Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Obstetrics and gynaecology

Fetal Oropharyngeal Teratoma: A Rare Tumour: A Case Report

Gagan Lata[1*], Liza Singla[2], Moneet Walia[3]

[1]Associate Professor, Department of Obgy, Adesh Medical College & Hospital, Kurukshetra, India
[2]PG Resident 3rd Year, Department of Obgy, Adesh Medical College & Hospital, Kurukshetra, India
[3]Professor, Department of Obgy, Adesh Medical College & Hospital, Kurukshetra, India

DOI: 10.36347/sjmcr.2023.v11i09.034 | **Received:** 20.08.2023 | **Accepted:** 25.09.2023 | **Published:** 29.09.2023

***Corresponding author:** Dr. Gagan Lata
Associate Professor, Department of Obgy, Adesh Medical College & Hospital, Kurukshetra, India

Abstract **Case Report**

Teratomas generally constitute 25-30% of congenital tumours with oropharyngeal tumours being very rare. The tumor typically arises from the palato-pharyngeal region around the basishenoid. Incidence is in 1 in 35,000-200,000 of live births. Most head-and-neck teratomas are incidental findings on routine second-trimester ultrasonography and are usually associated with polyhydramnios in 40% of cases. This case was also diagnosed with fetal oropharyngeal tumour at 20 weeks USG and was associated with polyhydramnios. After counselling, couple opted for termination of pregnancy in view of poor prognosis. After expulsion, products of conception were sent for histopathology and karyotyping.

Keywords: Oropharyngeal teratoma, polyhydramnios, Ultrasonography.

INTRODUCTION

Teratomas are rare tumours which are derived from the three germinal cell layers. These occur most commonly in the sacrococcygeal region, but they can arise anywhere in the body along the midline [1, 2]. Head-and-neck teratomas constitute 1%–9% of all teratomas with oropharyngeal teratomas being extremely rare (<1%) [3]. The tumor typically arises from the palato-pharyngeal region around the basishenoid. Incidence is in 1 in 35,000-200,000 of live births [4, 5]. Most head-and-neck teratomas are incidental findings on routine second-trimester ultrasonography and are usually associated with polyhydramnios in 40% of cases [6]. The polyhydramnios occurs as a result of very large tumour mass which causes oesophageal compression, thereby preventing amniotic fluid absorption because of failure of foetal deglutition [7]. The level of maternal serum alpha-fetoprotein (AFP) may be elevated. Ultrasound can help in the pre-natal diagnosis of this condition, although magnetic resonance imaging (MRI) is useful for further characterization of the lesion. Because of severe obstruction to airways, OPTs are associated with high morbidity and mortality rates during peripartum period. We present here a case of fetal OPT with imaging characteristics with respect to the antenatal diagnosis.

CASE REPORT

Thirty one years old female G2P1L1 came to antenatal OPD at 20+2 weeks with report of anomaly scan. Earlier she was booked at some other place from where she was referred to PGI Chandigarh for MTP in view of fetal facial teratoma which was incidental finding on USG. At PGI Chandigarh, she was advised MTP by paediatric surgeon. She came to AMCH, Shahbad for second opinion where she was admitted. She was married 3 years back and had previous caesarean section one and half year back in view of cephalopelvic disproportion. She had taken folic acid in first trimester. NT NB scan and double marker was not done in first trimester. She was not exposed to X Ray and unsupervised drugs in first trimester .No history of comorbities and no family history of congenital anomalies. On examination fundal height was 22 weeks which was more than period of gestation due to polyhydramnios. Repeat USG was done at AMCH which showed SLIUF with polyhydramnios with placenta covering the os, large solid cystic swelling on the face in region of lip and nose measuring 4.8x6.4 cm, heteroechoic mass involving face and neck? facial teratoma.She was advised MRI but couple refused for that. After appropriate counselling about the poor prognosis of baby, she was planned for hysterotomy in view of placenta covering the os. Female fetus weighing 425 grams was extracted with an exophytic tumor protruding through the oral cavity. Tumour was partly solid and partly cystic. Patient did well in post operative period. Products of conception were sent for histopathology which revealed clinical diagnosis of Immature Teratoma (Face). Further products of

conception were sent for chromosomal analysis. FISH analysis was performed which showed normal number of chromosomes 13, 18, 21 and normal sex chromosome pattern in the cells which showed that it was not associated with any other chromosomal abnormality.

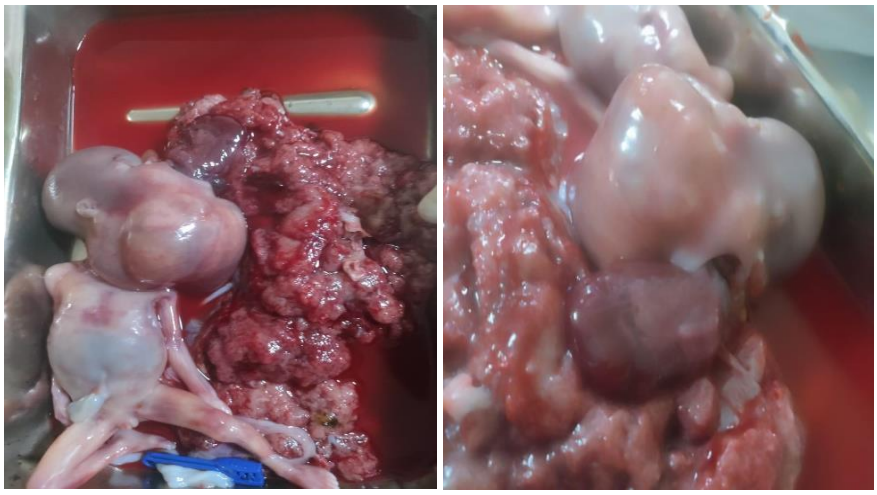

Figure 1: Aborted fetus showing facial mass

DISCUSSION

Teratomas account for around one-third of all neonatal tumours [8]. Teratoma is derived from the three developmental germ layers, called the ectoderm, mesoderm, and endoderm. These germ layers are made of germ cells, which are capable of forming a variety of tissues, including fat, muscle, teeth, nails, and hair. During fetal development, some of these germ cells do not differentiate, thereby retaining the ability to turn into other cell types. These germ cells then migrate through the midline until they reach the primitive gonads where they can remain for decades, eventually developing into eggs or sperm. It is thought that a problem during these cells' differentiation process could lead to the development of a teratoma, which explains why teratomas may occur in the gonads themselves or along the path of germ cell migration. So, teratomas are found primarily in the gonads, but they can develop anywhere in the body, especially in midline locations, such as the sacrum, coccyx, mediastinum, retroperitoneum, and central nervous system. Mature teratomas tend to be benign, whereas immature teratomas are more likely to undergo malignant transformation. Oropharyngeal teratoma are exceptionally rare fetal teratomas and present with oropharyngeal mass, polyhydramnios and elevated maternal serum AFP level [9]. It appears as heterogeneous mass of solid and cystic components which is believed to occur due to trapping of mesoderm and endoderm with ectoderm during embryogenesis [10]. The tumour typically arises from palate-pharyngeal region around the basisphenoid (Rathke's pouch). With progressive growth it fills the buccal cavity and finally protrudes out of the mouth. Due to both solid and cystic elements, Oropharyngeal teratoma usually appears as heterogeneous mass on ultrasonography. Magnetic resonance imaging (MRI) is, however, helpful in detailed characterization of lesion, excluding central nervous system (CNS) involvement and determining the tracheal anatomy for airway safety.

Maternal serum AFP is a commonly used test during second trimester to assess a possible neural tube defect. An elevated level of AFP in maternal serum can lead to sonographic investigation and diagnosis of OPT [11, 12]. AFP levels may, however, be within the normal range and are not specific to this condition. In our case, however, the patient had presented very late and the maternal serum AFP test had not been done during the second trimester, so the USG detected the condition first. This signifies that regular prenatal check-ups and screening tests are necessary in order to rule out a fatal defect earlier, a procedure that women living in rural and remote areas often ignore in the developing countries. Other conditions where maternal serum AFP levels may be elevated are pregnancies with foetal neural tube defects, autosomal trisomies, sacrococcygeal teratoma, and esophageal or anorectal atresia [13]. The prognosis oropharyngeal teratoma is generally very poor. There is no evidence to suggest environmental risk factors in the pathogenesis of pharyngeal teratoma. Furthermore, there are no karyotypic abnormalities involved, and these lesions are not thought to be inherited in a Mendelian or polygenic manner [14]. Therefore, parents should be reassured that they are not at increased risk of having another child with this lesion. Prenatal diagnosis is important because newborns that are born with pharyngeal teratomas that compress the airways can experience postnatal asphyxiation, brain damage and even death [15]. It was observed that if the tumour body was larger than the mouth, it was easy to identify it on prenatal ultrasound examination. However, if the tumour site was deeper, prenatal ultrasound may not be able to identify the tumour. Prenatal ultrasound examination is an essential and reliable method for identifying fetal oropharyngeal teratomas. Three-dimensional colour

ultrasound can clearly show the relationship between the tumour and the oral space [16].

CONCLUSION

Antenatal diagnosis of oropharyngeal teratoma is useful as it helps to decide the plan of management. Parents should be told about the risk and benefits of continuing the pregnancy and they can opt accordingly. Diagnosis ultimately is confirmed by histopathology and further chromosomal analysis can be done to rule out associated anomalies.

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