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Case report

Stridor caused by duplication cyst in a female infant and temporary vocal cord paralysis. A case report

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ABSTRACT

Introduction and importance: Congenital hypopharynx duplication cysts are a medical rarity; nevertheless, they offer an important differential diagnosis in neonatal dyspnea or feeding problems.
Case report: Herein, we describe a case of delayed diagnosis but successful surgical removal of a large congenital hypopharynx cyst in a 4-month-old infant presenting with stridor.
Clinical discussion: Early and proper diagnosis and surgical handling of hypopharynx cyst can help to achieve the best prognosis and outcome.
Conclusion: We present the successful management of a newborn with a pharyngeal duplication cyst. In our case, the temporary postoperative laryngeal nerve palsy resolved within four weeks. Overstretching of recurrent laryngeal nerve might have caused this complication.

1. Introduction

The first pharyngeal duplication cyst (PDC) was described by Blasius in 1711 [1]. Generally, gastrointestinal duplication cysts are defined by three main characteristics: a cyst attached to the gastrointestinal wall, coverage by two muscle layers and a gastrointestinal mucosa lining [1]. The aim of the present study was to describe the clinical course and management of a rare type of congenital cervical cyst. The current therapeutic standard focuses on surgical removal [5]. Our patient was operated in maxcare private hospital. Embryological conclusions were deduced from the histology.

2. Case report

A 3-month-old Caucasian, female, term infant was transferred by senior pediatric physician to our hospital with an ultrasound diagnosis of a large cyst located in the neck. The family and prenatal history (of intrauterine development) was uneventful. The child was delivered by cesarean section due to labor arrest at the 37th week. Acute neonatal respiratory distress occurred and the baby was ventilated for some hours. Postpartum, the newborn was suspected to have pneumonia. It recovered and was discharged after antibiotic treatment with ampicillin and gentamicin for pneumonia.

The baby presented with slight stridor for a couple of weeks, worsening during feeding and agitation. There was no visible or palpable swelling on the neck. Vital signs, routine laboratory tests and echocardiogram revealed normal findings.

However, cervical ultrasound detected centrally located cystic lesion. The cyst was located dorsally from the larynx and trachea, extending from the level of the hypopharynx down to the cervical part of the esophagus until the upper thymus.

Magnetic resonance imaging (MRI) confirmed simple cystic structure compressing the trachea (Fig. 1-a), measuring 54 × 27 × 24 mm. There was no evidence for communication with the tracheal or esophageal lumen. There were no diagnostic challenges. A diagnosis of duplication cyst was made. The differential diagnoses involved laryngomalacia, tracheomalacia, bronchogenic cyst and vascular malformations.

At the age of 4 months, laryngotracheoscopy and pharyngoesophagoscopy were performed in combination with surgical removal in the same session. Before intubation the upper pole of the cyst

Abbreviations: PDC, pharyngeal duplication cyst; MRI, magnetic resonance imaging; CPAP, continuous positive airway pressure; RLN, recurrent laryngeal nerve; RLNP, recurrent laryngeal nerve palsy.

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was visualized protruding into the oropharynx (Fig. 2). There were no other malformations.

In Rose position we used a transverse incision, dissected through the strap muscles and retracted the trachea to the right. The upper part of the cyst originated from the pharyngeal constrictor muscle and the upper esophagus and continued downwards to the entrance of the middle mediastinum. There was only a narrow tiny fibrous connection to the backside of the laryngotracheal funnel (Fig. 3-a). Invasive recurrent laryngeal nerve (RLN) monitoring was not performed.

The perioperative antibiotic prophylaxis with cefuroxime (50 mg per kg of body weight) was administrated as a single shot. The patient was operated in general anesthesia, standard neonatal equipment was used. The procedure was performed by second author (RBT), the senior consultant of pediatric surgery.

Since the thin-walled cyst shared its wall with the hypopharynx and the upper esophagus, the resection required the opening of both structures (Fig. 3-b). After complete resection of the epithelium-lined cyst, we reconstructed both tubular structures with a double-layer polyglactin 910 (Coated Vicryl 5/0, Ethicon ®) suture. The patient tolerated the procedure well and remained hemodynamically stable. The surgery lasted about 3 h, a revision was not indicated. No relevant blood loss was observed.

The surgical pathology report revealed an epithelial lining consisting of respiratory and squamous epithel surrounded by both smooth and striated muscle fibers (Fig. 4-a, b).

The postoperative course was prolonged by failed attempts to extubate, but the baby was finally weaned after 8 days. Noninvasive continuous positive airway pressure (CPAP) support was continued over a week. The baby was started on oral feeds through a nasogastric tube until full oral feeding. At the follow-up laryngoscopy on the 3rd day, we detected a suboptimal mobility of right vocal cord, suggestive for recurrent laryngeal nerve palsy (RLNP), grade II by Clavien-Dindo classification [18]. However, repeated flexible laryngoscopy after 4 weeks revealed normal movements and positions of the vocal cords. In addition, there were no further respiratory problems or any complications. Normal sucking and swallowing occurred, and the baby showed satisfactory weight gain. Follow-up MRI after 3 months demonstrated nearly normal surrounding structures (Fig. 1-b). Since we removed the complete cyst including its lining epithelium, recurrence is not expected.

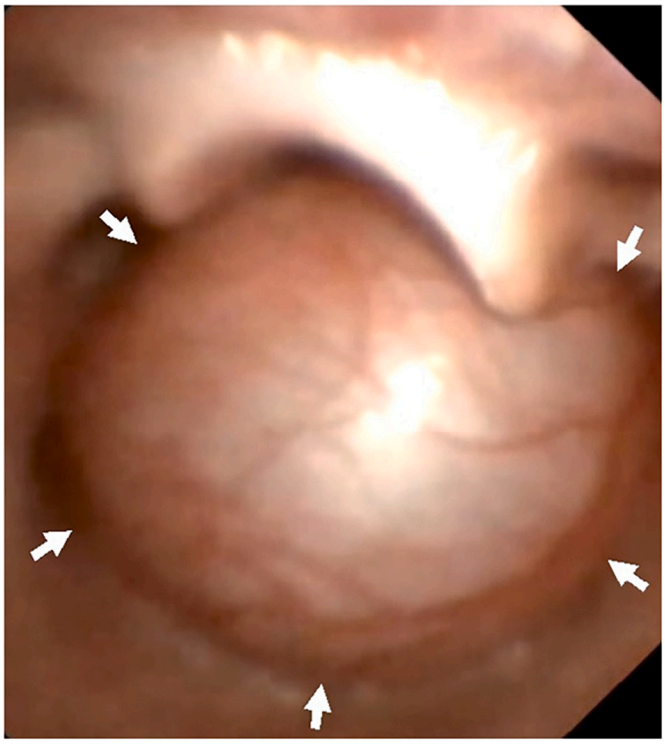

Fig. 2. Pharyngoscopic view of the pharyngeal protrusion from above (flexible pharyngoscopy). Arrows indicate the boundaries of the cyst.

No special follow-up was planned in the future, unless new symptoms appear.

All our work has been reported in line with the SCARE criteria and guidelines [17].

3. Discussion

The only curative therapy for cystic foregut malformations of the

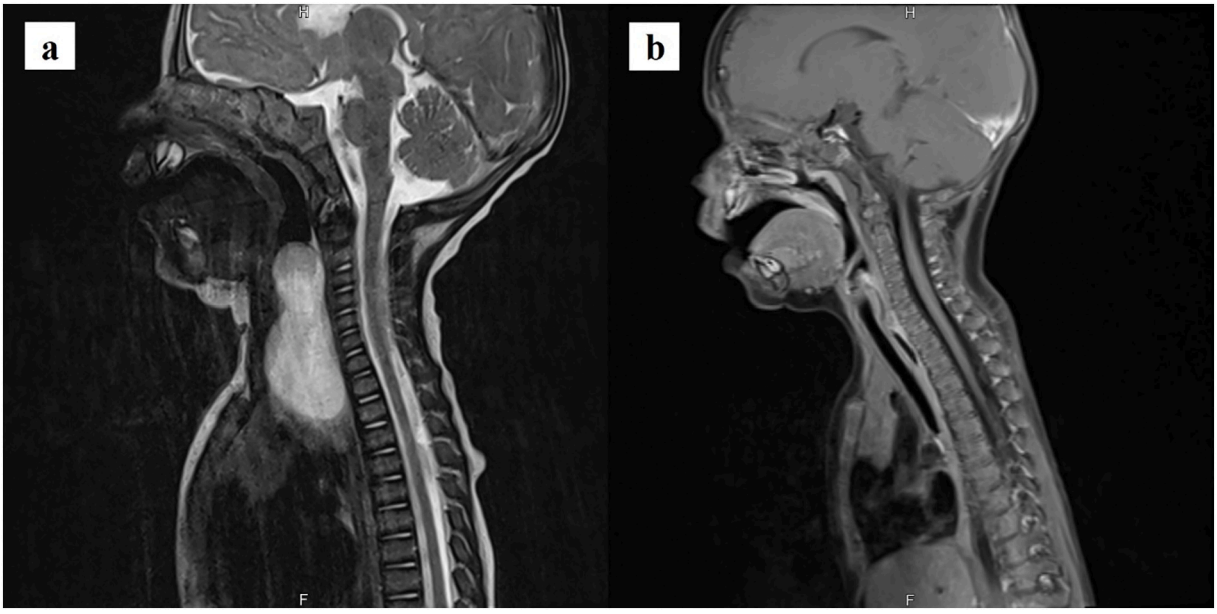

Fig. 1. MRI imaging of the neck. (a) Retrotracheal cystic mass between the esophagus and trachea. The upper pole protruded into the hypopharynx and was portrayed during pharyngoscopy (Fig. 2). The lower pole had contact with the upper mediastinum and was exposed during surgery (Fig. 3-a). (b) Follow-up MRI 3 months after the surgery.

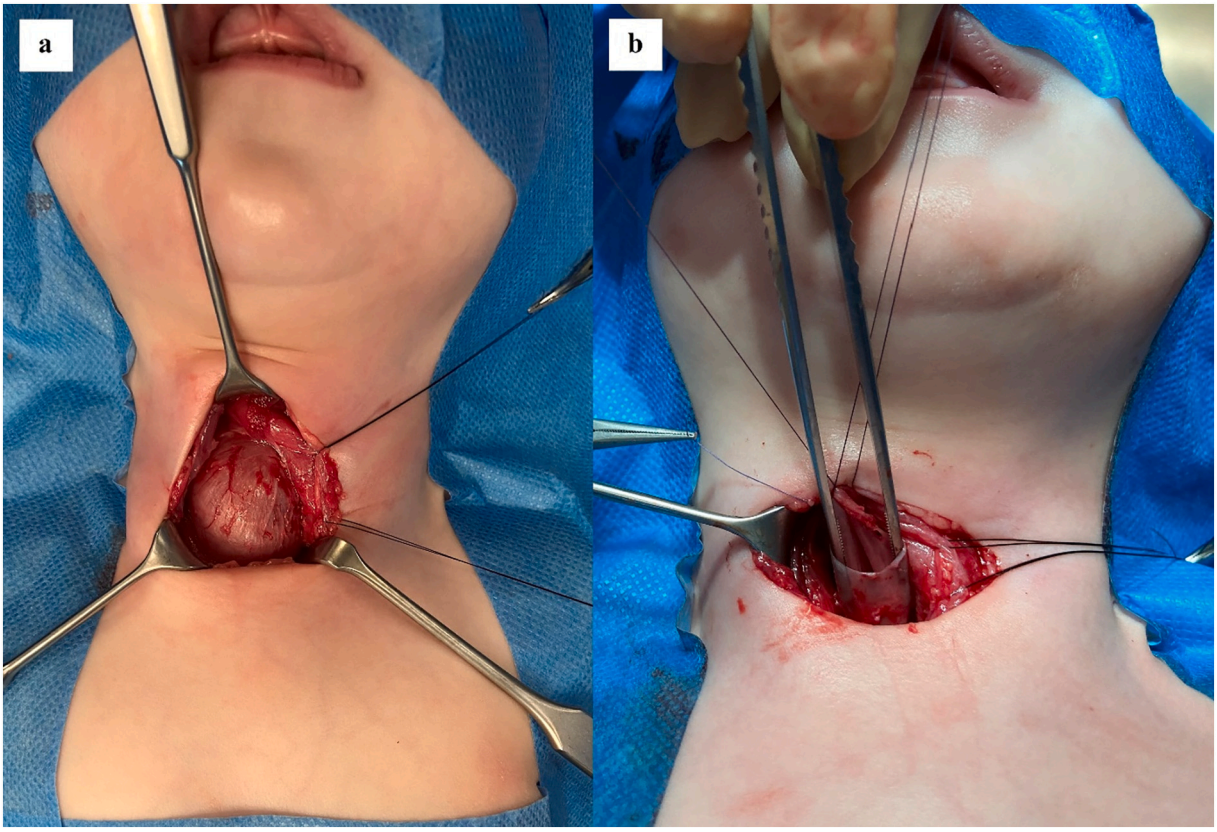

Fig. 3. Operative findings after ventral exposure. (a) The lower part of the cyst protruding into the upper part of the mediastinum. (b) The laryngoesophageal junction, upper part of the esophagus after removal of the cyst.

pharynx and upper esophagus is surgical removal. As we have reported, a common wall of the cyst and the upper gastrointestinal tract must be expected.

We identified twelve pediatric cases described in the Anglo-American literature to date [5–16]. The leading symptoms of hypopharyngeal duplication cysts are respiratory disorders such as stridor, aspiration, dysphagia and regurgitation. Infection may occur, which can lead to life-threatening complications. Hence, the differential diagnosis of stridor should include laryngo- and tracheomalacia, vascular malformations, subglottic tumors and infectious diseases. The most helpful tools in diagnosis are ultrasound and magnetic resonance imaging.

The development of PDC is far from clear. Several theories have been proposed, including the most popular split notochord theory by Veeneklass [2], the split notochord theory by Bentley and Smith [3], the bronchopulmonary foregut theory and the aberrant luminal recanalization theory [4]. The histological findings of this case report favor the bronchopulmonary foregut theory. This theory suggests the development of an additional budding of the foregut, which develops into the tracheobronchial tree anteriorly and the esophagus posteriorly. The epithelial lining found in the removed cyst indicates its origin during the budding phase of the trachea from the foregut. In addition, the muscular wall consisted of striated and smooth muscles, each occurring in the tracheal and upper esophageal wall.

The surgical options for PDC are marsupialization, CO_2 laser ablation and resection. We assume that marsupialization may lead to a high rate of recurrences by forming a blind sac. According to literature, complete surgical resection provides the best chance to remove the whole afflicted tissue [5].

Complications that appear rarely include regurgitation, excessive scarring, dysphagia and stridor or transient recurrent laryngeal nerve palsy (RLNP). To improve safety, we recommend invasive intraoperative RLN monitoring.

4. Conclusion

We present the successful management of a newborn with a PDC. In our case, temporary RNLP resolved within four weeks. Overstretching of both RLNs might have caused this complication.

Funding

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Ethical approval

N/A.

Consent

Written informed consent was obtained from the patient's parents for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

T. Baranski was the major contributor to the writing of this manuscript. R.-B. Tröbs performed the critical analysis, examination and scientific validation. T. Baranski, R.-B. Tröbs analyzed and performed the literature research. W. Piroth, P. Seiffert and M. William kindly provided their revised diagnostics for this work. All authors have approved the manuscript.

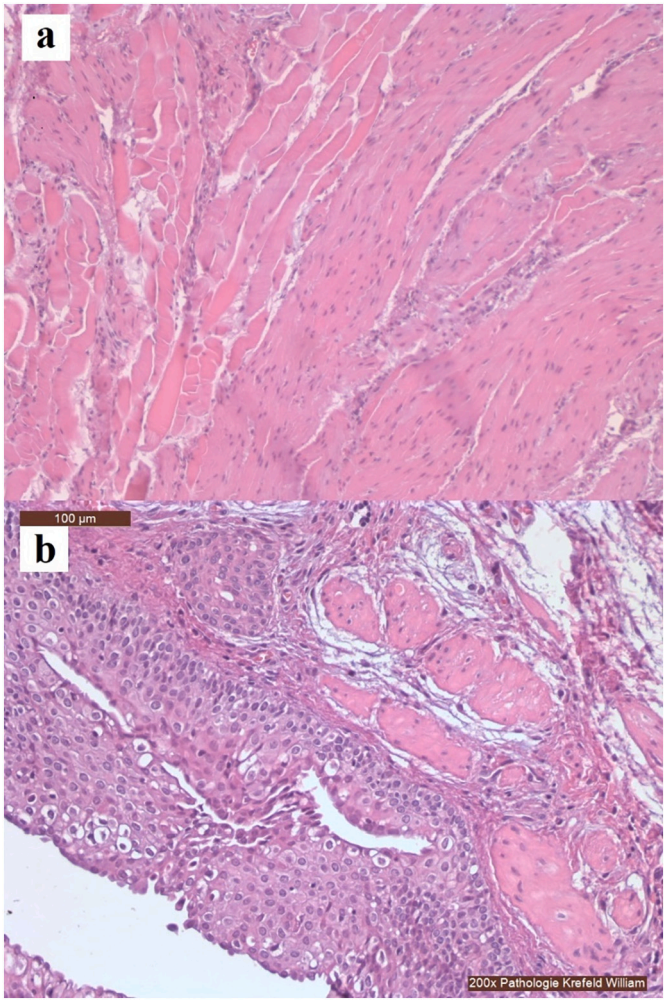

Fig. 4. Histologic appearance of the cyst wall (a, ×100; b, ×200). (a) The muscle layer is composed of striated and smooth muscle. (b) The mucosal layer consists of stratified, nonkeratinized epithelium.

Registration of research studies

N/A.

Guarantor

Tomasz Baranski, Ralf-Bodo Troebs.

Declaration of competing interest

Nothing to declare

References

[1] J.L. Arbona, J.G. Fazzi, J. Mayoral, Congenital esophageal cysts: case report and review of literature, Am. J. Gastroenterol. 79 (1984) 177–182. PMID: 6702802.
[2] G.M. Veeneklaas, Pathogenesis of intrathoracic gastrogenic cysts, AMA Am. J. Dis. Child. 83 (4) (1952 Apr) 500–507, https://doi.org/10.1001/archpedi.1952.02040080096010.
[3] J.F. Bentley, J.R. Smith, Developmental posterior enteric remnants and spinal malformations: the split notochord syndrome, Arch. Dis. Child 35 (179) (1960 Feb) 76–86, https://doi.org/10.1136/adc.35.179.76.
[4] R.I. Macpherson, Gastrointestinal tract duplications: clinical, pathologic, etiology and radiologic considerations, RadioGraphics 13 (1993) 1063–1080, https://doi.org/10.1148/radiographics.13.5.8210590.
[5] M.E. Ahmed, M.E. Ahmed, A.M. El Batawi, H.M. Abdelfattah, N. Jelassi, Internal hypopharyngeal cyst: a review of literature, Dysphagia 34 (4)) (2019 Aug) 487–498, https://doi.org/10.1007/s00455-019-10003-2.
[6] J.K. Kim, K.K. Park, Foregut duplication cyst of the hypopharynx: a rare cause of upper airway obstruction, J. Pediatr. Surg. 42 (6) (2007 Jun) E5–E7, https://doi.org/10.1016/j.jpeds urg.2006.11.007.
[7] M. Roy, V. Rupa, A.N. Deepthi, J. Chacko, Unusual cystic hypopharyngeal mass in a child with obstructive symptoms, Indian J. Otolaryngol. Head Neck Surg. 63 (Suppl. 1) (2011 Jul) 41–43, https://doi.org/10.1007/s12070-011-0184-z.
[8] J. Edwards, S. Pearson, G. Zalzal, Foregut duplication cyst of the hypopharynx, Arch. Otolaryngol. Head Neck Surg. 131 (12) (2005 Dec) 1112–1115, https://doi.org/10.1001/archotol.131.12.1112.
[9] J.M. Hsu, A.J. Mortelliti, Gastric choristoma of the hypopharynx presenting in an infant: a case report and review of the literature, Int. J. Pediatr. Otorhinolaryngol. 56 (1) (2000 Nov 30) 53–58, https://doi.org/10.1016/S0165-5876(00)00378-5.
[10] C. Johnston, B. Benjamin, H. Harrison, A. Kan, Gastric heterotopia causing airway obstruction, Int. J. Pediatr. Otorhinolaryngol. 18 (1) (1989 Sep) 67–72, https://doi.org/10.1016/0165-5876(89)90233-4.
[11] E.J. Picard, J.J. Picard, J. Jorissen, M. Jardon, Heterotopic gastric mucosa in the epiglottis and rectum, Am. J. Dig. Dis. 23 (3) (1978 Mar) 217–221, https://doi.org/10.1007/BF01072320.
[12] K.M. Van Abel, M.L. Carlson, S. Narendra, S.A. Cofer, D.M. Thompson, Hypopharyngeal gastric choristoma in an infant resulting in airway obstruction, Arch. Otolaryngol. Head Neck Surg. 137 (9) (2011 Sep) 947–951, https://doi.org/10.1001/archoto.2011.138.
[13] J. Steineger, K. Brøndbo, A congenital malformation of the piriform sinus mimicking a laryngeal cyst.A case report, Otolaryngology S4 (2012) 001, https://doi.org/10.4172/2161-119x.s4-001.
[14] P. Daher, E. Riachy, S. Zeidan, A. Saad, Upper airway obstructive symptoms because of ectopic gastric mucosa in a newborn: a case report, J. Pediatr. Surg. 41 (8) (2006 Aug) e7–e9, https://doi.org/10.1016/j.jpedsurg.2006.04.005.
[15] K.B. Hüttenbrink, W. Stoll, Stridor im Neugeborenenalter bei heterotoper Magenschleimhaut im Hypopharynx [Stridor in the neonatal period in heterotopic gastric mucosa of the hypopharynx], Laryngol. Rhinol. Otol. (Stuttg.) 66 (2) (1987 Feb) 67–69. PMID: 3573892.
[16] S. Frenkiel, K.A. Remsen, Foregut duplication arising from the pharynx, 279-8, J. Otolaryngol. 19 (4) (1990 Aug), https://doi.org/10.1001/archoto.2010.127.
[17] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A. Fowler, D.P. Orgill, For the SCARE Group, The SCARE 2018 statement: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 60 (2018) 132–136.
[18] D. Dindo, N. Demartines, P.A. Clavien, Classification of surgical complications: a new proposal with evaluation in a cohort of 6336 patients and results of a survey, Ann. Surg. 240 (2)) (2004 Aug) 205–213, https://doi.org/10.1097/01.sla.0000133083.54934.ae. PMID: 15273542; PMCID: PMC1360123.