (5-20 mm).

All children received radiofrequency surgery with a self-retaining laryngoscope. During the operation, the cyst was completely removed, and hyoid contouring was performed during the surgery. Eight patients were intubated and transferred to the ICU for treatment after the operation. The median intubation time was 4.5 days (2-5 days). The examination of the laryngoscope after the operation indicated that the white wound membrane was well formed. Histological examination confirmed the diagnosis of a congenital tongue base cyst (Figure 1-C). There was no recurrence after 6 months of follow-up (Figure 1-D).

Discussion:

Congenital tongue base cysts usually occur on the midline between the back of the tongue and the lingual foramen cecum. Most cysts are mucous retention cysts[2]. They are generally deep and covered with a thick layer of mucous membrane. Inside the cyst is serous liquid, and it is usually near and oppresses the epiglottis, which may lead to respiratory obstruction and feeding difficulty. The cysts originate from the endoderm and mesoderm of the embryonic stage. Because of the lack of specificity in clinical manifestations, they are easily misdiagnosed as congenital laryngomalacia and neonatal pneumonia. As a result, children cannot receive timely and effective treatment, resulting in serious consequences. It has been reported in the literature that 50% of congenital tongue base cysts are definitively diagnosed after autopsy[3], indicating that the rates of missed diagnosis and misdiagnosis of this disease are high.

This study shows that 26 children (70.3%) suffered from laryngeal wheezing, dyspnea and feeding difficulties after birth, which indicates that the clinical symptoms in these children occur early. The common clinical manifestations of this disease are laryngeal wheezing, inspiratory dyspnea, choking, and vomiting. These clinical manifestations lack specificity, so it is impossible to make a diagnosis based on these clinical manifestations. The diagnosis must be made through auxiliary examination. Because congenital tongue base cysts grow expansively, squeezing the epiglottis, they may cause dyspnea and dysphagia.

This study shows that electronic laryngoscopy has a high detection rate for congenital tongue base cysts and is the first choice for such children[4]. Electronic laryngoscopy has good flexibility and a wide field of vision and can more clearly demonstrate the scope and characteristics of the lesions. Electronic laryngoscopy also causes less stimulation for children and can detect and diagnose laryngeal diseases earlier, but it also has certain limitations. For example, the size and scope of the tumor cannot be accurately measured, and the anatomical relationship with the surrounding tissues cannot be determined. CT scanning can accurately display the anatomical location of the cyst and the relationship between the cyst and the hyoid bone. It is the most commonly used imaging examination method for diagnosing congenital tongue base cysts. MR examination is not recommended for this disease. Because MR examination takes a long time, it is not suitable for children with serious illness. In addition, MR examination requires sedatives, which may increase the risk of upper airway obstruction. Thyroid cartilage has not been ossified in the neonatal period. Ultrasound has good sound transmission and can show the throat and related areas well. It can identify the location,

boundary, shape, blood flow and other conditions of the lesions. It can identify cystic and solid properties and can exclude ectopic thyroid glands, hemangioma and other related diseases. Ultrasound also has the advantages of high safety, convenient inspection, no radioactivity, and no need for sedation and can be used as an important modality for screening congenital tongue base cysts[5]. However, the results of ultrasonic examination are often affected by the experience of the examiner and the performance of the machine, which may affect the detection rate. Therefore, electronic laryngoscopy, neck ultrasound and neck CT should be combined to make a correct diagnosis, to improve the diagnostic accuracy of congenital tongue base cysts and to reduce the incidence of missed diagnosis or misdiagnosis.

For children with congenital tongue base cysts, surgery should be carried out as soon as possible to remove the occupation and relieve the obstruction. The surgery should remove as much of the cyst wall as possible and contour the hyoid bone to reduce postoperative recurrence. When the child has obvious dyspnea, aspiration of the cysts can temporarily relieve the symptoms, and surgery can be performed when the situation permits. Although pure puncture aspiration can quickly relieve the clinical symptoms, after puncture aspiration, the cyst fluid flows out, and the cyst volume is significantly reduced. As a result, pure puncture aspiration is not conducive to surgery, and the cyst easily recurs after puncture; thus, it is not recommended to puncture the cyst before surgery.

Conclusion:

The symptoms of congenital tongue root cysts in infancy appear early but lack specificity. For children with dyspnea and feeding difficulty, electronic laryngoscopy, ultrasound, and neck CT should be performed in a timely manner to make a clear diagnosis. It is vital for these patients to be diagnosed and treated as soon as possible.

Figure-1

A Electronic laryngoscopy revealed that the cyst was located at the root of the tongue, compressed the epiglottis, and was hemispherical. The glottis was poorly exposed.

B Neck CT examination revealed a round cystic mass at the base of the tongue, with a clear boundary and a diameter of about 16mm

C Pathological examination showed irregular fissure structure under squamous epithelium with inflammatory cell infiltration

D Six months after operation, laryngoscopy showed that the cyst at the base of the tongue was completely removed, and there was no sign of recurrence.

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