REVIEW
3621  Autoimmunity as the comet tail of COVID-19 pandemic
   Talotta R, Robertson E

3645  Gender medicine: Lessons from COVID-19 and other medical conditions for designing health policy
   Machluf Y, Chaiter Y, Tal O

MINIREVIEWS
3669  Complexities of diagnosis and management of COVID-19 in autoimmune diseases: Potential benefits and
detrimentsof immunosuppression
   Georgiev T, Angelov AK

ORIGINAL ARTICLE
Retrospective Study
   D'Urbano F, Fabbri N, Koleva Radica M, Rossin E, Carcoforo P

3697  Somatostatin receptor scintigraphy in the follow up of neuroendocrine neoplasms of appendix
   Saponjski J, Macut D, Sobic-Saranovic D, Ognjanovic S, Bozic Antic I, Pavlovic D, Artiko V

3708  Efficacy of stool multiplex polymerase chain reaction assay in adult patients with acute infectious diarrhea

3718  Comparison of gemcitabine plus nab-paclitaxel and FOLFIRINOX in metastatic pancreatic cancer
   Han SY, Kim DU, Seol YM, Kim S, Lee NK, Hong SB, Seo HI

3730  Shear wave elastography may be sensitive and more precise than transient elastography in predicting
   significant fibrosis
   Yao TT, Pan J, Qian JD, Cheng H, Wang Y, Wang GQ

3743  Radioactive 125I seed implantation for locally advanced pancreatic cancer: A retrospective analysis of 50 cases
   Li CG, Zhou ZP, Jia YZ, Tan XL, Song YY

3751  Active surveillance in metastatic pancreatic neuroendocrine tumors: A 20-year single-institutional experience
   Gao HL, Wang WQ, Xu HX, Wu CT, Li H, Ni QX, Yu XJ, Liu L

3763  Clinical efficacy of tocilizumab treatment in severe and critical COVID-19 patients
   Zeng J, Xie MH, Yang J, Chao SW, Xu EL
## Contents

### Phosphatidylinositol-3,4,5-trisphosphate dependent Rac exchange factor 1 is a diagnostic and prognostic biomarker for hepatocellular carcinoma

*Cai Y, Zheng Q, Yao DJ*

### Observational Study

#### Awareness and attitude of fecal microbiota transplantation through transendoscopic enteral tubing among inflammatory bowel disease patients


### CASE REPORT

#### Cauda equina arachnoiditis – a rare manifestation of West Nile virus neuroinvasive disease: A case report


### REVIEW

#### Portal gas in neonates; is it always surgical? A case report

*Altokhais TI*

### CASE REPORT

#### Large lingual heterotopic gastrointestinal cyst in a newborn: A case report

*Lee AD, Harada K, Tanaka S, Yokota Y, Mima T, Enomoto A, Kogo M*

#### Osteochondral lesion of talus with gout tophi deposition: A case report

*Kim T, Choi YR*

#### Traumatic neuroma of remnant cystic duct mimicking duodenal subepithelial tumor: A case report


#### Autoimmune hepatitis in a patient with immunoglobulin A nephropathy: A case report

*Jeon YH, Kim DW, Lee SJ, Park YJ, Kim HJ, Han M, Kim IY, Lee DW, Song SH, Lee SB, Seong EY*

#### Diagnosis of an actively bleeding brachial artery hematoma by contrast-enhanced ultrasound: A case report

*Ma JJ, Zhang B*

#### Lung adenocarcinoma harboring rare epidermal growth factor receptor L858R and V834L mutations treated with icotinib: A case report

*Zhai SS, Yu H, Gu TT, Li YX, Lei Y, Zhang HY, Zhen TH, Gao YG*

#### Gastroduodenitis associated with ulcerative colitis: A case report

*Yang Y, Li CQ, Chen WJ, Ma ZH, Liu G*

#### Majocchi's granuloma caused by *Trichophyton rubrum* after facial injection with hyaluronic acid: A case report

*Liu J, Xin WQ, Liu LT, Chen CF, Wu L, Hu XP*
## Contents

**World Journal of Clinical Cases**  
Semimonthly Volume 8 Number 17 September 6, 2020

<table>
<thead>
<tr>
<th>Page</th>
<th>Title</th>
<th>Authors</th>
</tr>
</thead>
<tbody>
<tr>
<td>3881</td>
<td>Gallbladder sarcomatoid carcinoma: Seven case reports</td>
<td>Qin Q, Liu M, Wang X</td>
</tr>
<tr>
<td>3890</td>
<td>Surgical strategy used in multilevel cervical disc replacement and cervical hybrid surgery: Four case reports</td>
<td>Wang XF, Meng Y, Liu H, Hong Y, Wang BY</td>
</tr>
<tr>
<td>3911</td>
<td>Diagnosis and treatment of mixed infection of hepatic cystic and alveolar echinococcosis: Four case reports</td>
<td>A JD, Chai JP, Wang H, Gao W, Peng Z, Zhao SY, A XR</td>
</tr>
</tbody>
</table>
ABOUT COVER
Editorial board member of World Journal of Clinical Cases, Dr. Elia de Maria is Adjunct Professor of Arrhythmology Lab in the Cardiology Unit, Ramazzini Hospital in Carpi, Italy. He graduated in Medicine and Surgery from the University of Napoli in 1999, continuing on to obtain specialization in Cardiology in 2003. He also holds the distinction of High Degree Master in Electrophysiology and Cardiac Stimulation. Since 2005, he has practiced as a Permanent Consultant Cardiologist in the Italian Public Hospitals, and since 2015 as an External Contract Professor in the Faculty of Medicine and Surgery of University of Verona. His clinical and research interests encompass pharmacological therapy in acute and chronic cardiac conditions, temporary and definitive pacing, thoracentesis and pericardiocentesis, and hemodynamic monitoring. (L-Editor: Filipodia)

AIMS AND SCOPE
The primary aim of World Journal of Clinical Cases (WJCC, World J Clin Cases) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

INDEXING/ABSTRACTING
The WJCC is now indexed in Science Citation Index Expanded (also known as SciSearch®), Journal Citation Reports/Science Edition, PubMed, and PubMed Central. The 2020 Edition of Journal Citation Reports® cites the 2019 impact factor (IF) for WJCC as 1.013; IF without journal self cites: 0.991; Ranking: 120 among 165 journals in medicine, general and internal; and Quartile category: Q3.
Large lingual heterotopic gastrointestinal cyst in a newborn: A case report

Atsushi-Doksa Lee, Kazuma Harada, Susumu Tanaka, Yusuke Yokota, Takashi Mima, Akifumi Enomoto, Mikihiko Kogo

**ORCID number:** Atsushi-Doksa Lee 0000-0002-4347-8106; Kazuma Harada 0000-0003-1695-0687; Susumu Tanaka 0000-0002-0306-3627; Yusuke Yokota 0000-0001-9282-6544; Takashi Mima 0000-0001-8329-1592; Akifumi Enomoto 0000-0001-5804-8283; Mikihiko Kogo 0000-0002-4556-2491.

**Author contributions:** Lee AD, Harada K, Tanaka S and Kogo M were the patient’s surgeons, reviewed the literature and contributed to manuscript drafting; Yokota Y, Mima T and Enomoto A reviewed the literature and contributed to manuscript drafting; all authors issued final approval for the version to be submitted.

**Informed consent statement:** Informed written consent was obtained from the patient for publication of this report and any accompanying images.

**Conflict-of-interest statement:** The authors declare that they have no conflict of interest.

**CARE Checklist (2016) statement:** The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

**Open-Access:** This article is an

**Abstract**

**BACKGROUND**

Heterotopic gastrointestinal cysts have gastrointestinal epithelium in the cyst wall and rarely occur in the oral cavity. Most are found in the neonatal period. However, heterotopic gastrointestinal cysts that are diagnosed as a congenital tongue cyst by routine ultrasonography are extremely rare.

**CASE SUMMARY**

A 12-day-old female presented with swelling of the anterior tongue. The obstetrician had detected significant tongue swelling on fetal ultrasonography in the 35th gestational week. The female was born by cesarean delivery at gestational week 39. She soon became dyspneic, and the cyst was aspirated. After the aspiration, her breathing recovered and she started breastfeeding. The cyst was excised under general anesthesia on the 67th day. Histopathologic examination showed that that cyst wall consisted of a lining of columnar gastrointestinal-type epithelium and pseudostratified ciliated epithelium. The patient restarted breastfeeding 3 h after surgery. The postoperative course was uneventful.

**CONCLUSION**

Airway distress and feeding difficulty were successfully avoided by cyst aspiration, and surgical resection was performed with no perioperative complications.
Key words: Tongue cyst; Congenital cyst; Prenatal diagnosis; Airway management; Gastrointestinal epithelium; Case report

©The Author(s) 2020. Published by Baishideng Publishing Group Inc. All rights reserved.

Core tip: Heterotopic gastrointestinal cysts have gastrointestinal epithelium in the cyst wall and rarely occur in the oral cavity. When these cysts do occur in the oral cavity, most are found in the neonatal period. However, heterotopic gastrointestinal cysts that are diagnosed as a congenital tongue cyst by routine ultrasonography are extremely rare. In such cases, airway management immediately after delivery is particularly important, and intratracheal intubation or reduction procedures may be required. We successfully avoided airway distress and feeding difficulty by performing cyst aspiration. Cyst aspiration may improve the outcome in such rare cases.

URL: https://www.wjgnet.com/2307-8960/full/v8/i17/3808.htm
DOI: https://dx.doi.org/10.12998/wjcc.v8.i17.3808

INTRODUCTION
Heterotopic gastrointestinal cysts have gastrointestinal epithelium in the cyst wall and rarely occur in the oral cavity[1]. When heterotopic gastrointestinal cysts do occur in the oral cavity, they frequently occur in the anterior part of the tongue and are mainly found in the neonatal period; however, some cases become asymptomatic in adulthood[2,3]. The diagnosis of a heterotopic gastrointestinal cyst as a congenital tongue cyst by routine ultrasonography is extremely rare[4-10]. Congenital tongue cysts can cause respiratory distress and require multidisciplinary treatment for airway management. Intratracheal intubation and reduction procedures may be required immediately after delivery[5-7,9]. The treatment is complete surgical resection, and there are very few cases of recurrence[8]. However, there is no consensus on the optimal timing of surgery and the effectiveness of preoperative cyst reduction procedures. Herein, we report a rare case of a congenital heterotopic gastrointestinal cyst of the tongue. Cyst aspiration successfully avoided airway distress and feeding difficulty.

CASE PRESENTATION

Chief complaints
A 12-day-old female was referred to our hospital for tongue swelling.

History of present illness
The obstetrician had detected significant swelling of the tongue on fetal ultrasonography performed in the 35th gestational week. Because of the anticipated high risk of breathing difficulty, the female was born by cesarean delivery at gestational week 39 in the presence of a pediatric surgeon.

History of past illness
The medical and obstetric history of the mother was unremarkable, with no family history of any congenital anomalies. The patient had no other congenital anomalies.

Physical examination
The cystic legion was located hypoglossally and measured 15 mm × 25 mm. The cyst elevated the tongue and occupied the oral cavity, preventing the mouth from closing (Figure 1A). She became dyspneic soon after delivery, and so the pediatric surgeon aspirated the cyst (Figure 1B).
Figure 1 Neonatal appearance at birth. A: The tongue cyst occupies the oral cavity before needle aspiration; B: Tongue cyst aspiration with an 18-gauge indwelling needle.

Laboratory examinations
Histopathologic examination showed that the specimen was a true cyst, and that its wall consisted of a lining of columnar gastrointestinal-type epithelium and pseudostratified ciliated epithelium (Figure 2).

Imaging examinations
Magnetic resonance imaging (MRI) performed on the 24th day showed that the 25 mm circular-shaped cyst was located hypoglossally without expansion in the oral floor (Figure 3).

FINAL DIAGNOSIS
The final diagnosis was a heterotopic gastrointestinal cyst of the tongue.

TREATMENT
To avoid the repetition of aspiration, immediate surgical resection was recommended. However, the mother of the patient did not want her to undergo surgery in the neonatal period. Thus, a second aspiration was performed on the 7th day. However, the cyst was found to be gradually increasing in size, the excision procedure was performed with the patient under general anesthesia on the 67th day. The tongue was pulled forward and an incision was made on the anterior midline under the tongue. The submucosal tissue was bluntly dissected until the cyst wall was observed (Figure 4A). The cyst was excised by dissecting the connective tissue (Figure 4B). The cyst contained viscous dark brown fluid. The wound was sutured with absorbable thread.

OUTCOME AND FOLLOW-UP
The patient was able to breastfeed 3 h after surgery. The postoperative course was uneventful, and there has been no recurrence during 5 years of follow-up.

DISCUSSION
The reported congenital cystic lesions of the tongue include a ranula[11], teratoma[12], thyroglossal duct cyst[13], lymphangioma[14], and heterotopic gastrointestinal cyst. However, heterotopic gastrointestinal cyst of the tongue is rare[1]. Although this type of cyst is most commonly referred to as a heterotopic gastrointestinal cyst, it is also known by various terms such as a foregut duplication cyst, choristomatic cyst, ciliated epithelial cyst, lingual duplication cyst, cystic tumor of the tongue, unusual thyroglossal duct cyst, heterotopic large bowel cyst, enterocystoma, and alimentary...
Figure 2 Pathological findings of the cyst. A: Gross anatomy of the excised cyst. The cyst wall is elastic and soft, and the lumen is smooth; B: Hematoxylin-eosin stained specimen showing columnar gastrointestinal epithelium lining the cyst wall (× 400 original magnification); C: Ciliated pseudostratified epithelium is also seen in another part (× 400 original magnification).

Figure 3 T2-weighted magnetic resonance imaging shows that the cyst is located at the ventral part of the anterior tongue. A: Horizontal view; B: Sagittal view.

It is rare to see heterotopic gastrointestinal epithelium in the oral region, while it is more commonly reported in the duodenum, gallbladder, common bile duct, jejunum, Merkel diverticulum, ileum, appendix, colon, and rectum[3]. The wall of a heterotopic gastrointestinal cyst may consist of a lining of ciliated epithelium and squamous epithelium in addition to gastrointestinal epithelium[3]. The pathogenesis of a heterotopic gastrointestinal cyst is still unknown, but the cyst may arise from undifferentiated ectoderm lining of the primitive stomodeum that becomes entrapped within the oral cavity during the 4th week of fetal development[16]. Most heterotopic gastrointestinal cysts are discovered by 2 years of age[3]. The treatment is surgical resection. Possible surgical complications are Wharton’s duct injury and lingual muscle injury. Preoperative MRI is helpful to show the expansion to adjacent structures and the localization of the cyst. Recurrence is rare with complete resection[1].

Cases in which a heterotopic gastrointestinal cyst is detected in the prenatal period are extremely rare. To the best of our knowledge, there are only nine cases in which a lingual heterotopic gastrointestinal cyst has been diagnosed prenatally (Table 1)[4-10]. In
Table 1 Cases of heterotopic gastrointestinal cyst detected in the prenatal period

<table>
<thead>
<tr>
<th>Case</th>
<th>Ref.</th>
<th>Preoperative cyst size</th>
<th>Preoperative intratracheal intubation</th>
<th>Preoperative reduction procedure</th>
<th>Age at the time of surgery</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Chen et al. [6] (1997)</td>
<td>50 mm × 40 mm × 30 mm</td>
<td>Performed</td>
<td>Aspiration</td>
<td>0 d</td>
</tr>
<tr>
<td>2</td>
<td>Chen et al. [6] (1997)</td>
<td>25 mm × 20 mm × 15 mm</td>
<td>Performed</td>
<td>Aspiration and marsupialization</td>
<td>9 mo</td>
</tr>
<tr>
<td>3</td>
<td>Rousseau et al. [9] (2004)</td>
<td>4 cm diameter</td>
<td>Not performed</td>
<td>Aspiration</td>
<td>13 d</td>
</tr>
<tr>
<td>4</td>
<td>Hall et al. [8] (2005)</td>
<td>21 mm × 28 mm × 25 mm</td>
<td>Not performed</td>
<td>None</td>
<td>13 d</td>
</tr>
<tr>
<td>5</td>
<td>Karam et al. [4] (2007)</td>
<td>12 mm × 16 mm</td>
<td>Not performed</td>
<td>None</td>
<td>3 d</td>
</tr>
<tr>
<td>6</td>
<td>Hambarde et al. [5] (2011)</td>
<td>40 mm × 40 mm × 22 mm</td>
<td>Not performed</td>
<td>Marsupialization</td>
<td>2.5 mo</td>
</tr>
<tr>
<td>7</td>
<td>Houshmand et al. [10] (2011)</td>
<td>32 mm × 26 mm × 28 mm</td>
<td>Not performed</td>
<td>None</td>
<td>6 d</td>
</tr>
<tr>
<td>8</td>
<td>Houshmand et al. [10] (2011)</td>
<td>33 mm × 25 mm × 20 mm</td>
<td>Not performed</td>
<td>None</td>
<td>2 mo</td>
</tr>
<tr>
<td>9</td>
<td>Gantwerker et al. [7] (2014)</td>
<td>37 mm × 25 mm × 16 mm</td>
<td>Performed</td>
<td>None</td>
<td>6 d</td>
</tr>
<tr>
<td>10</td>
<td>Present case</td>
<td>15 mm × 25 mm</td>
<td>Not performed</td>
<td>Aspiration</td>
<td>67 d</td>
</tr>
</tbody>
</table>

Figure 4 Intraoperative findings. A: A smooth cyst wall is observed after dissecting the tongue mucosa; B: Loose connective tissue is attached to the cyst.

all of these cases, a lingual cyst was detected with ultrasonography. Large cysts on the tongue may cause airway obstruction. Therefore, prenatal ultrasonographic diagnosis is essential to predict the risk of upper airway obstruction at delivery. Multidisciplinary collaboration is also essential to formulate a careful delivery plan that ensures airway protection in the neonate[7]. Prenatal MRI is a useful way to evaluate the risk of airway obstruction[7-10]. If an airway problem is detected with echo, an ex-utero intrapartum treatment procedure may provide time to open the infant’s airway while maintaining the placental blood supply[8].

In previous reports of lingual heterotopic gastrointestinal cysts, intubation was required for airway management in three of nine cases[6,7]. The remaining seven patients were not intubated, as ultrasonographic imaging showed that there were no airway problems[4,6,10]. Preoperative aspiration or marsupialization was performed in four cases, while aspiration alone was performed in two cases[4,10]. All aspiration procedures were performed at birth. Our case was the only case in which aspiration was performed twice. The timing of cystectomy was within 2 wk in six cases[6-10]. Around two months in three cases (including the present case)[5,10], and at nine months in one case[8]. In all cases, there were no problems with the postoperative respiratory and nutritional statuses. Although previous reports did not describe the preoperative feeding status and intubation at the time of general anesthesia induction, the infant in our case was able to breastfeed just before the surgery, and oral intubation was easy despite the remaining swelling of the tongue.

In our case, immediate cyst aspiration was performed at birth, and the respiratory
condition and feeding function were stable; therefore, surgery was postponed. There is no consensus on the optimal timing of surgical removal of congenital tongue cysts, including heterotopic gastrointestinal cysts. Kaneko et al. suggested that surgical resection of a congenital mucous cyst of the tongue is safely performed at 1 year of age, although the timing of surgery depends on the degree of breathing and feeding impairments, and the child’s development. However, all reported cases of prenatally diagnosed heterotopic gastrointestinal cysts, including our case, involved cyst excision within 1 year and had a good outcome. Thus, aspiration is effective for initial airway and feeding management, but frequent aspiration should be avoided because of the risk of infection and adjacent tissue damage. If the cyst grows and repeated aspiration is difficult, it may be better to perform excision at that time.

CONCLUSION

We described the surgical treatment of a lingual heterotopic gastrointestinal cyst in a newborn. Airway distress and feeding difficulty were successfully avoided by cyst aspiration, and surgical resection was performed on the 67th day with no perioperative complications.

REFERENCES
