Pyriform Sinus Fistula Presenting as Fetal Cervical Cyst

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Abstract

Background: Pyriform sinus fistula, congenital anomaly originating from the third or fourth brachial pouch remnant, rarely occurs during the prenatal period. Case Report: A pregnant woman was referred to our hospital at 30 weeks of gestation with ultrasonographic finding of fetal cervical cyst. Fetal ultrasonography had revealed a left unilocular cystic lesion in the neck. Although it remained unchanged in size, deviation of the trachea by the cyst was later demonstrated by magnetic resonance imaging. A male child was born with a good Apgar score by elective cesarean delivery. Mild respiratory dysfunction was resolved by immediate cyst aspiration. Our diagnosis was pyriform sinus fistula with subcutaneous cyst formation based on catheterization performed under laryngoscopy. Our hospital pediatric surgeons removed the cyst and fistula, and the postoperative course was uneventful. Conclusion: The possibility of pyriform sinus fistula must be kept in mind for fetal cystic lesions of the neck.

Keywords: Cysts, Pregnancy, Pyriform Sinus, Trachea, Ultrasonography.
Discussion

Most cases of pyriform sinus fistula are detected in childhood or older years, and rarely during the prenatal period. Kakogawa et al. [3] reported three cases of prenatal detection of pyriform sinus fistula, each of which resulted in a cystic lesion in the fetal neck. A pyriform sinus fistula in addition to congenital goiter, solid thyroid tumor, thyroid cyst, brachial cleft cyst or laryngocele should be considered for differential diagnosis of a fetal cervical cystic lesion [4].

Although a pyriform sinus fistula often causes infectious events including thyroiditis in childhood or older patients [5], those detected during the antenatal period will have cyst formation in an area near the trachea, which can cause airway obstruction. Although there are no reports of complete airway obstruction in related cases, Kakogawa et al. [3] introduced EXIT in their report. Deviation of the trachea in the present case was not so severe and we anticipated that decompression could be attained by immediate aspiration of the cyst, thus EXIT was not thought to be suitable. However, the cooperation of pediatric surgeons was needed during the elective cesarean delivery procedure, as the newborn showed mild respiratory dysfunction, which resolved by
immediate aspiration of the cyst by the pediatric surgery team.

Fetal intervention can be considered for avoidance of cesarean delivery with or without utilizing EXIT. However, since the cyst can easily refill through the fistula, as observed in our case, the interval between intervention and delivery may be important, and further evaluation is warranted.

Conclusion

A cystic lesion detected in the neck of a fetus was diagnosed as pyriform sinus fistula after birth. Immediate aspiration of the cyst was highly effective for neonatal respiratory distress due to tracheal deviation.

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