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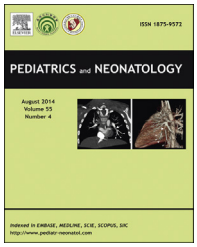

CASE REPORT

Huge Neck Masses Causing Respiratory Distress in Neonates: Two Cases of Congenital Cervical Teratoma

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Congenital cervical teratomas are rare and usually large enough to cause respiratory distress in the neonatal period. We present two cases of congenital huge cystic neck masses in which distinguishing cervical cystic hygroma and congenital cystic teratoma was not possible through radiologic imaging techniques. Experience with the first case, which was initially diagnosed and treated as cystic hygroma by injection sclerotherapy, led to early suspicion and surgery in the second case. The masses were excised completely and histopathologic diagnoses were congenital teratoma in both patients. Our aim is to review congenital huge neck masses causing respiratory distress in early neonatal life to highlight this dilemma briefly with these interesting cases.

1. Introduction

Fetal neck masses are rare and may be encountered during fetal anomaly screening scan during the second trimester. It is essential to distinguish the different pathologies as they influence prenatal counseling, antenatal, and postnatal management. In the neonatal period, differentiating the various neck masses and reaching an accurate diagnosis are important for the prognosis and appropriate timing of surgical treatment.[1] The anatomic location of the mass can yield a clue about probable origin of the mass; anterior neck masses include teratoma, epignathus, goiter, bronchogenic cyst, and hemangioma; and posterior masses include cystic hygroma (cystic lymphangiomas located in the head and neck), cervical meningocele, occipital encephalocele, and hemangioma.[1] Lymphangiomas and teratomas may be seen as giant fetal neck masses.[2] Complete and meticulous surgical excision is the recommendation for each of them; however, several medical treatments have been preferred in the treatment of lymphangioma to avoid surgical complications.[3] Herein, we report two cases of congenital cervical teratoma, one of which was initially

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treated as lymphangioma according to radiological diagnosis.

2. Case Reports

2.1. Case 1

A 1-day-old newborn was referred to our unit for a huge neck mass. The mass was first detected at 12 weeks' gestation as a small cyst, but the subsequent ultrasonography (US) scan was normal. Physical examination revealed a huge neck mass (Figure 1). The chest radiography, abdominal US, and echocardiography were normal. The tumor involving both sides of the neck extended from the auricle to the clavicle. US revealed a 13 cm × 9.5 cm × 7 cm multiloculated, multiseptate mass. Blood flow in the tumor was fairly unremarkable, and pulsating flows were not detected. Computed tomography (CT) revealed an 8 cm × 13 cm × 7 cm multiloculated cystic mass, which extended from the left maxilla to bilateral submandibular area, and to the anterior chest surrounding the trachea at both sides with scattered microcalcifications (Figure 2A).

Based on the US findings, we considered the tumor to be lymphangioma. We planned injection of OK-432 (Picibanil, Chugaiv Pharmaceutical Co., Tokyo, Japan). During the 3rd postnatal week, the baby developed respiratory distress and was intubated. On postnatal Day 27, OK-432 was injected into the largest cysts under ultrasonographic guidance in the interventional radiology unit. After fluid aspiration from the lesion, the same amount of OK-432 was introduced up to a maximum of 20 mL with a concentration of 0.01 mg/dL. Cervical magnetic resonance imaging (MRI) obtained 10 days after repeated sclerosing therapy revealed multiloculated cystic lesions with septae of varying thickness and no reduction in the size of the mass (Figures 2B and C). Surgical intervention was undertaken through a left-sided incision over the tumor. In the midline, the mass tightly adhered to the tracheal cartilages. The tracheal cartilage was injured during excision in vertical plane in a 1.5 cm length and primarily repaired. The huge mass was totally excised. Respiratory distress required tracheotomy on postoperative Day 29, and feeding was maintained via a gastrostomy tube. The patient did not experience any recurrent mass during 1-year follow-up. The postoperative MRI showed normal anatomic structures without recurrent disease (Figure 2D).

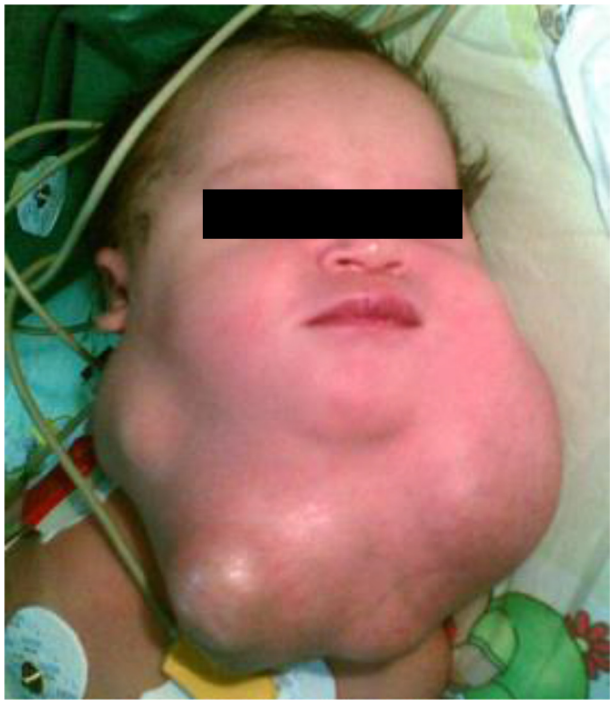

Figure 1 Huge lobulated irregular neck mass.

Microscopic examination of the specimen revealed skin adnexal structures, mature glial tissue, cartilage, smooth muscle, fibrous and fatty tissue, minor salivary glands, respiratory and gastrointestinal structures, and thyroid tissue. These findings were consistent with mature teratoma that developed from all three germ layers (Figure 3A).

2.2. Case 2

A 2-day-old male child was referred to our hospital for a large cervical mass. The child developed respiratory distress soon after birth and, therefore, he was intubated. On the first antenatal US, which was performed at 12 weeks' gestation, no abnormality was detected. However, polihydroamnios was detected on US in the 3rd trimester.

The tumor was located predominantly on the left side of the neck from the level of mandible to the supraclavicular space extended to the right side. The α-fetoprotein (AFP) level was 226.689 ng/mL. The chest radiography, abdominal US, and echocardiography did not reveal any malformations except the neck mass. MRI confirmed a 5 cm × 3.5 cm multiloculated cystic mass, which was hyperintense on the T2-weighted images, and demonstrated peripheral wall enhancement on the T1-weighted image, extending from the left hypopharynx—parapharyngeal space to the caudal anterior midline, submandibular region, and partly to the right sternocleidomastoid muscle (Figure 4). On the 2nd day, surgical intervention was undertaken through a left-sided incision over the tumor. A multiloculated cystic mass, which had a definite capsule, was totally resected with preservation of the significant locoregional structures such as thyroid and parathyroid glands. The AFP level dropped to 63.125 ng/mL on postoperative Day 5. He was extubated on postoperative Day 7.

The mass was composed predominantly of immature neuroepithelial tissue forming rosette-like structures, tubules and immature cartilage (Figure 3B), bone, smooth muscle, and fatty tissue in histopathological examination. The diagnosis was immature teratoma.

The patient was discharged on Day 45. During the 5-month follow-up, the patient remained well, developing normally with no evidence of recurrence.

3. Discussion

Routine antenatal US in populations reduced the unexpected cervical masses presenting soon after delivery. Antenatal diagnosis may also be made following targeted US evaluation

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