

Rapidly growing cervical teratoma – fetal death during delivery

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Abstract

Here we present a rapidly growing fetal cervical teratoma associated with agenesis of the corpus callosum that caused an early delivery resulting in rupture of the mass and neonatal death during Cesarean section. Large cervical lesions may threaten the life of the fetus during delivery of the head.

Keywords

Cervical teratoma, fetus, cesarean section, delivery, fetal death, agenesis of corpus callosum.

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Introduction

Giant cervical teratoma which arises from the oral cavity or pharynx is a very rare disease in neonates. The estimated incidence is between 1/35,000 and 1/200,000 [1]. These tumors are benign but they may cause severe morbidity and mortality due to airway obstruction. Successful management of prenatally diagnosed giant cervical teratomas by Cesarean section (C-section) and *ex utero* intrapartum treatment (EXIT) procedure has been reported in many case series [2, 3].

Cesarean section is the preferred mode of delivery for the safety of the fetus and to avoid dystocia. We report a case of giant cervical teratoma which caused fetal death during delivery due to rupture of the fragility of the tumor.

Case

A 24-year-old pregnant woman at 20 weeks of gestation was referred to our Perinatology Clinic with a diagnosis of fetal cervical mass. Ultrasound revealed a left lateral heterogeneous neck mass, measuring 30 × 42 × 35 mm that consisted of cystic and solid areas. Magnetic resonance imaging showed a solid-cystic heterogeneous mass beginning in the pre auricular area and extending to the anterior chest wall that compressed the fetal airway (**Fig. 1**). The Council of Perinatology decided to follow up the pregnant patient with serial ultrasound/MRI examination. The delivery mode of the fetus was planned as an elective C-section using the EXIT procedure.

During the follow-up, rapid enlargement of the mass and heavy polyhydramnios were observed at 24 weeks of gestation. At the detailed ultrasound exam, the mass was measured as 110 × 97 × 96 mm and agenesis of the corpus callosum was also detected.

During week 25, an emergency C-section was performed due to an early rupturing of the membrane and a breech presentation. However, the mass ruptured during the C-section due to its large size and fragile nature. The rupture of the mass caused fetal hemorrhage during the delivery

of the fetus. A male infant (birth weight: 790 g; birth length: 28 cm) was delivered with Apgar scores of 2 and 0 at 1 and 5 minutes, respectively. Neonatal resuscitation failed and the baby died. The macroscopic appearance of the newborn is shown in **Figures 2-4**. The family refused an autopsy and complete excision of the mass. Thus, we only received large biopsies from different areas of the mass. The histopathological examination was consistent with a teratoma and genetic analysis revealed a normal karyotype.

Discussion

Cervical tumors are rare in fetal life. Cystic hygromas, teratomas, hemangiomas, lymphangiomas congenital goiters, solid thyroid tumors, thyroid cysts, branchial cleft cysts, neuroblastomas, epignathi and hamartomas are the common types of lesions located in this area [4]. Cystic hygroma, branchial cleft cyst, thyroglossal duct cysts are all noncalcified and mainly cystic and hemangiomas are calcified and mainly solid. Developmental branchial and thyroglossal cyst are often multilocular with only a thin rim of enhancement and do not contain fat [5].



Figure 1. MRI.



Figure 2. The ruptured mass.



Figure 3. The fetus.

Tumors located in the cervical region are very important due to the contiguous organs. These tumors have the potential to obstruct the airway, which can cause hypoxia and death after delivery. Large masses in this area can also cause impaired fetal swallowing, polyhydramnios, and preterm labor during the prenatal period [6, 7]. Ultrasound and MRI may be used together, prenatally, to determine the relationship between the fetal mass and the airway. Fetal sonography is useful to determine the vascularity and consistency of the mass and to identify the associated signs, such as polyhydramnios. The extent of the lesion, infiltration into the surrounding organs, and the obstruction pattern can be visualized using fetal MRI [5]. Antenatal fetal MRI and ultrasonography together may be useful in identifying fetal neck masses that require the EXIT procedure [8]. After a prenatal diagnosis, multidisciplinary management is advised. Counseling, serial im-



Figure 4. Lateral view.

aging, and planned deliveries are the mainstays of this management.

In our case, the patient was referred to our clinic with a cervical mass that measured 30 × 42 × 35 mm. We discussed the case with pediatricians and pediatric surgeons. Serial ultrasounds of the lesion and a close follow-up were planned; however, the tumor measured 110 × 97 × 96 mm only 4 weeks later. A few days later, an emergency C-section was performed due to rupturing of the membranes and a breech presentation. Heavy polyhydramnios was found, probably due to swallowing difficulty caused by the rapid growth of the tumor. The pathologic investigation revealed a cervical teratoma.

Teratomas occur in 1 out of 4,000 live births, demonstrating a female predominance [5]. Teratomas were classified in four different groups as dermoid cysts, teratoid cysts, true teratomas and epignathi (cervical-oro-pharynx) [9].

Congenital cervical teratomas occur one in every 20,000-40,000 live births [10]. Teratomas in this location are large, bulky tumors and likely to cause airway compression, mandating close prenatal

monitoring of the mass [11]. These rare tumors may also grow aggressively and cause obstruction in the esophageal lumen by external compression, leading to polyhydramnios and an increased risk for preterm delivery. Polyhydramnios and hydrops, along with tumor size > 5 cm, carry a poor prognosis for the fetus [12].

Hirose et al. reported on seven patients with a prenatal diagnosis of a large cervical teratoma. Four patients had fetal hydrops; of these, two died *in utero* and a third underwent an elective termination [13].

Associated anomalies are also signs of a poor prognosis. We also detected agenesis of the corpus callosum in our case. The association of cervical teratomas with agenesis of the corpus callosum has been previously reported as a case report [14].

An elective, planned delivery is essential in cases with a cervical teratoma. Large cervical lesions may threaten the life of the fetus during delivery of the head. In our case, the fetus was delivered by an emergency C-section. Despite the experience of the surgeon and general anesthesia, the mass ruptured during delivery. This could be attributed to the discordancy between the head and the body of the fetus and uncontrolled uterine contractions. Larger or vertical incisions of the uterus and uterine relaxing agents may be preferable for these types of cases.

Fetal cervical teratomas are rare tumors. Close monitoring of the mass and a planned delivery with pediatric surgeons are the mainstays for the management of these cases. In addition, one must consider that rapidly growing cervical masses can rupture due to their fragility, even during C-sections that are performed by experienced hands.

Declaration

The Authors state that parents consent and ethical approval were taken and that patient anonymity was maintained. The Authors state that ethical standards were followed.

Declaration of interest

The Authors declare that there is no conflict of interest. Funding: none.

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