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Prenatal Diagnosis of a Large Oropharyngeal Teratoma and Airway Management with EXIT: A Case Report

Case Report

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Abstract

We present a rare case of a large fetal oropharyngeal teratoma (epignathus) diagnosed during the third trimester and managed successfully with a planned ex utero intrapartum treatment (EXIT) procedure followed by neonatal surgical resection. A 30-year-old pregnant woman was referred to our department at 29 weeks of gestation due to polyhydramnios and the detection of an oropharyngeal mass on ultrasound. Fetal magnetic resonance imaging confirmed a 5×5 cm heterogeneous mass filling the oral cavity, raising concern for airway obstruction at birth. At 32 weeks, spontaneous pre-term labor necessitated urgent EXIT. While fetoplacental circulation was maintained, a tracheostomy was performed to secure the airway, allowing for safe delivery and ventilation of the neonate. The newborn subsequently underwent successful transoral surgical excision of the mass, which was confirmed histologically as an immature teratoma. Postoperative recovery was uneventful, and the infant remained free of recurrence during a 24-month follow-up period. This case highlights the importance of prenatal diagnosis, fetal imaging, and multidisciplinary planning in the management of airway-compromising lesions. It also introduces the EXIT procedure to otolaryngologists as a critical and effective approach for ensuring airway patency in selected high-risk cases of congenital head and neck tumors.

Keywords: Teratoma, oropharyngeal neoplasms, airway management, tracheostomy, EXIT, case report

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Introduction

Head and neck teratomas are uncommon, accounting for only about 10% of all teratomas. Among these, the nasopharynx and cervical region are the most frequently affected sites. When the lesion arises in the oropharyngeal area, it is referred to as epignathus, with a reported incidence ranging between 1 in 35,000 and 1 in 200,000 live births. These tumors are derived from all three germ layers (ectoderm, mesoderm, and endoderm) and are classified as mature (benign) or immature (malignant) (1). Although often benign, they can pose a life-threatening risk to neonates due to potential airway obstruction. Large masses in the oropharynx may interfere with fetal swallowing, leading to polyhydramnios and severe respiratory distress at birth (2).

Advances in prenatal imaging, especially ultrasound and fetal magnetic resonance imaging (MRI), allow for early detection, anatomical characterization, and evaluation of airway compromise (3,4). This enables multidisciplinary delivery planning, often involving the ex utero intrapartum treatment (EXIT) procedure. EXIT maintains fetoplacental circulation during partial delivery, providing critical time to secure the airway via intubation or tracheostomy (5). The purpose of sharing this case is to introduce the EXIT procedure to otolaryngologists and emphasize its life-saving potential in cases of anticipated neonatal airway obstruction. We present a rare case of a large fetal oropharyngeal teratoma managed successfully with EXIT and early postnatal surgical resection.

Case Presentation

A 30-year-old woman at 29 weeks of gestation presented with lower abdominal pain. Targeted ultrasound revealed a large heterogeneous mass occupying the fetal oropharynx, with accompanying polyhydramnios (Figure 1a). Fetal biometry was consistent with gestational age, and no other anomalies were identified. Maternal serum alpha-fetoprotein was markedly elevated to 48,000. Fetal echocardiography showed normal cardiac anatomy and function.

Fetal MRI demonstrated a 5×5 cm heterogeneous mass originating from the oropharynx, expanding the oral cavity and displacing airway structures, suggesting a high risk of airway obstruction at birth (Figure 1b). Although elective cesarean delivery was initially planned, spontaneous preterm labor occurred at 32 weeks, prompting urgent delivery. An EXIT procedure was performed, and a tracheostomy was established while fetoplacental circulation was maintained. The neonate was then separated from the placenta and transferred to the neonatal intensive care unit.

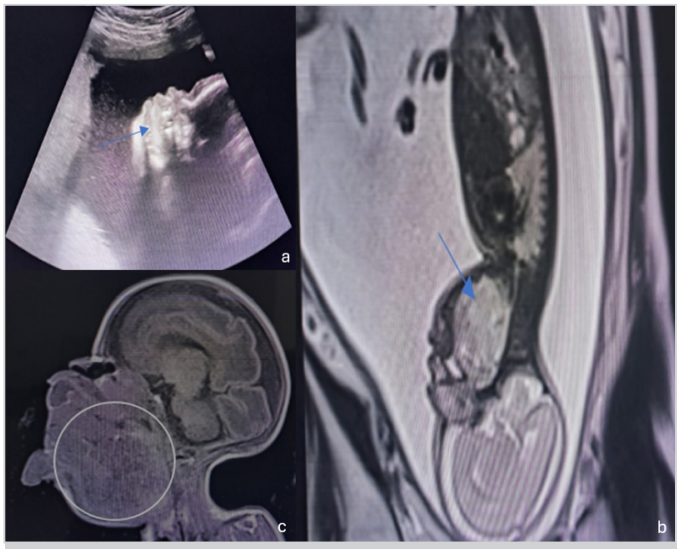

Figure 1. Prenatal and postnatal imaging showing a fetal oropharyngeal mass detected by ultrasound and magnetic resonance imaging (MRI) before birth and confirmed by computed tomography (CT) on postnatal day 2. **a)** Ultrasound image obtained 30 days prior to cesarean section showing a hyperdense mass in the fetal oropharynx. **b)** Fetal MRI obtained 25 days prior to cesarean section showing a heterogeneous mass measuring approximately 5×5 cm located in the oropharynx. **c)** Sagittal CT image of the fetus obtained on postnatal day 2 following cesarean section

Postnatal computed tomography imaging further characterized the mass, revealing extension into the prevertebral space (Figure 1c). On the second postnatal day, a transoral surgical excision was performed in collaboration with neurosurgery and the mass was completely resected (Figure 2). Histopathology confirmed an immature teratoma. Postoperative recovery was uneventful. The infant was initially fed via nasogastric tube for one week, followed by successful oral feeding. No postoperative complications occurred. Follow-up at 24 months showed no evidence of recurrence. The tracheostomy tube was successfully removed at the 5th postoperative month after confirming adequate airway patency. Written informed consent was obtained from the patient's father for publication of this case report and the accompanying images.

Discussion

This case highlights the critical importance of prenatal diagnosis and coordinated multidisciplinary planning in managing fetal oropharyngeal teratomas. Teratomas may arise in multiple anatomical locations, with the sacrococcygeal region representing the most common site,

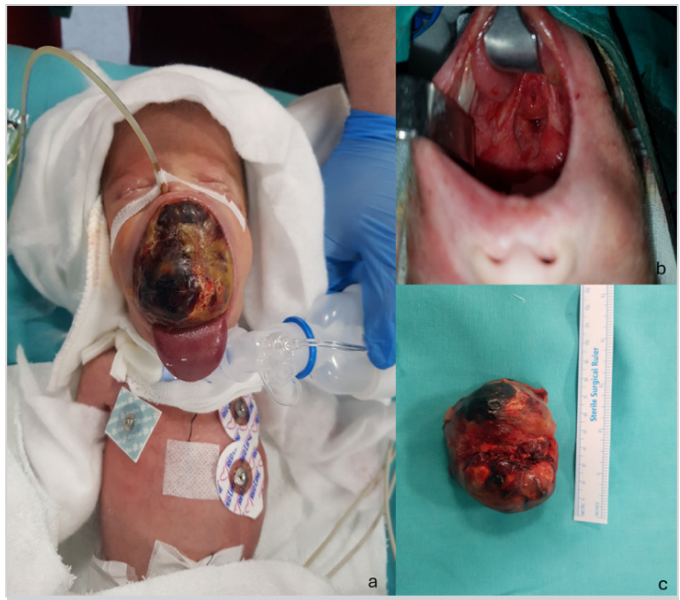

Figure 2. Intraoperative and postoperative views showing the oropharyngeal mass before resection, the surgical field after excision, and the gross appearance of the resected specimen. **a)** Pre-resection image of the mass. **b)** The appearance of the oral cavity and laryngeal structures following mass excision. **c)** The resected mass

accounting for approximately 40% of all cases. Less than 5% are found in the head and neck. These tumors demonstrate a higher incidence in females, and when identified during early childhood, they are generally benign. Epignathus denotes a teratoma originating in the oropharynx, which, when large, can result in craniofacial distortion and severe respiratory compromise at birth. Prenatal ultrasound, particularly when complemented by fetal MRI, allows for early detection and detailed evaluation of the mass, including its size, composition, relationship to airway structures, and vascularity. In our case, the presence of polyhydramnios and a large oropharyngeal mass suggested a high likelihood of postnatal airway compromise. These findings prompted early planning and involvement of a multidisciplinary team, which contributed to the favorable outcome (4,6).

The EXIT procedure was essential in managing the airway in this case. Originally described in 1990, the EXIT-to-airway technique has become an established approach for fetuses with anticipated airway obstruction, including cervical masses, large goiters, congenital high airway obstruction syndrome, and oropharyngeal teratomas (7). The procedure involves partial cesarean delivery with ongoing placental perfusion to preserve oxygenation while securing the neonatal airway (8). This controlled setting provides a crucial window of time typically 30-60 minutes for safe intubation or tracheostomy, reducing the risk of hypoxia and emergency resuscitation (9).

In our case, we anticipated that conventional intubation would be extremely difficult due to a large mass almost completely occupying the fetal oropharynx. EXIT provided a critical window for performing a calm, controlled tracheostomy while the fetus remained oxygenated via placental support. Once the airway was secured, the neonate was fully delivered and ventilated. This approach prevented hypoxia and enabled safe transition to postnatal care. The literature strongly supports the use of EXIT in similar high-risk airway cases. King et al. (8) described twins with oropharyngeal teratomas who both underwent EXIT: one was intubated, and the other required partial tumor debulking and tracheostomy, demonstrating EXIT's flexibility. Hu et al. (2) similarly emphasized that EXIT improves survival in infants with epignathus, especially when airway obstruction is anticipated. In contrast, smaller oropharyngeal teratomas without evidence of airway compromise may be managed with routine delivery followed by prompt intubation (10). Our review showed that most large oropharyngeal teratomas in the literature were successfully managed with EXIT, which has been associated with improved outcomes compared to historical cases without it (6,9).

Definitive treatment of oropharyngeal teratomas is complete surgical excision, ideally during the neonatal period. In our case, the tumor was removed on the second postnatal day. Surgery not only resolves airway obstruction but also facilitates feeding and normal orofacial development. Resection is curative in most cases, particularly for histologically immature teratomas. For immature teratomas, which carry malignant potential, surgery remains the first-line treatment. Adjuvant chemotherapy is considered in high-grade lesions, but low-grade (grade 1,2) immature teratomas that are completely excised can often be managed conservatively with serial tumor marker surveillance (1,9). Our patient followed this conservative approach, with no recurrence observed to date.

Review of similar cases shows favorable outcomes with early diagnosis and multidisciplinary care. Zhu and Li (9) reported two postnatally diagnosed cases that were successfully resected with no recurrence. Hu et al. (2) described a case managed with EXIT and early surgery, also with good outcome. Conversely, Garg and Singh (6) presented a fatal case of undiagnosed teratoma where delayed airway management led to neonatal death. The literature emphasizes early prenatal detection, comprehensive antenatal imaging, multidisciplinary planning, and use of EXIT when airway compromise is expected. Following these principles transforms a potentially fatal condition into a survivable one (6,9).

Conclusion

We presented a rare case of a large fetal oropharyngeal teratoma diagnosed in the third trimester. Successful management included a planned EXIT procedure with tracheostomy and complete neonatal tumor resection. Histopathology confirmed an immature teratoma. The purpose of sharing this case is to introduce the EXIT procedure to otolaryngologists and emphasize its life-saving potential in anticipated neonatal airway obstruction through timely, multidisciplinary planning.

Ethics

Informed Consent: Written informed consent was obtained from the patient's father for publication of this case report and the accompanying images.

Footnotes

Authorship Contributions

Surgical and Medical Practices: M.A., F.C.E., Concept: M.A., F.C.E., Design: M.A., A.T.D., Data Collection and/or Processing: M.A., A.T.D., Analysis and/or Interpretation: F.C.E., Literature Search: M.A., Writing: M.A.

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Main Points

- Oropharyngeal teratomas (epignathi) are rare congenital tumors that may cause life-threatening neonatal airway obstruction.
- Prenatal imaging, especially ultrasound and fetal magnetic resonance imaging, plays a crucial role in early diagnosis and delivery planning.
- The ex utero intrapartum treatment (EXIT) procedure allows for safe airway management while maintaining placental circulation.
- In this case, a large fetal oropharyngeal teratoma was diagnosed at 29 weeks and successfully managed with EXIT and neonatal tumor resection.
- This case demonstrates the importance of a multidisciplinary approach and introduces the EXIT technique to otolaryngologists as a life-saving option in similar scenarios.

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