Epigastric Heteropagus: A Case Report
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Citation

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Abstract
Purpose: To report a cephalus epigastric heteropagus case and its surgical issues.

Case Report: A thirteen day old full term male baby was brought to the neonatal emergency unit with an attached disformed twin. A parasitic twin with an hydrocephalic head and a rudimentary trunk with limb buds and a rudimentary lower limb was attached to the epigastrium. The parasitic hydrocephalic head contained a dysmorphic face with anophthalmia and dysgenesic ears. There was a supra-umbilical evagination without omphalocele. No heart activity was found in the parasite.

Separation was performed at day 26 with parietal refection, leaving evagination for a subsequent procedure. The parasite was 960g and exploration showed one small bowel without any other organ.

Postoperative care was conducted at the neonatal intensive care unit. The baby was fed the same day. The course was complicated by a skin infection treated by local care.

The patient was discharged from the hospital on day 53 weighing 2.45 Kg after skin healing and satisfactory weight gain. Eventration care was scheduled to about 2 years.

Conclusion: We describe an extremely rare anomaly which is usually diagnosed antenatal. In our case the diagnosis was made upon birth. The separation succeeded with limited explorations and technical platform.

INTRODUCTION
Heteropagus or « parasitic » conjoined twins are asymmetric conjoined twins. The tissues of the severely defective twin are dependent on the cardiovascular system of the other twin (autosite) [1,2,3]. Parasitic conjoined twins are an extremely rare condition that represents 1% - 2% of all conjoined twins, and less than 0,1% in 100,000 birth [4]. When parasite’s attached to host epigastrium, anomaly is called epigastric heteropagus twins (EHT) and it is exceedingly rare [1]. We are reporting a cephalic epigastric heteropagus case and its surgical issues.

CASE REPORT
A thirteen day old full term male baby was sent from a regional hospital to the Neonatal Intensive Care Unit (NICU) of the Mother & Child Academic Hospital in N’djamena City (CHAD), with an attached, deformed twin.

The mother was 24 years old with 3 older children, apparently in good health. Pregnancy was not booked with an unassisted vertex delivery at home.

The newborn was seen at day 13 of life with a weight of 2,760 g. There was attached to the epigastrium a parasitic twin with a hydrocephalic head, a rudimentary trunk with limbs buds and a rudimentary lower limbs. The parasite hydrocephalic head contained a dysmorphic face with anophthalmia and dysgenesic ears. There was a supra-umbilical evagination without omphalocele. No heart activity was found in the parasite.

Abdominal ultrasound showed no anomaly in the host. Abdominal CT scan was not performed. The neonate was admitted to NICU where laboratory tests were performed; fluids and electrolytes balance were corrected.

A separation was done on day 26 and the surgical procedure lasted for an hour. We proceeded by circumferential incision leaving about 1,5cm of skin flap on all sides. Careful dissection led us to find a vascular trunk from the falciform ligament of the autosite including a cartilaginous bridge. The
separation was performed with parietal reflection leaving eventration for a later subsequent procedure.

The parasite weighed 960g and exploration showed one small bowel without any other organs.

Postoperative care was conducted in the NICU. The baby was fed the same day. Postoperative follow up was complicated by a skin infection which was treated with some medical ointment.

The patient discharged from the hospital on day 53 weighing 2.450 Kg after skin healing and satisfactory weight gain. Eventration care was scheduled to occur in about 2 years.
DISCUSSION

According to the literature, EHT are predominantly male. Willis reported that « such twins have an acardiac, acephalic trunk with limbs attached to the host and occurs as a result of deoxygenated blood from the host truncus, flowing into the parasite through the umbilical artery of the autosite »[5].

Other groups have argued that ischemic atrophy by itself cannot account for the pathologies seen in parasitic twins; most parasitic twins are acephalic [6].

As for our case, it was an acardiac cephalic parasite with rudimentary limbs. We did not found omphalocele as reported in 60% of cases by Gupta et al. [3].

They did not share bowel or other organs. Nasta et al. reported a case with a connexion of the parasite’s bowel with a Meckel’s diverticulum of the autosite [7]. Gupta et al. reported 2 cases with a hinge joint between the upper limb bones of the parasite and the xiphoid-sternum of the autosite [3].

In our case, the hinge joint was between the xiphoid-sternum of the autosite and the rudimentary chest of parasite.

The vascular supply to the parasite has been reported to arise from the liver, left internal mammary artery, epigastric vessel, umbilical vessels and falciform ligament [8].

No angiography was performed in our case but we discovered intra-operatively vessels from liver falciform ligament of the autosite.

Abdominal wall defects are commonly seen in omphalopagus; this is not surprising and defect corresponds to the site of attachment of the parasite [6].

Mode of delivery of heteropagus twins is only described in 34 articles from 1984 to the present; 53% of twins were delivered vaginally and 29% by caesarean. 12% of spontaneous abortion and 6% of elective termination [6].

According to Cury et al. early diagnosis, close prenatal management and a proper route of delivery will ensure the best possible outcome for the mother and affected neonates [9]. In our case, no prenatal diagnosis was made and delivery was performed at home without medical assistance. The neonate was received in our hospital only after 13 days of life.

Preoperative imaging can be quite extensive [6]. Ultrasound and echocardiography was performed in our case and showed no sharing organs and no cardiac anomaly.

Separation is granted not only on cosmetic and psychosocial grounds but also because parasites often cause respiratory distress and may restrict growth of the autosite [6]. Timing of surgery varies depending on condition of the autosite. We proceeded to a separation on day 26 of life after imaging and correcting fluids and electrolytes.

We did on one-step procedure and left the eventration for a subsequent procedure.

CONCLUSION

We describe an extremely rare anomaly which is usually diagnosed antenatal. In our case the diagnosis was made upon birth. The separation succeeded with limited explorations and technical platform.
References

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