ECRONICON OPEN ACCESS

EC PAEDIATRICS
Case Report

Gastroschisis in a Premature Infant: A Case Report and Review of Literature

Volkan Sarper Erikci[1]*, Mehmet Mert[1], Tunç Özdemir[1] and Gökhan Köylüoğlu[2]

[1]Departments of Pediatric Surgery, Tepecik Training Hospital, Sağlık Bilimleri University, Izmir, Turkey

[2]Departments of Pediatric Surgery, Tepecik Training Hospital, Katip Çelebi University, Izmir, Turkey

***Corresponding Author:** Volkan Sarper Erikci, Departments of Pediatric Surgery, Tepecik Training Hospital, Sağlık Bilimleri University, Izmir, Turkey.

Received: February 26, 2018; **Published:** April 09, 2018

Abstract

Abdominal wall defects are rare anomalies and gastroschisis (GS) is relatively common with respect to omphalocele. The mortality has decreased over the years but morbidity still remains high. GS provides a considerable challenge to the pediatric surgeon. In this report management of a preterm infant with GS using Bianchi repair is presented and discussed under the light of relevant literature.

Keywords: *Gastroschisis; Morbidity; Surgical Treatment*

Introduction

Abdominal wall defects are rare anomalies. Gastroschisis (GS) is relatively common with an incidence rate of ranging from 1/4000 to 4 - 5/10000 live births [1,2]. It is also stated that there is a high prevalence rate of gastroschisis in babies of young mothers [3]. Although the mortality has decreased over the years from 90% to 10%, morbidity still remains high and GS continues to provide challenge to pediatric surgeon dealing with these babies [4-6].

There are multiple reports of different techniques of GS closure. These are primary closure with umbilical preservation [7], elective delayed midgut reduction without anesthesia as described by Bianchi and Dickson [8,9], delayed repair with a preformed silo [10]. In this report, the management of a preterm infant with GS using Bianchi repair is presented and discussed under the light of relevant literature.

Case Report

A 1720-g pre-term female infant at 36 weeks gestational age was admitted to our neonatal intensive care unit (NICU) with a diagnosis of GS. Elective cesarean delivery was performed in the 18-year-old G1P1 otherwise healthy mother. Antenatal history revealed evisceration of abdominal viscera out of the abdominal cavity compatible with GS. After transfer to our center, the baby was inspected and entire gastrointestinal tractus including stomach, small bowel, entire colon, both tuba uterinas with uterus were found to be eviscerated (Figure 1). The defect diameter was 2.0 cm right to the umbilicus. The herniated organs were covered with sterile gauze bandages soaked in warm saline during the transport of baby to our NICU. Before attempting Bianchi reduction, a nasogastric tube was inserted into the stomach for evacuation of gastrointestinal secretions proximally apart from intravenous fluid resuscitation with the use of broad spectrum antibiotics. A rectal tube was also inserted into the rectum with much return. After sterile preparation and draping, reduction of eviscerated viscus into the abdominal cavity without anesthesia using Bianchi technique was performed. The technique itself took 15 minutes and at the end of the procedure, a single purse-string suture using nonabsorbable (3/0 polypropylene) material was inserted to close the abdominal defect (Figure 2). During her stay in the NICU, there was no need for mechanic ventilation. Following this reduction, the baby

did well and received total parenteral nutrition (TPN) and she began oral intake 12 days after reduction. During her follow-up in NICU, a gradual increase in the abdominal distention and failure to defecate despite rectal irrigations was observed and oral intake was stopped and parenteral nutrition was restarted using a central venous line. Direct roentgenogram of the baby taken at this stage, demanstrated a distended abdominal cavity with dilated loops of small bowel suggesting no passage of feces from ileum into the colon distally (Figure 3). There was no resolution abdominal distention and a laparotomy was performed on the 38th postoperative day with the diagnosis of intestinal obstruction. Intraoperatively, in addition to severe distention of the small bowel, diffuse adhesions between the abdominal wall and loops of small intestine and multiple adhesions between the intestinal loops were detected and adhesiolysis was performed (Figure 4). During the examination at surgical intervention, there was no atretic intestinal segment in the whole gastrointestinal tractus. But the colon was found to be small caliber in diameter totally with respect to small bowel and multiple levelling colonic biopsies for a possible diagnosis of Hirschsprung's disease and incidental appendectomy were performed. Due to huge diameter discrepancy between the small bowel and colon impairing the passage of feces distally, a loop ileostomy was performed. Early gastrointestinal feeding was started on the first postoperative day. Histopathological examination of the excised specimens did not reveal Hirschsprung's disease. Now the patient is at follow-up and has a steady increase in body weight and ileostomy closure is planned to be performed.

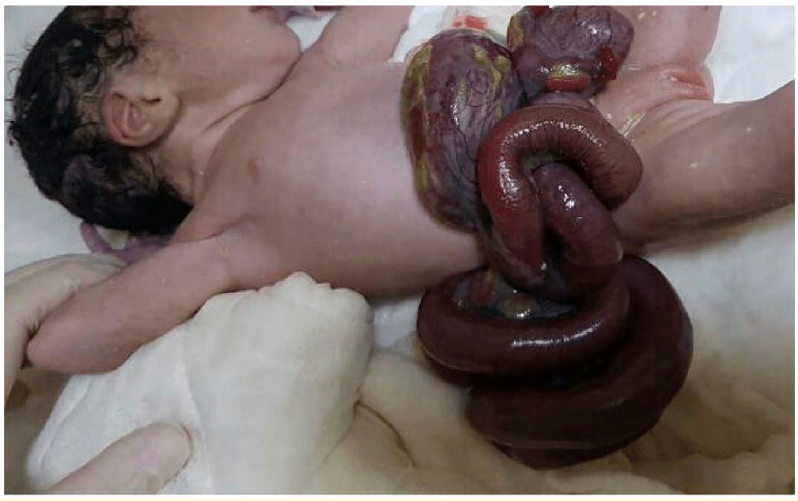

Figure 1: The premature infant with gastroschisis.

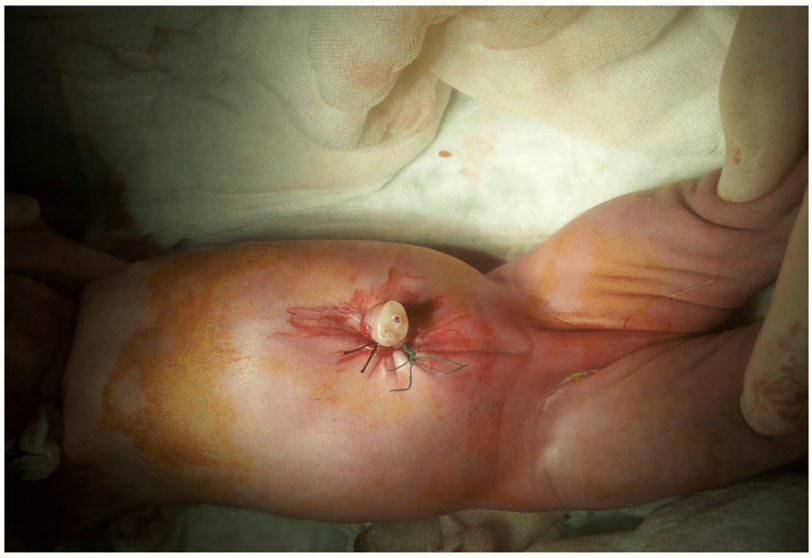

Figure 2: *Postoperative view following Bianchi procedure.*

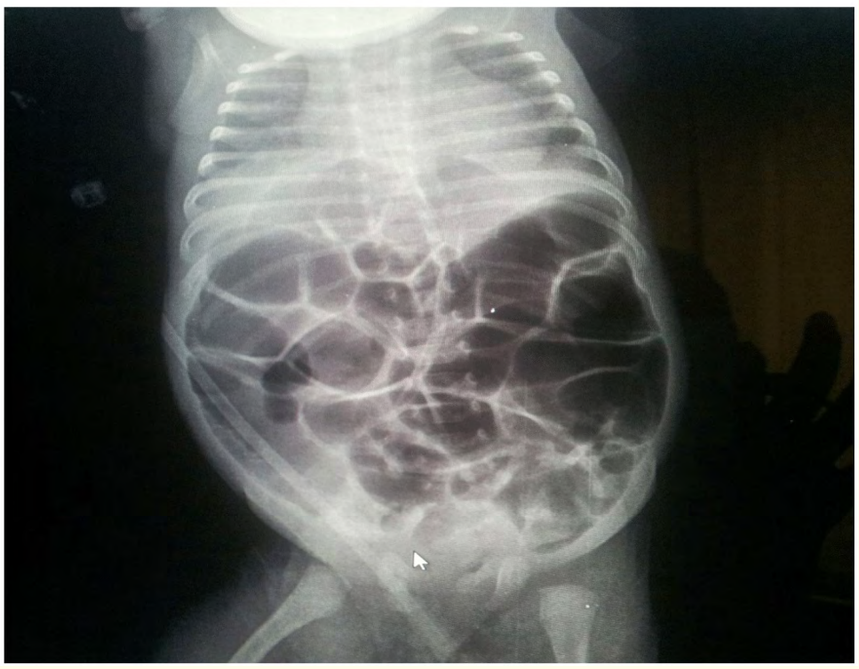

Figure 3: *Abdominal X-ray showing abdominal distention with multiple dilated intestinal loops.*

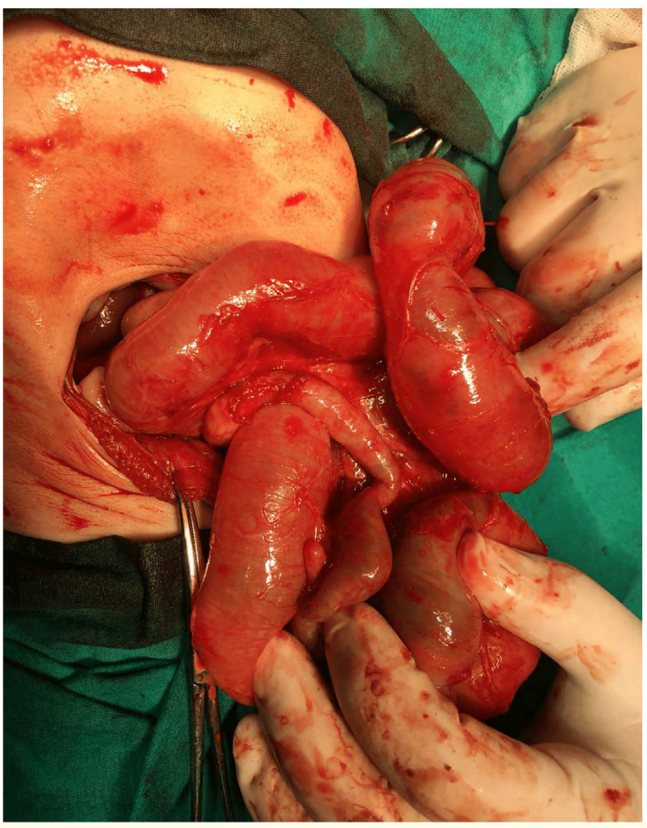

Figure 4: *Operative view after adhesiolysis. Note there is diffuse distention of small bowel and the colon totally. small in caliber.*

Discussion

Gastroschisis is one of the most common anterior abdominal wall defects. It has been reported that unlike omphalocele, the incidence of GS has been increasing consistent with regional reports form Australia and other developed countries [11-17]. Young maternal age has been identified as a risk factor for gastroschisis and with a maternal age of 18 years, the presented case in this report is in accordance with literature [3]. Despite the mortality rate is decreasing considerably, concerning the morbidities that may be seen during the postoperative management of these babies, GS provides a considerable challenge to the pediatric surgeon. GS is one of the major causes of the short bowel syndrome in infancy and childhood and it is also a major cause of prolonged, costly, complicated, and hazardous NICU stays with requirements for TPN. GS may present as simple or complicated. When complicated patients with GS present with intestinal atresia/s, stenosis, necrosis, perforations. Although it did not appear to be a microcolon, due to the diminished luminal caliber in the whole colon, the presented case has been considered as a complicated GS and a loop ileostomy was performed after closure of the abdominal wall defect for intractable failure of defecation.

Morbidity is a major problem in certain amount of patients with GS [18]. The most common causes of morbidity include a very low gestational age, presence of accompanying additional anomaly, abdominal compartment syndrome after closure of the abdominal defect and necrotising enterocolitis development [19,20]. Although there was no associated anomaly, prematurity with low birth weight together with diminished total colonic luminal caliber seems to be the major factor of morbidity in the presented case. Intestinal dysmotility after reduction of eviscerated viscus is another cause of morbidity and there are conflicting reports on the return of intestinal motility in GS. It is suggested that there is no difference between preterm and term births of patients after surgical treatment with respect to intestinal function [21]. However other studies report that preterm and low birth weight infants with GS need longer time to start oral intake and longer hospitalization [5].

Regarding the surgical treatment of GS, there are different techniques in the closure of the abdominal wall defect. These are namely immediate operative closure, elective delayed midgut reduction without anesthesia as described by Bianchi, delayed repair with a preformed silo [8,10]. In a survey of EUPSA delegates in Europe on the management of GS, it has been stated that primary closure was found to be the preferred choice of surgical treatment and was achieved by operative closure in the majority of patients rather than by Bianchi technique [22]. It has also been reported that staged reduction and closure was less popular and was achieved by custom-made silo or preformed silo followed by surgical closure [22]. Although primary fascial closure is currently the preferred method of GS closure across Europe, reduction of eviscerated viscus and closure with Bianchi technique was feasible in our patient and Bianchi reduction is our first choice of treatment in these sick babies.

There are conflicting reports on the delivery method of babies with GS. It has been suggested that prenatally diagnosed patients with GS can be delivered vaginally [23]. Other studies also failed to document any advantage of routine cesarean delivery in GS [24-26]. On the contrary to these reports, it has been recommended that cesarean delivery before 36 weeks allowed earlier enteral feeding and was associated with less complications and higher incidence of primary closure with statistical significance [27]. No matter which type of delivery and the surgical treatment to be performed, once the baby is born, for the avoidance of venous congestion of the eviscerated bowel due to the tight abdominal wall defect producing vascular compression, there should not be a delay for closure of the abdominal wall defect.

Conclusion

In conclusion, our case suggests that reduction of eviscerated viscus with Bianchi technique in GS without anesthesia appears safe and it is our preferred first choice of treatment for children with GS. If development of respiratory distress and metabolic acidosis occurs after reduction of viscera into the abdominal cavity, conversion to a staged reduction with silo avoids serious bowel injury. Delayed acquisition of intestinal function is the major morbidity that may prompt the attending surgeon even for a performance of temporary proximal intestinal ostomy. Morbidities in the management of GS should be kept in mind and should be treated accordingly.

Bibliography

1. Schwartz MZ. "Gastroschisis pediatric surgery". In: Puri P, Höllwarth M, editors. Berlin: Spring-Verlag (2006): 161.

2. Holland AJA., *et al*. "Gastroschisis: an update". *Pediatric Surgery International* 26.9 (2010): 871-878.

3. Rittler M., *et al*. "Gastroschisis and young mothers: what makes them different from other mothers of the same age?" *Birth Defects Research Part A: Clinical and Molecular Teratology* 103.6 (2015): 536-543.

4. Swift RI., *et al*. "A new regime in the management of gastroschisis". *Journal of Pediatric Surgery* 27.1 (1992): 61-63.

5. Driver CP., *et al*. "The contemporary outcome of gastroschisis". *Journal of Pediatric Surgery* 35.12 (2000): 1719-1723.

6. Vegunta RK., *et al*. "Perinatal management of gastroschisis: analysis of a newly established clinical pathway". *Journal of Pediatric Surgery* 40.3 (2005): 528-534.

7. Olsen L., *et al*. "Gastroschisis: primary closure is possible in most newborns". *Annals of Surgery* 203 (1992): 512-515.

8. Bianchi A and Dickson AP. "Elective delayed reduction and no anesthesia: "minimal intervention management" for gastroschisis". *Journal of Pediatric Surgery* 33.9 (1998): 1338-1340.

9. Bianchi A., *et al*. "Elective delayed midgut reduction-No anesthesia for gastroschisis: selection and conversion criteria". *Journal of Pediatric Surgery* 37.9 (2002): 1334-1336.

10. Fischer JD., *et al*. "Gastroschisis: a simple technique for staged silo closure". *Journal of Pediatric Surgery* 30.8 (1995): 1169-1171.

11. Reid KP., *et al*. "The epidemiologic incidence of congenital gastroschisis in Western Australia". *American Journal of Obstetrics and Gynecology* 189.3 (2003): 764-768.

12. Salihu HM., *et al*. "Omphalocele and gastroschisis". *Journal of Obstetrics and Gynaecology* 22.5 (2002): 489-492.

13. Rankin J., *et al*. "Congenital anterior wall defects in the North of England, 1986-1996: occurrence and outcome". *Prenatal Diagnosis* 19.7 (1999): 662-668.

14. Tan KH., *et al*. "Congenital anterior abdominal wall defects in England and Wales 1987-93: retrospective analysis of OPCS data". *British Medical Journal* 313.7062 (1996): 903-906.

15. Hemmenki K., *et al*. "Gastroschisis and omphalocele in Finland in the 1970's: prevelance at birth and its correlates". *Journal of Epidemiology and Community Health* 36.4 (1982): 289-293.

16. Martinez-Frias ML., *et al*. "Epidemiological study of gastroschisis and omphalocele in Spain". *Teratology* 29.3 (1984): 337-382.

17. Penman DG., *et al*. "Secular rates in incidence of gastroschisis in south west of England in 1995". *British Journal of Obstetrics and Gynaecology* 105 (1998): 328-331.

18. Snyder CW., *et al*. "Early severe hypoalbuminemia is an independent risk factor for intestinal failure in gastroschisis". *Pediatric Surgery International* 27.11 (2011): 1155-1158.

19. Snyder CL. "Outcome analysis for gastroschisis". *Journal of Pediatric Surgery* 34.8 (1999): 1253-1256.

20. Blane CE., *et al*. "Gastrointestinal complications of gastroschisis". *American Journal of Roentgenology* 144.3 (1985): 589-591.

21. Soares H., *et al*. "Gastroschisis: Preterm or term delivery?" *Clinics* 65.2 (2010): 139-142.

22. Zani A., *et al*. "Is there unity in Europe? First survey of EUPSA delegates on the management of gastroschisis". *European Journal of Pediatric Surgery* 23.1 (2013): 19-24.

23. Abdel-Latif ME., *et al*. "Mode of delivery and neonatal survival of infants with gastroschisis in Australia and New Zealand". *Journal of Pediatric Surgery* 43.9 (2008): 1685-1690.

24. Puligandla PS., *et al*. "Routine cesarean delivery does not improve the outcome of infants with gastroschisis". *Journal of Pediatric Surgery* 39.5 (2004): 742-745.

25. Singh SJ., *et al*. "Gastroschisis: determinants of neonatal outcome". *Pediatric Surgery International* 19.4 (2003): 260-265.

26. Eggink BH., *et al*. "Outcome of gastroschisis: a 20 year case review of infants with gastroschisis born in Galveston, Texas". *Journal of Pediatric Surgery* 41.6 (2006): 1103-1108.

27. Hadidi A., *et al*. "Early elective cesarean deliver before 36 weeks vs late spontaneous delivery in infants with gastroschisis". *Journal of Pediatric Surgery* 43.7 (2008): 1342-1346.

Volume 7 Issue 5 May 2018