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OROPHARYNGEAL FETUS IN FETU AT DR GEORGE MUKHARI ACADEMIC HOSPITAL. REVISITED

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ABSTRACT

Background: Fetus in fetu is a rare malformation in which a parasitic twin is within a more mature twin. Most of the fetus in fetu are located in the retroperitoneum and are acardiac and anencephalic.

Case presentation: Here, a presentation is made of an oropharyngeal fetus in fetu, which was a referral from Rustenburg hospital with suspected gastroschisis, pre-eclampsia, preterm labour at 28 weeks gestation.

Conclusion: Prenatal ultrasound remains the cornerstone in diagnosing fetus in fetu and thus provide the essential information for management and parental guidance.

Keywords: Fetus in fetu, Oropharyngeal, prenatal ultrasound.

1. INTRODUCTION

Fetus in fetu is a rare congenital abnormality which occurs in 1 /500.000 birth.

Firstly, described by Meckel in the 18th Century, as the presence of a parasitic twin in the body of its sibling twin.

Less than 200 cases have been reported so far, with the majority of them being located in the retroperitoneal region and only few oropharyngeal fetus in fetu cases. A preponderance of 2:1 has been reported in male. [(1)]

This is a benign condition with good prognosis in 97% of cases after complete surgical resection. Care must be taken to differentiate with a teratoma.

Oropharyngeal foetus in fetu carries a high risk of airway obstruction, hence an antenatal diagnosis by ultrasonography is paramount, which may lead to a multidisciplinary team management planning, for an Ex utero Intrapartum procedure.

This report presents an oropharyngeal Fetus in fetu case, which was managed at Dr George Mukhari Hospital, in November 2019.

2. CASE REPORT

A 36 years old patient P2G3, a referral from Rustenburg Provincial Hospital with the following problem list: pre-eclampsia, polyhydramnios, gastroschisis, pre-term labour and previous caesarean section at 28 weeks gestation.

On arrival at Dr George Mukhari Academic Hospital, the patient was reassessed and misdiagnosis of gastroschisis was repeated and the rest of the problem list was confirmed by the attending practitioner.

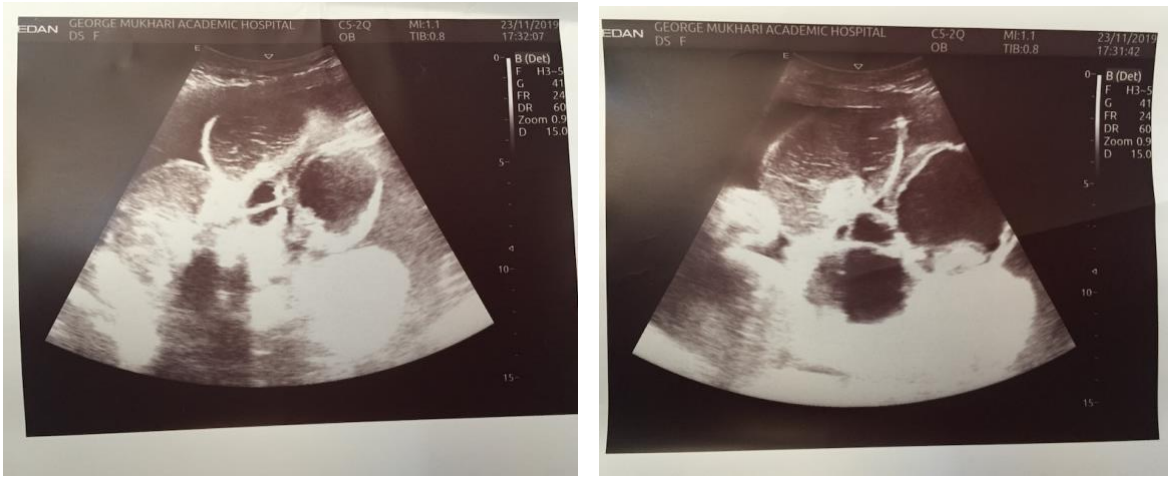

Figure 1: Multiple cystic masses and polyhydramnios on ultrasound.

The decision to perform an emergency caesarean section was made in conjunction with the consultant.

A female neonate was delivered through a classical incision with multiple large cysts originating from the mouth and attached to the nose thus obstructing the respiration. The presence of a parasitic twin which was anencephalic with a cornual tissue and bowel was identified.

The neonate died of inadequate oxygenation due to failed intubation.

The fetal blood was sent for karyotyping and the result came as 46 XX.

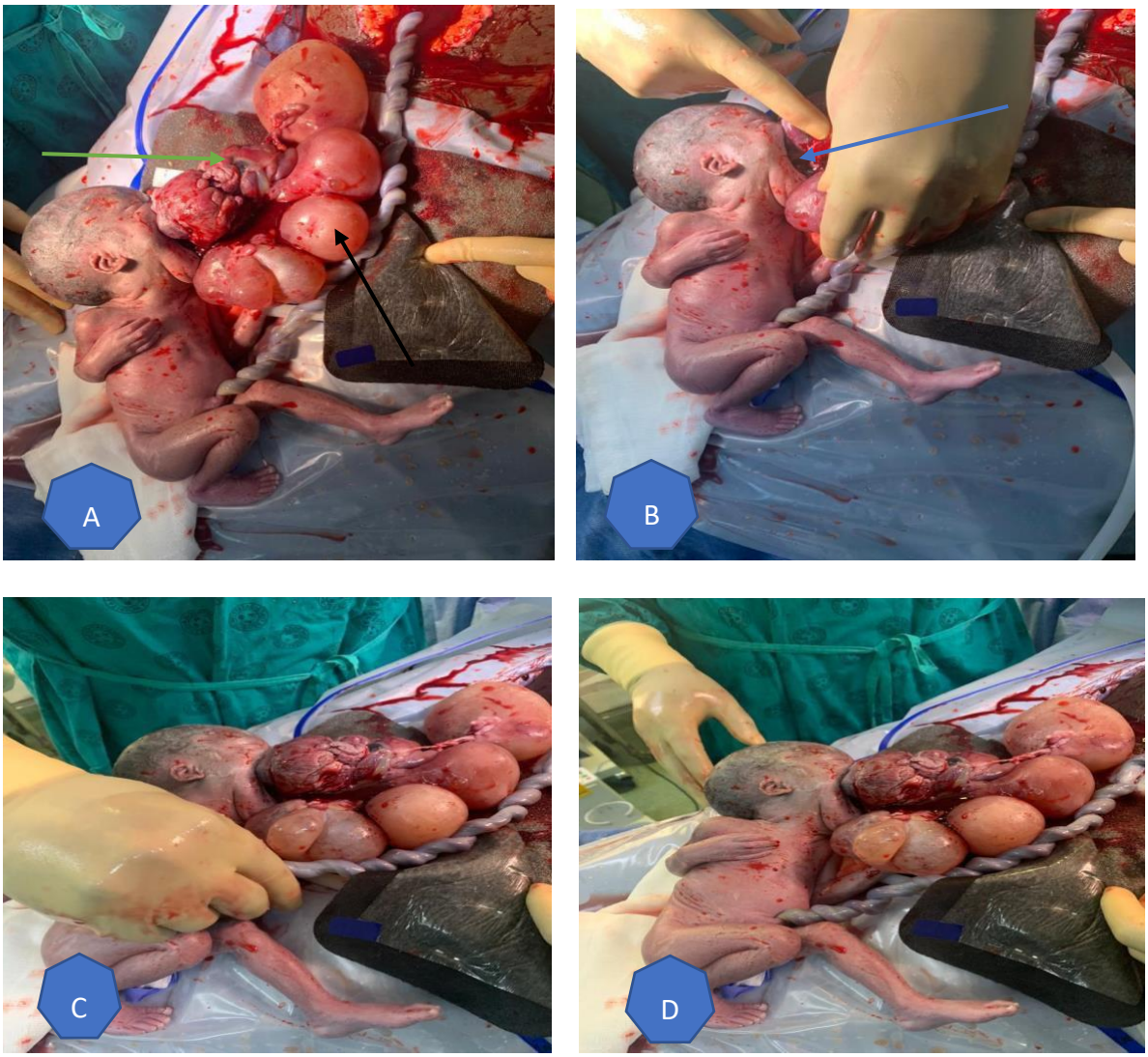

Figure 2: Female baby delivered with multiple cystic masses obliterating the airway. Note a corneal tissue highlighted by the green arrow and the bowels by the black arrow in A, Note how deep the mass is attached to the oral cavity of the host twin (blue arrow) in B.

3. DISCUSSION

Fetus in fetu is a rare congenital malformation, which occurs in diamniotic monochorionic pregnancy, resulting from unequal division of the totipotent inner cell mass of the developing blastocyst, leading to the inclusion of a smaller cell mass within a maturing sister embryo. [2]

Fetus in fetu is a term coined by Johann Friedrich Meckel in the 18th century, [3,4] initially reported in 1935, [5] and defined by Willis in 1953, as a mass characterized by vertebral axis often associated with other organs or limbs around the axis. [3]

It is a rare entity, with an incidence of 1 in 500.000 deliveries, [5] whereby a monozygotic diamniotic parasitic twin is incorporated into its sibling early in embryonic development and grows inside it. [3]

Various locations have been reported in the literature, 80% in the abdomen, 8% in the skull, 8% in the sacral region, and only few cases of oral or oropharyngeal cases, and complications vary according to locations, with abdomen distention, emesis and peritoneal inflammation for an abdominal location, obstructive hydrocephalus and mental retardation for a cranial location, and dysphagia and airway obstruction for an oropharyngeal location, which requires an emergency surgery. [4]

It has been reported that 97% of fetus in fetu had a good prognosis after complete surgical resection of the parasitic twin. [4] Therefore, the definitive management of foetus in fetu is complete surgical excision of the parasitic twin.

However, there have been isolated cases of malignancy following resection of a fetus in fetu, prompting some surgeons to recommend complete resection on an urgent basis followed by post-operative surveillance of tumour markers such as alfa-fetoprotein (AFP) and human chorionic gonadotrophin (HCG). [5]

In cases of oropharyngeal location, the prognosis depends on how deep the mass infiltrates the oral cavity of the host twin and how fast the fetus's airway is secured.

The diagnosis in this case was made retrospectively and hence led to poor management of the neonate.

Prenatal diagnosis is a useful tool to identify well defined organs in the fetus in fetu, help to guide the ex-utero intrapartum management procedure for the new-born fetus in fetu or offer the reasonable option of termination of pregnancy for severe cases.

The patient booked early at the local clinic at Rustenburg for antenatal visit. Her age classified her as an advanced maternal age, and the timeous referral was mandatory.

The patient could have benefited from an early fetal anomaly scan at a tertiary hospital. Multidisciplinary approach to this case could have been planned.

Ling Wang et al reported a case of a 28 years old primi-gravida patient, with oropharyngeal fetus in fetu, discovered at 16 weeks during a routine sonographic examination. A nonhomogeneous mass of 32mm x 27mm x 28mm protruding out of the mouth was identified in foetus (figure 3), with the lowest part of the mass located in the foetal chest and upper part in the foetal oral cavity. Parents opted for termination of pregnancy. [4]

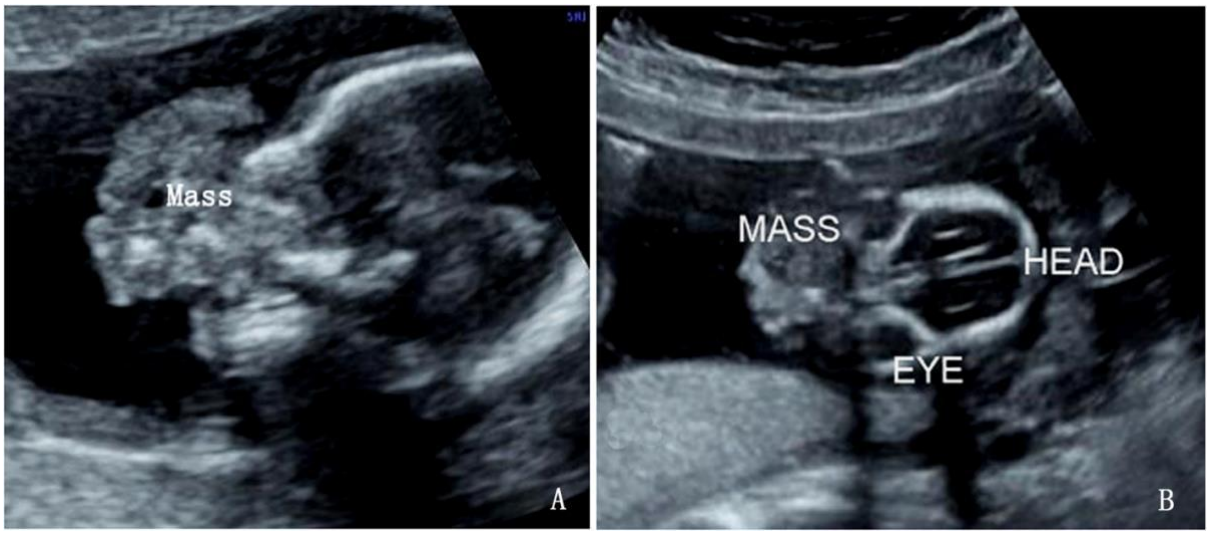

Figure 3: A nonhomogenous mass protruding out of the mouth, identified in the foetus oropharyngeal section. A sagittal view, B Coronal View.

Ex utero intrapartum (EXIT) procedure would have been the better option for the baby but again it required an accurate antenatal diagnosis of the condition and a proper multidisciplinary team management planning. Multidisciplinary team composed of Obstetricians, Paediatrics, Paediatrics surgeons, Anaesthesiologists, and Ear and Nasopharyngeal-Throat (ENT) specialists.

The EXIT procedure is a surgical procedure used for airway control in variety of entities, including extrinsic (teratomas, lymphangiomas) and intrinsic (laryngeal atresia, congenital upper airway obstruction) obstructive malformation of the upper airways. Interventions performed with the EXIT procedure include intratracheal intubation, tracheotomy, tracheoplasty, and ablative surgery for tumours. [6]

4. CONCLUSION

Fetus in fetu remains a challenging and rare condition. The need for timeous referral to tertiary hospital for a multidisciplinary approach and treatment is mandatory.

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