Original Article

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11-Year Experience with Gastroschisis: Factors Affecting Mortality and Morbidity

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Abstract

Objective: The aim of the study was the evaluation of patients treated with a diagnosis of gastroschisis and to establish the factors which affected the morbidity and mortality.

Methods: Twenty-nine patients, managed for gastroschisis during 2000-2010 were reviewed retrospectively. Patients were analysed in respect to gestational age, birth weight, associated anomalies, type of delivery, operative procedures, postoperative complications, total parenteral nutrition (TPN) related complications. The factors affecting mortality and morbidity were determined.

Findings: Associated abnormalities were present in 24% of the patients. Eleven patients underwent elective reduction in the incubator (Bianchi procedure) without anesthesia. Eight patients had delayed reduction with silo and ten patients had primary closure. Although the type of delivery had an effect on morbidity but not mortality, gestational age, birth weight, and the operative procedure performed had no effect on morbidity or mortality. Duration until tolerance of oral intake, and of TPN and hospitalization were found to be statistically significantly shorter in the group of babies delivered by cesarean section.

Conclusion: In our study the most important cause of mortality was the abdominal compartment syndrome and multi-organ failure in the early years. Long hospitalization periods and sepsis were the main causes of mortality in recent years.

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Key Words: Gastroschisis; Morbidity; Mortality; Neonate

Introduction

Abdominal wall defects are rare anomalies. The incidence of gastroschisisis is 4-5/10000[1]. It can occur isolated or associated with other congenital anomalies. The survival rate of gastroschisis cases is over 90% in the literature due to developments in antenatal care, neonatal intensive care and total parenteral nutrition (TPN)[2]. The mortality has decreased over the years whereas the morbidity still remains high. The aim of this study was to evaluate patients who were followed-up with a gastroschisis diagnosis between 2000-2010 and determine the factors affecting morbidity and mortality.

Subjects and Methods

Twenty-nine patients who were operated with a gastroschisis diagnosis in our clinic between 2000 and 2010 were retrospectively analysed. The

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evaluation of patients was performed in terms of gestational age, birth weight, additional anomalies, applied surgical procedures, complications, TPN duration and related complications and the factors affecting morbidity and mortality were investigated. The chi-square test was used to determine the relationship between two qualitative data. The t test and Mann-Whitney U test were used to compare averages of two independent groups. One-way ANOVA and Kruskal-Wallis analysis was used in comparing averages of more than two groups. $P<0.05$ was considered as statistical significance. The SPSS 15.0 package program was used.

Findings

There were 16 male and 13 female patients. The delivery method was cesarean section in 55.2% and vaginal delivery in 44.8%. Once admitted, the patients were taken into a heated incubator, a nasogastric catheter (NG) was inserted, and the herniated organs were covered with sterile gauze bandages soaked in warm saline. Bianchi reduction was performed in the incubator without anaesthesia when the condition of the patient was stabilized. These are the patients who deemed suitable according to their general condition, the diameter of the defect and the herniated organs after intravenous fluid replacement and antibiotic treatment. The other patients underwent primary closure or silo application. Silo established by prolene mesh or vicryl mesh was used for this procedure and the reduction was performed with daily wound dressings.

The mean age of the mothers was 20.6±3.85 (16-32) years. Average gestation was found to be 34.9±2.7 weeks (30-39 weeks). There were 15 term and 14 preterm infants. The birth weight ranged between 1340g and 3460g and the mean value was 2248±454g. Three patients were under 1500g. Duration of hospital stay ranged between 1 and 18 hours, with a mean value of 6.0±4.0 hours.

Average defect diameter was 3.0±0.65cm (2-4cm). When herniated organs were reviewed, the small intestines were outside the body in 100%, colon in 68.9%, and stomach in 37.9% of the patients while the tuba was involved in 23% of female infants. Average length of hospitalization was 37.5±40.1 (1-150) days.

Seven patients (24%) had additional anomalies. Three had a cardiac anomaly and two had intestinal atresia. One patient had congenital lobar emphysema, while one patient osteogenesis imperfecta and limb contracture (Bruck syndrome).

One of the patients with intestinal atresia was repaired during Bianchi reduction, while the other patient was primarily closed and re-opened about three weeks later to perform atresia repair.

Elective delayed reduction (Bianchi reduction) was tried in 17 patients and was successful in 11. Ten patients underwent primary repair. Two of these had elective delayed reduction but the treatment was not successful. Staged reduction with silo was performed in eight patients. All four of the eight patients who had staged reduction with silo had undergone failed elective delayed reduction before.

A second operation was needed in 12 of the patients. Seven of these had undergone silo and then abdominal closure, a resection anastomosis was used in one patient because of a stercoral fistula, and surgery was needed in three patients due to intestinal adhesions and in one patient due to delayed intestinal atresia repair. One patient was operated one more time to remove the previously implanted mesh. Five patients required a third operation. Four of these were due to intestinal adhesions, and one was for lobectomy for congenital lobar emphysema. The duration of primary closure in patients who had silo repair was 10.3±6.38 (3-21) days.

TPN was administered to the patients until they were able to tolerate oral intake. The mean duration of TPN was 38.8±40.2 days. Oral intake started after 18.5±13.1 days. Gestational age, type of surgery and birth weight of the infant under 2500g were found to be statistically insignificant regarding the time of oral intake. Similarly, duration of TPN was found to be independent from birth weight of the infant under 2500g and gestational age. Duration of patient hospitalization was also not affected by these three parameters Interestingly, time until oral intake, TPN duration

Table 1: Comparison of gastroschisis patients by morbidity

Parameter		Oral Intake (days)	TPN duration (days)	Length of hospitalization (days)
Prematurity	**Yes**	16.1	19.3	25.3
	No (term)	18.8	21.1	32.2
	***P* value**	0.7	0.7	0.4
Type of Operation	**EDR**	18.7	18.1	27.1
	PC	14.0	19.1	24.1
	S	21.4	24.2	38.0
	***P* value**	0.2	0.4	0.6
Delivery Method	**Caesarean section**	11.4	13.9	20.2
	Vaginal delivery	27.2	29.4	45.0
	***P* value**	0.01	0.03	0.007
Additional Anomaly	**Yes**	20.5	55.6	50,8
	No	17.7	32.5	39.7
	***P* value**	0.9	0.2	0.3

EDR: Elective delayed reduction, PC: Primary Closure, S: Silo

duration and length of hospitalization were significantly shorter in infants born by cesarean section than of those born by spontaneous vaginal delivery (Table 1).

Kidney dysfunction developed in the postoperative period in 14 (48.2%) patients: in seven patients who had elective delayed reduction (EDR), in five patients who had silo method, and in two patients who had primary repair. One of these patients required dialysis. Three patients died on the first postoperative day. Hepatic dysfunction developed in 12 patients. Five of them died of sepsis. TPN-related hepatic dysfunction developed in two patients who were hospitalized for a long time, and one of them died of sepsis.

Intestinal perforation developed in four patients. Two were during EDR and primary repair was performed. These two patients died due to acute renal failure in the early postoperative period within 24 hours. The other two patients had undergone primary repair and silo. Perforation developed in one due to necrotizing enterocolitis (NEC). These patients did not develop related complications in the follow-up after being operated for perforation but one of them died following the lobectomy performed for congenital lobar emphysema and the other one died of sepsis.

Ten patients died in total. The survival rate was 65.5%. Three patients died in the first 48 hours with abdominal compartment syndrome, acute renal failure, acidosis and multi-organ failure in the early period. All three of these patients under went EDR. Another patient who was in poor general condition from the first few postnatal hours and whose clinical picture deteriorated even more after the operation with silo was performed died in the early postoperative period. Sepsis and respiratory failure developed in one patient following the lobectomy operation; five patients died due to sepsis following long hospitalization, three or more operations performed because of intestinal obstruction and perforation, and requirement of long TPN duration. One of these patients had jejunal atresia and another one had intestinal perforation because of NEC.

No statistically significant difference was found between dying and living patients in terms of birth weight, additional anomaly, method of delivery in the analysis. Although mortality appears to be high with the EDR, it was not statistically significant when mortality was evaluated by procedure (Table 2).

Discussion

Mortality rates keep falling but problems related to morbidity persist in patients with gastroschisis[3]. A very low gestational age, presence of accompanying additional anomaly and NEC

Table2: Comparison of gastroschisis patients by mortality

		Alive (n=19)	Deaths (n=10)	*P* value
Premature		9	5	0.4
Term		10	5	
Birth weight (gr)		2259 (1350-2850)	2226(1340-2600)	0.5
Delivery Method	**Caeserean Section**	12	5	0.4
	Vaginal Delivery	7	5	
Type of Operation	**EDR**	6	5	0.2
	PC	8	2	
	Silo	5	3	
Additional anomaly	**Yes**	5	2	0.8
	No	14	8	

EDR: Elective delayed reduction, PC: Primary Closure, S: Silo

development in patients have been reported to affect morbidity and cause an increase in mortality[4,5].

The major morbidity is delayed acquisition of intestinal function[6]. This situation is accepted to occur as a result of intestinal fluids encountering amniotic fluid and to depend on the compression effect of the abdominal defect on the herniated organs[7,8]. Some papers report that better results were achieved as a result of less contact between amniotic fluid and intestinal structures in preterm infants with gastroschisis and support preterm birth for these babies[9]. There are studies suggesting no difference between preterm and term births of patients with gastroschisis[10] while other studies report that preterm and low birth weight infants with gastroschisis need longer time to start oral intake and longer hospitalization[11]. Prematurity was present in 48.2% of our patients treated for gastroschisis. The survival rate was 64.2% in premature patients. The effect of gestational age on mortality and morbidity was not determined in our cases. We also found the very low birth weight of our patients to have no effect on morbidity and mortality. One patient had intestinal perforation due to NEC and died of sepsis. The NEC incidence was low in our patient group.

Even though there are various opinions on the delivery method of babies with gastroschisis, the general opinion is that prenatally diagnosed patients with gastroschisis can be delivered by vaginal delivery[2]. However, we can conclude that delivery with cesarean section significantly affects morbidity as a result of the evaluation of patients born by cesarean section and vaginal delivery in terms of morbidity. There was no difference between gestational ages of infants born by cesarean section or vaginal delivery in our series but a significant difference was found in infants born by cesarean section in terms of duration of oral intake, length of hospitalization and TPN duration. This finding supports other authors recommending infants diagnosed with gastroschisis to be delivered by cesarean section.

The mortality rate of our patients is higher than that in the world literature. Prior to 2005, the development of abdominal compartment syndrome and multi-organ failure in the early postoperative period was determined as the most important causes of mortality. These patients belong to the period before 2005 when EDR was preferred. These are patients who died in the first 48 hours. EDR was not the preferred method after 2005 and no patient died in the early period due to abdominal compartment syndrome in our clinic. Although we prefer to perform primary closure in patients with gastroschisis, we use staged reduction with silo if the volume of herniated organs is too high and the abdominal cavity is not developed. Intestinal hypomotility is one of the major problems after gastroschisis operations[12]. Patients are followed-up by administering TPN due to lack of full bowel function and they can be operated multiple times with a diagnosis of intestinal adhesions. In later years, the cause of death in our patients was very long-term hospitalization and sepsis. We lost five patients with septic complications after multiple operations. The average length of hospitalization was 113.5±44.6 days in these patients. Mortality has been associated with long hospitalization, multiple operations, long TPN duration and sepsis. Additional anomalies have no effect on mortality.

A recent study demonstrated that the majority of septic events in this group of patients occur as a result of skin flora[13].

In conclusion, the available articles report conflicting results regarding the effect of gestational age, delivery method, additional anomaly, and birth weight on morbidity and mortality in the treatment of infants diagnosed with gastroschisis.

The limitation of our study is that it is a retrospective study with a small number of cases but gestational age, birth weight, and the presence of an additional anomaly were found to have no effect on the mortality and morbidity of gastroschisis.

The survival rate was 80% in patients with primary closure, 54.5% in patients with elective delayed reduction and 62.5% in patients who had staged reduction with silo. Although no statically significant difference was found between the effects of operation types on morbidity and mortality, primary closure should be preferred and the silo application should be used otherwise. Considering the additional problems caused by prematurity, infants with gastroschisis should wait for term birth.

Conclusion

The present study supports the notion that birth by cesarean section has a positive effect on morbidity. This can be explained by the prevention of compression and ischemic effects on the herniated organs during vaginal birth by choosing cesarean section. The study should be repeated in a larger series by considering the morbidity caused by cesarean section in the mother.

Acknowledgment

Ethical approval for the present retrospective study was granted by Ankara University Ethics Committee.

Conflict of Interest: None

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