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GRADE III IMMATURE TERATOMA OF PLACENTA: DIAGNOSIS OF SURPRISE

Histopathology

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ABSTRACT

Placental teratoma is rare non-trophoblastic tumor of placenta, for which definitive prenatal diagnosis poses a great challenge for clinician and radiologist as well. We report an extremely rare case of grade III immature placental teratoma (second case in English literature searched, to the best of our knowledge) in a 36 years old female. USG revealed a large lobulated heterogenous solid-cystic mass measuring 16.9x10.5x10.5 cm. in uterine cavity focally abutting placenta. After preterm elective LSCS, intact placenta with attached mass was sent for histological examination which confirmed the diagnosis of grade III immature placental teratoma. Besides morphological features and radiological findings, molecular alterations and evidence of twining by molecular genotyping may prove very useful in arriving at the diagnosis and provide further insight to the ongoing dilemma.

KEYWORDS

Acardius, immature, placenta, teratoma.

INTRODUCTION

Placental teratoma is rare non-trophoblastic tumor of placenta, only a fewer than 30 cases have been reported after the first case report of placental teratoma by Morville in 1925.[1,2] Immature teratoma of placenta is even rarer. Only single reported case of placental immature teratoma was found in the literature searched.[2] Although teratomas can be suspected antenatally by ultrasound but in view of extreme rarity, unusual site, variegated presentation, and long list of differential diagnoses, the definitive prenatal diagnosis poses a great challenge for clinician and radiologist as well. At times, these tumors may cause variable clinical impact; from compression to fetal and placental hemodynamic changes to fetal growth retardation to even fetal demise.[3]

We report an extremely rare case of grade III immature placental teratoma (second case in English literature searched, to the best of our knowledge) in a 36 years old female.

CASE REPORT

A 36 years old booked pregnant female (G2P1L1 with previous LSCS) presented to OPD at 28 weeks of gestation period, perceiving fetal movements well and with clinical suspicion of angiomyxoma of umbilical cord. She had no complaints of bleeding per-vaginum or pain abdomen or history of chronic illness. She had history of eclampsia in previous pregnancy. Hemogram revealed: Hb-10.4 gm%, TLC-11,310 cells/cumm, DLC-N88%, L10%, M02%, Platelet count-1.69 lac/cumm. Ultrasound examination revealed a large lobulated heterogenous predominantly solid mass with few internal cystic areas and calcific foci, measuring 16.9x10.5x10.5 cm. present in uterine cavity focally abutting placenta near the insertion site of umbilical cord. Surrounding anechoeic fluid is seen with a thin membrane separating it from fetal amniotic sac. It shows internal vascularity on Color Doppler with low resistance arterial flow. This mass was of approximately 6.8x5.5x3.6 cm. size in previous ultrasound done at 20 weeks of gestation, with normal fetal echocardiogram. At that time of antenatal checkup, further quadruple screening test was done which was suggestive of Neural tube defects. Magnetic resonance imaging (MRI) was also suggested but not done.

Patient underwent elective preterm LSCS delivery. Peroperatively, scar dehiscence of about 1 cm. is noted in left lower uterine segment and a live baby newborn was delivered as vertex. Baby was sent to NICU and kept on ventilator, later baby expired and was found to have clinical suspicion of congenital pulmonary airway malformation (CPAM). Intact placenta with attached umbilical cord and solid to cystic mass was sent for histopathological examination.

Macroscopic examination revealed placental disc measuring 18x17x3.8 cm. with eccentrically attached 16 cm. long umbilical cord and attached mass measuring 17x17x9 cm. (Figure 1a). Outer surface of mass was multinodular, solid to cystic to translucent at places and partially covered by membranes. The nodules varied in size from 2.5x2 cm. to 10.5x8 cm. Some of the cystic areas were filled with dirty white necrotic material, hemorrhagic fluid and mucoid secretions as well. Focal presence of bony hard area was also noted. No gross separate attached umbilical cord, axial skeleton, recognizable limb formation or internal organ formation in the mass was observed.

Microscopic examination of the mass showed variable admixture of mature tissue types including adipose tissue, cartilage, bone, smooth muscle bundles, choroid plexus like tissue, seromucinous and adnexal glands. Some cysts showed lining of gastrointestinal, respiratory and stratified squamous epithelium with pilosebaceous units. At places, multifocal calcification, horn cysts, oligodendroglioma like areas, immature neuroepithelial tissue (>3 low power field) forming rosettes, sheets and ependymal canal like structures were identified. The tumor was seen arising in between fetal membranes. No microscopic evidence of attached umbilical cord like structure to the mass was observed. Adjacent placenta and umbilical cord showed unremarkable histology. Histopathological diagnosis of Grade 3 Immature teratoma of placenta was considered.(Figure 1 & 2)

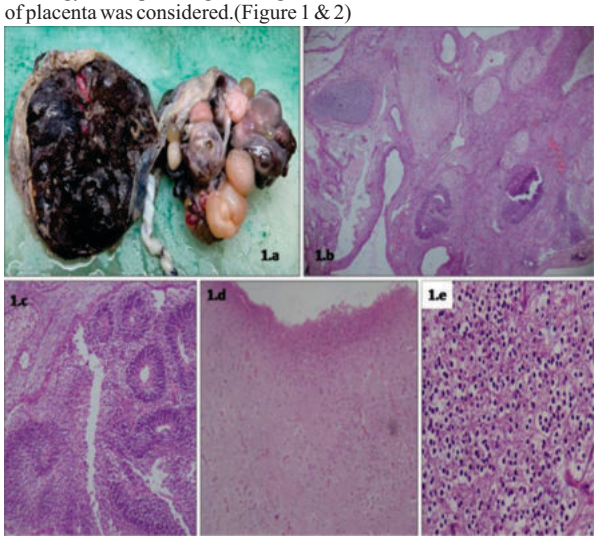

Figure 1.

a. Photograph showing gross appearance of the mass along with placenta.
b. Photomicrograph showing immature neuroepithelial tissue, cartilage & pseudostratified tall columnar epithelium (H&E,40X).
c. Photomicrograph showing immature neuroepithelial tissue forming tubules (H&E,100X).
d. Photomicrograph showing mature glial tissue (H&E,100X).
e. Photomicrograph showing oligodendroglioma like areas (H&E,400X).

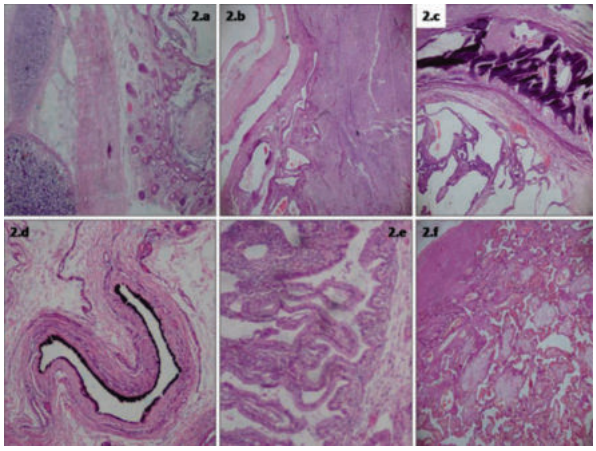

Figure 2.

a. Photomicrograph showing cartilage & cyst lined by skin with pilosebaceous units (H&E,40X).
b. Photomicrograph showing mature glial tissue with overlying fetal membranes (H&E,40X).
c. Photomicrograph showing bone & choroid plexus like tissue (H&E,40X).
d. Photomicrograph showing focal presence of neuromelanin (H&E,100X).
e. Photomicrograph showing presence of gastrointestinal type epithelium (H&E,400X).
f. Photomicrograph showing placental tissue with unremarkable histology (H&E,100X).

DISCUSSION

Teratomas are common germ cell tumors frequently arising in gonads although extragonadal locations are relatively less commonly involved. These tumors contain variable disorganized admixture of different tissue types originating from one or more germ cell layers. These are classified into mature (mature tissue types only), immature (immature tissue elements particularly neuroepithelium, primitive glomeuli, blastomatous tissue) and monodermal type (one specialised tissue type like thyroid tissue). Placental teratoma, moreover, immature type is extremely rare entity.

The origin and pathogenesis of occurrence of placental teratoma is still not clear and debatable. Several hypotheses have been proposed but two theories are favoured as possible mechanism; one is "germ cell theory" by Fox according to which, in early stages of embryogenesis, the primitive gut evaginates into the umbilical cord, and germ cells from the primitive gut aberrantly migrate through the connective tissue of the cord, reaching upto the extra-placental membranes, giving rise to an extra-placental membrane teratoma between the amnion and chorion. Second theory "included-twin hypothesis," by Nicholson suggested that teratoma originates from a twin fetus through some embryological mishap and may merge into it's co-twin. Tavares and Oporto believed that the teratoma cell line arises from two fused germ cells.[4-7]

Prenatal diagnosis of placental teratoma is even a challenge for radiologists and may be suspected by USG on the basis heterogenous admixture of tissue with varied echogenicity, such as calcification, adipose tissue and fluid filled cysts.[8] Acoustic shadowing on USG may indicate the presence of bone.[9] A teratoma appears as a well-circumscribed complex lesion, heterogeneous on T1- and T2-weighted sequences. 3D ultrasound and MRI volumes enhance characterization of the tumor tissues and produce 3D virtual physical models of different types of congenital anomalies.[10] However, in our case, the antenatal ultrasound did not consider a differential dignosis of teratoma but instead clinicoradiological provisional diagnosis of placental chorioangioma was considered.

The main differential diagnosis in the present case scenario was fetus acardius amorphous, which was excluded by the absence of gross or microscopic evidence of separate attached umbilical cord, cranio-caudal poles, axial skeleton organisation, recognizable limb like structure formation or internal organ formation. In case of any diagnostic dilemma, antenatal ultrasound findings in early gestation period may be correlated for any cardiac activity or evidence of multiple pregnancies. Since there are only limited numbers of reported cases of placental teratomas and not all cases are subjected to karyotyping or genetic sequencing, that's why associated chromosomal abnormality or specific genetic variations have not been demonstrated. However, Austin et al described a case of placental teratoma (based on above mentioned morphological criteria) by DNA genotyping investigation of the "teratoma" and its corresponding normal placental tissue which revealed an identical genetic profile at all microsatellite polymorphic loci with exception of one locus demonstrating loss of heterozygosity involving 1 of 2 "teratoma" samples tested.[11]

Awareness of this entity and clinicoradiological suspicion is of prime importance for an early antenatal diagnosis. In case of large tumors close antenatal follow-up and intra-partum observation are mandatory as there is risk of fetal asphyxia. From our perspective it is important for parents to know whether it is a true neoplasm or an extreme form of fetus acardius. As well this differentiation is also a diagnostic challenge for pathologists and radiologists. Besides morphological features and radiological findings, molecular alterations and evidence of twining by molecular genotyping may prove very useful in arriving at the diagnosis and provide further insight to the ongoing dilemma.

In literature searched, most of the reported cases of placental teratomas were benign or mature type and have favourable prognosis. All suspicious placental masses must be examined histopathologically supplemented with genetic studies to learn more about these cases including immature teratomas. Although, larger studies are not possible to be carried out due to the rarity of the disease, however, more accumulative and comparative diagnosis and follow-up of such cases along with genetic or ancillary studies, may help in resolving the issue in the future and help us in formulating a management plan.

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