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Case Report

Acardiac twin presenting as obstructed labour: A case report

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Abstract: The twin reversed arterial perfusion (TRAP) sequence is a rare complication unique to multiple pregnancies. Here we report a case of a 24 year old primigravida who delivered first twin at a peripheral hospital and was referred as a case of Obstructed labour of second twin with breech presentation. No USG was done throughout her antenatal period. She underwent caesarean section and delivered an Acardiac Twin at our hospital. The first twin was a female baby of 2.4kg. Mother and the first baby were discharged healthy after 7 days. With early diagnosis by USG and proper antenatal care this would have been prevented.
Keywords: Acardiac twin, Obstructed Labour, Caesarean Section, Ultrasound.

INTRODUCTION

Multifetal pregnancies represent a potentially perilous journey for the mother and her unborn children[1].The incidence of almost all the complications of pregnancy is increased with an increase in both maternal and fetal morbidity and mortality. The development of the acardiac anomaly is a rare complication of monozygotic multiple pregnancies. Acardiac fetuses were first described by Benedetti in 1533[2]. It occurs in 1% of monochorionic twin pregnancies and in 1 in 35,000 pregnancies[3].TRAP sequence is characterized by a structurally normal pump twin perfusing an anomalous twin. The acardiac twin is transfused by the normal co twin by means of reversal of circulation through large vein to vein and artery to artery anastomosis and has no direct communication with placenta. The outcome is 100% fatal for the acardiac twin with a 50–75% mortality of the normal pump twin. Proper timing of the delivery is of prime importance for the survival of the normal fetus. The risk of recurrence was estimated 1:10,000[4].

Multi fetal pregnancies should be meticulously followed up at a tertiary care centre for early identification and treatment of such complications. But still in the rural areas of our country the maternal health is neglected and we see such complications at advanced stages of pregnancy. We here present a case of an acardiac twin presenting as obstructed labour and diagnosed at the time of delivery.

CASE REPORT

A 24 yr primigravida was referred to our hospital at 38 wks of pregnancy after delivering the first twin at a peripheral hospital. The baby was a female weighing 2.4 kg with no visible anomaly (figure.1).Delivery of the first twin was uneventful. After the delivery of the first twin, a globular mass was felt in pervaginal examination and there was no progression of labour even with adequate uterine contractions. Early features of obstructions started developing. They diagnosed the case as breech presentation of second twin with early features of obstructions and referred it to our hospital. The patient had two antenatal check ups without having any USG scan. On clinical examination, patient was dehydrated and was having tachycardia. Her uterus was 34 weeks size with regular contractions. Cord was seen outside the vagina which was clamped. On P.V. examination, vagina was dry and hot, liquor was not draining. Cervix was effaced and fully dilated. Cord was felt coming out of vagina. A globular edematous structure was felt which mimic breech. Emergency USG was done in labour room which showed abnormal mass with difficulty in visualization of the fetal parts and gender. First twin was sent to NICU. Patient was resuscitated with I.V. hydration, antibiotics and was prepared for emergency caesarean section for the obstructed labour. Second twin was delivered(figure.2). It was grossly abnormal with no differentiation of the body parts. It looked like a mass weighing 3.5 kg which was divided in to 3 parts. One small round representing the head

with no eyes &nose with small skin tags on both sides representing the ears and one larger elongated part representing the rest of the body with no clear differentiation of chest, abdomen, pelvis or genitalia and there was presence of a single lower limb. Placenta was 500 grams. Two Umbilical cords were present. The normal twin cord was longand edematous, twin B had a short cord and both shared the same Placenta. Autopsy of the acardiac twin showed that the cardiac and pulmonary tissues were absent. There was a rudimentary cranium and vertebral column and some cerebral tissue and spinal cord. In both arms and legs, some bone formation was present. Patient was transferred to ward in a satisfactory condition. Catheter was removed on 7thpost operative day. She was discharged from hospital on7th postpartum day after urination with a healthy female baby and advised for follow up.

Fig-1: First twin delivered at periphery

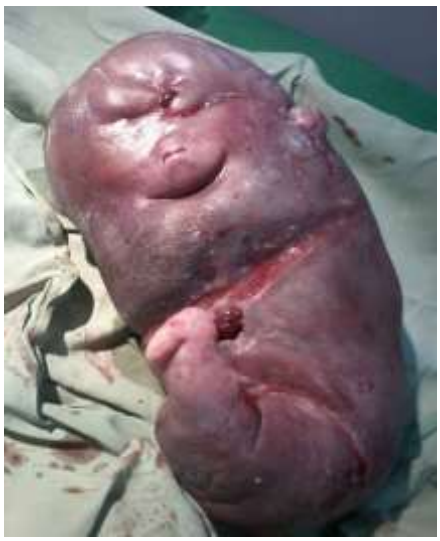

Fig-2 : Second Acardiac twin delivered at our hospital

DISCUSSION

TRAP sequence which stands for twin reverse arterial perfusion sequence is a rare complication of monochorionic twin pregnancy. The definition of the TRAP sequence was given by Van Allen *et al.* [5], who clarified the pathophysiological aspects of the condition. In this, there is a reversed perfusion from pump to acardiac twin *via* single anastomosis. This single vein-to-vein or arterial-to-arterial connection is between the two cords or indirectly through the chorionic plate.

The arterial perfusion pressure of the donor twin exceeds that of the recipient twin. Thus the donor twin receives reverse blood flow of deoxygenated arterial blood from its co-twin. This used blood reaches the recipient twin through its umbilical arteries and preferentially goes to its iliac vessels. Thus only the lower body is perfused and disrupted growth and development of the upper body results with well formed lower part of the body[6].

The anomalies in acardiac twin can be in the head(partial/total absence of cranial vault, holoprosencephaly), in face(absent facial structures, anophthalamia, microophthalmia, cleft lip, cleft palate), in the limbs(absent or rudimentary limbs), in viscera (diaphragmatic defects, absent lungs & heart, oesophageal atresia, ascites, absent liver & gall bladder), ventral wall defects, oedema of the skin, single umbilical artery, markedly different sized umbilical artery, inconsistent membrane development between the twins and occasionally umbilical artery drawn directly into superior mesenteric artery[7,8]. In our case the acardiac twin had poorly formed head with indistinguishable facial structures, rudimentary spinal cord and limbs.

There are different types of acardiac twin according to the degree of abnormal development of cephalic and truncal portions as proposed by Bianchi *et al.[9]*. The first type is acardius-acephalus, where no cephalic structures present. The second is acardius-anceps where some cranial structure and/or neural tissue present. Our case may belong to this category. The third is acardius-acormus with cephalic structure but no truncalstructures present. The fourth type is acardiusamorphus with no distinguishable cephalic or truncal structure[10]. As proposed by Malone &D' Alton, the acardiac twin can also be classified as hemiacardius in cases where the heart is incompletely formed and holocardius in cases where the heart is absent[11].

Management of acardiac fetus is a challenge as the continuous growth of the acardiac fetus is deleterious to the healthy pump twin. It can lead to complications such as cardiac insufficiency, polyhydramnios, prematurity and even death of the structurally intact twin in up to 50% of cases [12].

Twin reversed arterial perfusion(TRAP) cannot be prevented but an early diagnosis of this disorder can possibly save the donor twin. This can be detected in the early stage of pregnancy by ultrasound scanning and Doppler velocimetry [13]. The sequence can manifest as early as 11 weeks of gestational age [14]. First trimester scan is necessary to determine chorionicity and is preferably done before 14 weeks gestational age (100% sensitivity, 99% specificity) [15] as still the amniotic membrane is separate from the chorion. Our case went to a peripheral hospital for antenatal checkups and did not get even a single USG done.

Treatment is minimally invasive intervention to occlude vascular supply to the acardiac twin through cord occlusion techniques or intrafetal ablation[16]. Cord occlusion can be done by embolization, cord ligation, laser coagulation, bipolar diathermy, and monopolar diathermy, while intrafetal ablation has been performed with alcohol, monopolar diathermy, interstitial laser, and radiofrequency ablation. Recently High intensity focused ultrasound(HIFU) has been described as a non-invasive treatment for occlusion of blood flow in the acardiac twin [17].

Sepulveda W, et al. advocated expectant management in all cases [18]. They suggested use of close fetal surveillance with serial ultrasound. It is important for detecting any worsening of the condition, which may suggest the need for interventions to optimize the pump-twin's chance for survival.

This case have been reported to highlight the dangers inherent in long term neglect of a twin pregnancy and the need to educate women especially in developing countries on the importance of seeking medical help early. In our case it was not even diagnosed as a case of twin pregnancy until term because of lack of diagnostic facilities at the peripheral hospital and the negligent attitude towards female health in rural areas.

CONCLUSION

Early diagnosis and management decreases maternal morbidity and increases the survival chance of the donor twin. In developing countries like India proper health education, awareness regarding high risk cases like twin pregnancy, availability of diagnostic modalities should be there upto grass root level. In our case although the donor twin survived but the Caesarean delivery would have been avoided if it had been diagnosed earlier.

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