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Case report

Successfully separated conjoined twins at French Medical Institute for Mothers and Children Kabul Afghanistan

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ARTICLE INFO	A B S T R A C T
Keywords: Conjoined twins Afghanistan Omphalopagus Complication	Conjoined twins are a rare malformation and few cases have been reported in the literature. There are different types of conjoined twins and the most common type is the thoracopagus. Prognosis is related to the type of conjoined twins and the organs they share. We are reporting an Omphalopagus, in which there was sharing of the liver, sternum, and abdominal wall. The complications in this kind of cases are Bleeding, Bile leakage, and infection.

1. Introduction

The incidence of conjoined twins is rare. If the split happens after embryonic disk formation (after 12th days of conception), and the division is incomplete, it leads to conjoined twins formation [1,2].

This situation happens in 1% of monozygotic twin pregnancy [2], with the incidence of 1.5/50000 to 1/500000 births with a 3:1 male to female ratio [3]. Conjoined twins can be divided into two main categories: parasite and symmetrical. The symmetrical conjoined twins could be attached whether ventrally, laterally, caudally, or dorsally [3]. It is important to diagnose conjoined twins as early as possible. Ultrasonography and magnetic resonance imaging (MRI) are effective modalities for diagnosing early in pregnancy [3]. Based on the type of attachment and organs that are involved, different treatment policies can be considered. In mild cases, surgical separation would be helpful, in contrast in more severe cases termination of pregnancy as soon as possible would be a wise choice [1,3]. There are a variety of short-term and long-term complications associated with separation surgery [3]. Based on Surgical Case Report, 2020 (SACRE) guidelines, we discussed a case report of symmetrical 9 days old omphalopagus twins with attached liver who underwent successful surgical separation [4].

2. Case presentation

A 35 years old housewife multigravida (G5P4L4) from Herat, Afghanistan went to the office for a prenatal checkup. In obstetrical history, all 4 previous children were delivered vaginally. They were all full-term and healthy with a birth interval of 1.5 years. The youngest one was 5 years and 6 months old at the time of visit. Her LMP was 2020.8.13. She had a family history of twin pregnancy in her mother, twins died on the first day of birth. Her past medical and social history were unremarkable. Serial prenatal check-up sonographies all showed normal twin pregnancy. Delivery was done at 2021.5.6 by cesarean section. The omphalopagus twins (Fig. 1) were 9 days old when they were brought to the children's hospital. Upon their arrival to the emergency room (ER), their vital signs were normal and their combined weight was 4 kg. The twins were admitted to NICU. In abdominal X-ray conjoined twins with the fusion of anterior wall was seen and no abnormal opacity was noted in the region of kidneys, ureters, and urinary bladder. In abdominal CT scan livers were not separable and a vessel seen coming from the hepatic region of baby A to baby B likely portal vein branch and a separate biliary duct drainage system was seen. There was a connection at the sternum (Fig. 2). Blood investigation of each neonate is demonstrated in Table 1.

The only abnormality was hyperbilirubinemia in both twins that

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Fig. 1. Photo show the omphalopagus twins.

phototherapy, phenobarbital, and ursodeoxycholic acid were started. The BLCS was also sent in which baby B culture had no growth but baby A culture was positive for staphylococcus to whom antibiotics (piperacillin-tazobactam + vancomycin) was started. Twins with the diagnosis of Thoraco-Omphalopagus went under surgery on the sixteenth day of birth (Fig. 3). Patients anesthetized and controlled by two consultant anesthesiologists with different anesthesia systems and controlled regularly according to conjoined twins' anesthesia protocol. Patients Prep done from Knee of both children till nipples front and back, they covered accordingly too. It was also decided to start and finish the procedure over the same table to prevent post-surgery infection related to transferring one child to another operating table. Children had continuity of liver to each other and no border between livers was determined. There was a strong connection at sternum; Diaphragms had a defect at sternum level and had connectivity to each other; also diaphragm had a connection at one point with pericardium too. The connections were separated and both abdomens were closed. During surgery, both of them were transfused with 30 ml of packed RBC s. The surgery took about 6.5 h which performed by attending pediatric surgeon, and finished with very little bleeding (Fig. 3) and the twins were shifted to NICU intubated. Children extubated on the second day of surgery which they tolerated well with only 0.2 l/min oxygen demand and on the third day feeding of 2 ml/3 h started by nasogastric tube and was increased to 10 ml/3 h which tolerated well. After 4 days of operation, children shifted to normal feeding and were discharged after observation for one week. Their weight on the day of discharge was 3 kg each. They had two outpatient visits a week later and then left Kabul to Herat and are under online follow-up.

3. Discussion

Conjoined twins are among the rarest phenomenon. There are different types of conjoined twins. The most common type is thoracopagus(42%), while piopagus twins are the least common (1%). The rest of conjoined twins types based on anatomy are parapagus dicephalous (11.6%), cephalopagus (5.5%),omphalopagus (5.5%), craniopagus (3.4%), parapagus diprosopus (2.9%), ischiopagus (1.8%) and

rachipagus (1%). About 24.1% of conjoined twins cannot be categorized anatomically (unspecified) [2]. Conjoined twins can be diagnosed as early as 8 weeks of gestation [5]. Ultrasonography is the first mean for diagnosing conjoined twins in the first trimester [2]. Performing MRI is important since it provides crucial information about anatomy and prepares the surgical team for better separation [3]. Analyzing symmetrical ventrally attached twins with MRI is of high importance because it supplies significant information about the liver, the most involved organ, as in our discussed case. It is also very important to make the situation of the cardiac system obvious [3,6]. With the advent of 3-dimensional printing, surgical planning and organ orientation have improved [7]. Applying separation surgery depends on the condition of patients; Emergency separation is indicated when one twin is dead or dying and is threatening the life of another twin. Surgery is not suggested when there is a complex cardiac fusion. Elective surgery is indicated when the twins are stable and there is enough time to evaluate the fusions by imaging [8].

It is important to bear in mind that diagnosing conjoined twins may put parents under psych-social pressure [3].

Furthermore, prenatal management of attached twins could be religiously and ethically challenging [1,3].

The prognosis of conjoined twins depends on concomitant anomalies and the extent of the fusion of structures and abnormal vascular connections [9]. The experience of the surgery team, use of tissue expanders, and antenatal imaging can improve the prognosis [10,11]. Only about 60% of conjoined twins who undergo separation surgery survive [12].

One of the most significant short-term complications is infections which were not observed in any of our twins. Prolonged treatment and wound complexity rise the chance of infection. Long-term complications include associated malformations [3].

4. Conclusion

We reported an omphalopagus twin, who were brought to the hospital after birth. There was a vessel that connected twins' livers. In addition, their sternums and diaphragms were attached. The discussed twins were successfully separated with no short-term complications.

Funding

None.

Ethical approval

This is a case report paper.

Consent

Informed consent was obtained from the patient, for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal upon request. Informed consent was obtained from the patient's legal guardian (his mother and father), for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

AT, MAS, NB conceived and designed the study and wrote the manuscript; SHM, AA helped collect data; AA wrote the manuscript; SHM and NB confirmed the eligibility of the participants' for the study; MAS Supervised the whole study and approved the final version of the manuscript.



Fig. 2. The abdominal slice of CT-scan of twins revealed a connection from the sternum.

Table 1

This table illustrate	s blood	investigations	of baby	A and	baby	в
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Blood investigations	Neonate A	Neonate B	Normal range	
c-Reactive protein	3	0	Normal <10 mg/L	
Blood group	A^+	A^+	-	
Magnesium	2	1.4	1.7–2.8 mEq/L	
BUN	5	?	6–20 mg/dL	
Creatinine	0.4	0.5	0.65–1.1 mg/dL	
Na	135	137	135–145 mEq/L	
K	4.5	4.1	3.6–5 mEq/L	
Total bilirubin	14.6	9.8	0.1–1 µmol/L	
Direct bilirubin	0.4	0.3	0–0.2 µmol/L	
Indirect bilirubin	14.2	9.5	0.1–0.8 µmol/L	
Gamma-glutamyl transferase	83	92	58.5-254.9	
Alanine aminotransferase (ALT)	27	34	7–56 Unit/L	
Alkaline phosphatase	235	119	42–121 Unit/L	
Ca	8.9	8.5	8.4–10.2 mg/dl	
Hemoglobin	15.1	15.8	14.5–22 g/dL	
Leukocyte count	9.2	11.1	7–23 /mcL	
Platelet	439	484	1140 10 ^{^3} to 400 10 ^{^3}	
Prothrombin time (PT)	12.5	12.5	<14.5 Sec	
Partial prothrombin time (PTT)	12	12.2	Normal 23 s to 35 s	
International normalized ratio (INR)	0.9	0.9	Normal 0.8 to 1.1	



Fig. 3. The photos babies during and after separation via surgery.

Registration of research studies

Not applicable.

Guarantor

Sayed Hamid Mousavi, the corresponding author, accepted full responsibility for the work and/or the conduct of the study, had access to the data, and controlled the decision to publish.

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Declaration of competing interest

The authors report no declarations of interest.

References

- M. Kapoor, N. Sachdev, M. Agrawal, Conjoined twins [cited 2021 May 16], J. Obstet. Gynecol. 63 (1) (2013 Mar) 70–71. Available from: https://pubmed.ncbi. nlm.nih.gov/24431606/.
- [2] N. Gica, N. Gana, C. Mat, A.M. Panaitescu, G. Peltecu, A.M. Vayna, Conjoined twins—early prenatal diagnosis [cited 2021 May 16], J. Obstet. Gynaecol. (Lahore) 40 (5) (2020 Jul 3) 723–724. Available from: https://pubmed.ncbi.nlm.nih.gov/ 31607199/.
- [3] P. Teixeira Castro, H. Werner, A.P. Matos, P. Daltro, E. Araujo Júnior, Symmetric and ventrally conjoined twins: prenatal evaluation by ultrasound and magnetic resonance imaging and postnatal outcomes [cited 2021 May 16], J. Matern. Neonatal. Med. 34 (12) (2019). Available from: https://pubmed.ncbi.nlm.nih.gov/ 31387430/.
- [4] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, A. Thoma, A.J. Beamish, A. Noureldin, A. Rao, B. Vasudevan, B. Challacombe, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 1 (84) (2020 Dec) 226–230.
- [5] X.-W. Liang, Y.-Y. Cai, Y.-Z. Yang, Z.-Y. Chen, Early ultrasound diagnosis of conjoined twins at eight weeks of pregnancy: a case report [cited 2021 May 16], World J. Clin. Cases 8 (21) (2020 Nov 6) 5389–5393. Available from: https:// pubmed.ncbi.nlm.nih.gov/33269274/.

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- [6] K. McHugh, E.M. Kiely, L. Spitz, Imaging of conjoined twins [cited 2021 May 16], in: Pediatric Radiology, Pediatr Radiol, 36, 2006, pp. 899–910. Available from: https://pubmed.ncbi.nlm.nih.gov/16622666/.
- [7] J.A. Villarreal, D. Yoeli, P.M. Masand, Galvan NTN, O.O. Olutoye, J.A. Goss, Hepatic separation of conjoined twins: operative technique and review of threedimensional model utilization [cited 2021 May 16], J. Pediatr. Surg. 55 (12) (2020 Dec 1) 2828–2835. Available from: https://pubmed.ncbi.nlm.nih.gov/32792165/.
- [8] L. Spitz, E.M. Kiely, Experience in the management of conjoined twins [cited 2021 May 16], Br. J. Surg. 89 (9) (2002) 1188–1192. Available from: https://pubmed. ncbi.nlm.nih.gov/12190687/.
- [9] Early prenatal diagnosis of conjoined cephalopagus twins PubMed [Internet] [cited 2021 May 16]. Available from, https://pubmed.ncbi.nlm.nih.gov/ 12901255/.
- [10] L. Spitz, M.D. Stringer, E.M. Kiely, P.G. Ransley, P. Smith, Separation of brachiothoraco-omphalo-ischiopagus bipus conjoined twins [cited 2021 May 16], J. Pediatr. Surg. 29 (4) (1994) 477–481. Available from: https://pubmed.ncbi.nlm. nih.gov/8014797/.
- [11] Conjoined twins in Cameroon: issues inherent in diagnosis and management in the African context - PubMed [Internet] [cited 2021 May 16]. Available from, https:// pubmed.ncbi.nlm.nih.gov/20690268/.
- [12] [Conjoined twins: prenatal diagnosis, delivery and postnatal outcome] PubMed [Internet] [cited 2021 May 16]. Available from, https://pubmed.ncbi.nlm.nih.gov/ 21860927/.