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ORIGINAL ARTICLE

Initial Conservative Management of Exomphalos Major with Gentian Violet

Ashrarur Rahman Mitul,* K M N Ferdous

Department of Pediatric Surgery, Dhaka Shishu (Children) Hospital Dhaka, Bangladesh.

ABSTRACT

Aim: The purpose of the study was to assess the results of topical use of gentian violet (GV), among the babies with exomphalos major in our institute.

Methods: The study was carried out retrospectively in a tertiary care hospital during the period from 2005 to 2010 inclusive. Exomphalos patients were classified as major if diameter was >5 cm and/or had liver in the sac as content. These patients were initially preferentially treated conservatively with topical 1% GV over the sac resultig a ventral hernia to be repaired later.

Results: A total of 84 exomphalos patients were admitted during the study period. Among them, 37 neonates (26 males and 11 females) had exomphalos major (EM). Ten of them were prenatally diagnosed. The mean gestational age at delivery was 35 weeks, and mean birth weight was 2.1 Kg. Mean age at presentation was 3.7 days. Thirty (81%) had other associated anomalies, mostly cardiac (66.6%) and pulmonary (46.6%). Ten patients with EM needed early operation because of ruptured sac, and other anomalies. There were 2 preoperative and 8 postoperative deaths in this subgroup. Twenty seven patients were treated conservatively, among these 4 died of overwhelming sepsis. Remaining 23 patients left the hospital with a ventral hernia planned to be repaired at 1 year of age. Overall mortality in our series was 37.83%.

Conclusion: Initial conservative treatment of the sac with GV results in satisfactory outcome for infants with EM who cannot undergo immediate closure.

Key words: Exomphalos major, Conservative treatment, Gentian violet

INTRODUCTION

The incidence of exomphalos is approximately 1 in 3000 to 6000 live births [1, 2]. Some studies have classified this into 'minor' or 'major' (or alternatively 'giant') depending on the diameter of the abdominal wall defect (for instance, <5 and >5 cm or based on nature of contents (e.g. liver), whereas others have not discriminated [3]. Ein et al defined giant exomphalos as those having diameter of >10 cm [4]. Children with exomphalos major (EM) pose considerable problem. The most important issue is to choose between operative and non-operative treatment when simple closure is not possible in patients with EM. In the developing countries, management of babies with exomphalos remains a challenge to the pediatric surgeons because of lack of adequate neonatal intensive care facilities and total parenteral nutrition (TPN) [5]. Non-operative treatment using drying agents was first described by Ahlfeld in 1899 [6]. It still has its advocates. If managed successfully, the child is left with a soundly healed abdominal wall with a ventral hernia. The hernia can be repaired at an appropriate older age. A variety of agents have been used for epithelialisation

* Corresponding Author

(alcohol, mercurochrome, povidone iodine, gentian violet (GV), silver sulfadiazine, silver nitrate [2, 4-9]. In the index study, conservative treatment of EM with GV local application was preferred to operative management because of the lack of adequate neonatal intensive care facilities.

MATERIALS AND METHODS

This was a retrospective study in our institute from 2005 through 2010 inclusive. Ethical permission was taken from the institutional review board (IRB). Demographics, Clinical data, management, length of hospital stay (LOS), age at full enteral feeding, measures of mortality were studied. An informed written consent was taken from the attending guardians after detailing the disease process, condition of the baby and treatment options. After resuscitation, the babies were investigated for hemogram, blood sugar, electrolytes and blood culture. Investigations for associated anomalies included whole body X-ray, ultrasonography of brain and abdomen and 2-D echocardiography. Sepsis was defined as a positive blood culture. After admission, the babies were kept on nothing per oral. They were on partial parenteral nutrition with intravenous 10% glucose and electrolyte containing fluid along with amino acids, intravenous broad spectrum antibiotics. Application of 1% GV was started for all patients with EM with an intact sac, 1 to 2 ml at a time by a resident for 3-4 days, and later the mother was instructed to apply. For initial 10 days, the dye was applied twice daily; later once daily application was initiated when the eschar became firm (Fig. 1). Among EM patients, surgical treatment was performed if they presented with a ruptured sac (n=5) or in cases the sac ruptured while on treatment with GV (n=3). Neonatal intensive care unit (NICU) facilities could not be provided to any of them. Breast feeding was started depending on the condition, and was gradually weaned off intravenous fluids when feed was tolerated and the baby was settled. The patients were discharged when healthy granulation tissue formed and inward epithelialisation started, with the advice to continue applying GV at home. The parents were instructed to come for regular follow up, weekly for four weeks, fortnightly for 2 months, monthly for 6 months and 6 monthly thereafter. At follow up the following points were noted: growth and weight gain, overall condition, condition of the skin over the defect, any side effects attributable to GV. The repair of ventral hernia was undertaken at or after one year of age. All the repairs were possible as a single stage procedure. At surgery, the scar was incised via a midline incision, ventral hernia scar flaps were mobilized laterally with cautery until both the recti were either visualized or palpated. The liver was stuck to most of the upper hernia scar, was carefully identified and protected. If the lateral scar flaps along with the recti could be approximated without tension (n=7) to the midline, closure was accomplished. If the recti often could not be brought to the midline (n=4), a prolene mesh was used to complete the gap.

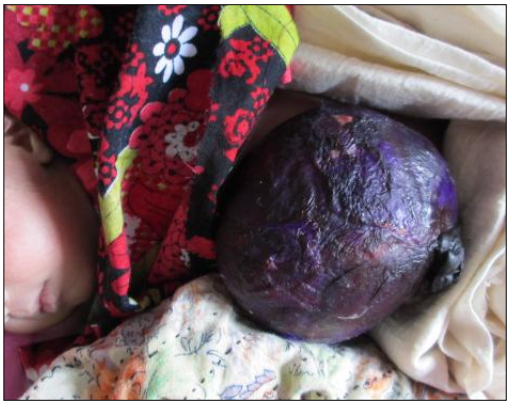

Figure 1: GV application over omphalocele.

RESULTS

Eighty four neonates with exomphalos were admitted in our institute over the study period. Among them, we had 37 EM. The demographics of the study group are shown in Table 1. All (n=10) prenatally diagnosed patients were delivered by caesarian section (CS), 3 at term and 7 between 34 and 36 weeks. Among the remaining (n=27), 19 neonates underwent CS for obstetrical reasons; the rest (n=8) had vaginal delivery. Thirty (81%) patients had other associated anomalies, mostly cardiac (66.6%) and pulmonary (46.6%) (Table 2). Karyotyping was not available in this series. The majority of babies presenting late were septicemic, hypothermic, dehydrated, lethargic

and reluctant to feed. Five babies with EM presented with ruptured sac and 3 had sac ruptured while on conservative treatment. Ten babies with EM needed early operation for sac rupture (n=8) or intestinal obstruction (n=2).

Table 1: Patient demographics (N=37)

Variables	**N**
Male	26
Female	11
Prenatal diagnosis	10*
Mean (wk)	24
Range(wk)	18-32
Gestational age	
Mean (wk)	35
Range (wk)	34-37
Weight	
Mean (kg)	2.1
Range (kg)	1.7-2.7
Age at presentation(days)	
Mean	3.7
Range	6 hours-8 days
Death	14

*All delivered by elective caesarian section

Table 2: Associated anomalies and/or medical problems (n=30)

Variables	**N**
Pulmonary hypoplasia	14
Congenital heart disease	20
Feeding problem	11
Beckwith Wideman Syndrome	3
Inguinal hernia	6
Undescended testis	8
Intraventricular hemorrhage	9
Club hand	7
Miscellaneous	13

Two patients who required surgery died during the preoperative period. Eight patients underwent surgery with silo formation (n= 5) or skin closure only (n=3). All of these babies died postoperatively because of respiratory and cardiac failure, silo separation, associated anomalies and/or overwhelming sepsis. Twenty seven babies were treated with GV were doing well, 4 of them succumbed in the 2nd week due to cardiac failure; echocardiographic diagnoses were Tetralogy of Fallot, ventricular septal defect, patent ductus arteriosus and pulmonary stenosis. Enteral feeding was started on 3rd to 7th postoperative day depending on the condition of the patient. Granulation tissue formation started by 3rd to 4th week and the gap started reducing in size. Epithelialisation and skin coverage usually completed by 4 to 6 weeks with formation of a ventral hernia (Fig. 2). Length of hospital stay was from 3 to 4 weeks. GV is cheap; total cost of 4 to 6 weeks treatment was only USD 2-3. Overall mortality was 37.83%. Survival of patients treated with GV was 85.18%.

Figure 2: Defect epithelialised to form ventral hernia.

Follow up

Twenty patients with EM were under follow up for maximum of 18 months. There was satisfactory skin coverage over the ventral hernia; no adverse effect related to GV was noted. Eleven of them have already undergone ventral hernia repair with a satisfactory scar and no postoperative complications. Seven ventral hernias could be repaired without any prosthesis; rest 4 required prolene meshes. No prosthetic mesh related complication was observed till date. Remaining 9 patients are still living with the ventral hernia; repair of hernia is being delayed for other medical causes like poor general health and coexisting diseases. Three patients were lost to follow up.

DISCUSSION

The object of treatment of patients with exomphalos is primarily to save life, but an important consideration is to achieve a sound abdominal wall and as short a hospital stay as possible [10]. Despite current advances in neonatal care, nutritional support, and surgical and anesthetic technique, overall mortality re-

mains significant. The prognostic indicators identified include size and content of the defect, rupture of the sac, low birth weight, gestational age, associated anomalies and respiratory insufficiency [11, 12]. The optimal treatment of exomphalos babies remains controversial [3]. For small omphalocele the widely accepted standard of care is primary surgical closure [2]. The management of EM remains more controversial, with many centers in the United Kingdom and elsewhere advocating a conservative nonsurgical treatment for larger exomphalos [3]. For EM, initial primary closure is often impossible and Lee et al. believe it should not be even attempted [2]. The viscero-abdominal disproportion gives rise to volume mismatch, severe life threatening squeal like sudden increase in intra abdominal pressure, with elevation of diaphragm and respiratory compromise, hemodynamic instability, inferior vena cava compression and distortion, acute renal failure, bowel obstruction and ischemia [13]. A divergence in treatment of EM has arisen among different authors between surgical closure and non-operative conservative therapy. The surgical options are primary closure, staged repair, or a temporary enclosure in an artificial silon chimney (SC) formation, delayed closure when the cavity permits using a progressive pneumoperitoneum [10]. Some centers have aggressive policy of surgical closure [3, 14, 15], whereas others advocate a much more liberal conservative option [2, 4, 5, 8, 13, 15]. The silon technique was devised to allow for gradual reduction of the contents into the abdominal cavity without acute rise of the intra abdominal pressure. But the SC has frequent complications like, wound infection related to a foreign body leading to dehiscence, fistula formation, delay in enteral feeding, and high incidence of systemic sepsis [15]. We also encountered similar complications in our study with ultimate demise of the patients.

Our data suggests that non operative treatment leads to early enteral feeding and short hospital stay. Conservative treatment has the advantage of avoiding abdominal surgery in the neonatal period, and it averts the risk of tight abdominal closure as well as the particular complications of SC [15]. Nuchtern et al recommends that nonoperative treatment of large omphalocele is safer than SC and results in early enteral feeding and shorter hospital stay [15].

Conservative treatment, our mainstay of therapy, has been described by others too [5, 13]. A variety of agents have been used to paint the sac to promote eschar formation and help epithelialisation (alcohol, mercurochrome, povidone iodine, silver sulphadiazine, silver nitrate) [8, 13, 16]. But all of these agents are not free of risks. Mercury poisoning with mercurochrome, hypothyroidism with Povidone Iodine, silver toxicity with silver nitrate and silver sulphadiazine, alcohol toxicity have all been reported [5, 9, 17]. GV has been used by other authors as well [8, 9, 13]. GV is a triarylmethane antiseptic dye, has been in use since 1890. The name is due to its colour, it is not made from gentian or violet flowers. It has antibacterial, antifungal, and anthelmintic properties and has traditionally been used for treatment of several dermatological conditions, e.g., fungal infections (oral thrush, vaginal candidiasis), superficial bacterial infections like boils, chronic leg ulcers, methicilline resistant staph. aureus (MRSA), oral leukoplakia in HIV positive individuals. It is thought to work by inhibiting reactive oxygen species. Other uses include prevention of umbilical sepsis in newborns, control threadworms, to prevent blood transmission of Chaga's disease, as basis of Gram stain. It has some industrial uses as well. Side effects include irritation of mucous membrane at high concentration, oral ulceration, necrotic skin reaction at high concentration, staining of cloths. Studies have shown that GV is capable of causing cancer in mice [20], but there is no evidence of this occurring in humans. Serious side effects are rare. It is available as 0.5-2% solution [21]. Chan [7] and Mullin [8] have mentioned it to be safe for use in exomphalos, only disadvantage being that it stains linen. In the index study, we have used 1% GV as have others [7]. It is inexpensive; a total cost of 4 to 6 weeks treatment was 2-3 USD. We did not experience any adverse effects related to GV. Associated anomalies in babies with exomphalos are a major concern reported in several series ranging from 30% to 77% [13-15, 18]. In the present study associated anom-

alies were significantly high. We encountered significant number of cardiac and pulmonary anomalies. This is similar to the series reported by Lee et al [2] and Kumar et al [18] Overall mortality rate in author's study corroborated with those reported by other researchers which is between 30-50% [2-4]. Mortality in this study was related to sepsis, complications related to delayed presentation, associated anomalies, prematurity, respiratory and cardiac failure and other medical problems, inadequate intensive care facilities, and lack of total parenteral nutrition. This has been reflected in the report by several other authors [2, 4, 5, 19]. We encountered encouraging outcome with satisfactory survival rate, reduced LOS, early enteral feeding, shorter hospital stay with conservative treatment and no adverse effects with gentian violet. These results correlate with several other studies [2-4, 5, 8, 9, 13, 15].

CONCLUSION

From the study in our institute, we can conclude, in resource challenged situations, conservative management of EM with delayed closure of the ventral hernia should be the choice of management. Associated anomalies and other medical problems should be addressed as well at the same time for better out come. A multidisciplinary approach involving the neonatologists, intensivist and neonatal surgeons may be more rewarding. Gentian violet is an inexpensive and safe escharotic agent, is effective in the treatment. It can be a better alternative to other agents with proven toxicity.

REFERENCES

1. Lakasing L,Cicero S, Davenport M, Patel S, Nicolides KH. Current outcome of antenatally diagnosed Exomphalos: an 11 year review. J Pediatr Surg. 2006; 41: 1403-06.
2. Lee SL, Beyer TD, Kim SS, Waldhausen JH, Healey PJ, Sawin RS, et al. Initial nonoperative management and delayed closure for treatment of giant omphaloceles. J Pediatr Surg. 2006; 41:1846-9.
3. Rijhwani A, Davenport M, Dawrant M, Dimitriuo G, Patel S, Greenough A, et al. Definitive surgical management of antenatally diagnosed exomphalos. J Pediatr Surg. 2005; 40: 516-22.
4. Ein SH, Langer JC. Delayed management of giant omphalocele using silver sulphadiazine cream: an 18-year experience. J Peditr Surg. 2012; 47: 494-500.
5. Wakhlu A, Wakhlu AK. The Management of Exomphalos. J Pediatr Surg. 2000; 35: 73-6.
6. Jones PG. Exomphalos (syn. Omphalocele): a review of 45 cases. Arch. Dis. Childh. 1963; 38: 180-7.
7. Debnath B, Biswas SK. Chylous ascites following repair of exomphalos major--a report. J Indian Med Assoc. 2010; 108:517-8.
8. Chan MCK. Giant omphalocele- Conservative or operative treatment. Correspondence. Arch Dis Child. 1980: 55: 167.
9. Mullins ME, Horowitz BZ. Iatrogenic neonatal murcury poisoning from mercurochrome of a large omphalocele. Clin Pediatr 1999; 38: 111-2.
10. Sonmez K, Onal E, Karabulut R, Turan O, Turkyilmaz Z, Hirfanoglu I, et al. A strategy for treatment of giant omphalocele. World J Pediatr 2010; 6: 274-7.
11. Tsakayannis DE, Zurakowski D, Lillehei CW. Respiratory insufficiency at birth: a predictor of mortality for infants with omphalocele. J Pediatr Surg. 1996; 31:1088-91.
12. Cooney DR. Abdominal defects in Welch. In: O'Neil Jr JA, Rowe MI, Grosfeld JL, Fonkalsrud EW, Coran Ag, editors. Pediatric Surgery, Vol 2. 5th edition. St Louis(Mo): Mosby-Year Book, Inc; 1998; p. 1045-71.
13. Adam AS, Corbally MT, Fitzgerald RJ. Evaluation of conservative therapy for exomphalos. Surg Gynecol Obstet. 1991; 72:394-6.
14. Dann JC, Fonkalsrud EW. Improved survival of infants with omphalocele. Am J Surg. 1997; 173: 284-7.
15. Nuchtern JG, Baxter R, Hatch EI. Nonoperative initial treatment management versus silon chimney for management of giant omphalocele. J Pediatr Surg. 1995; 30:771-6.
16. Bas NMA: Exomphalos and Gastroschisis. In: Freeman NV, Burge DM, Griffiths MP, Malone PSJ. (eds): Surgery of the Newborn. England, Churchill Livingstone 1994; p 301-20.
17. Lewis N, Kolimarala V, Lander A. Conservative management of exomphalos major with silver dressing: are they safe? J Pediatr Surg. 2010; 45: 2438-9.
18. Kumar HR, Jester AL, Ladd AP. Impact of omphalocele size and associated conditions. J Pediatr Surg. 2008; 43:2216-9.
19. Vachharajani AJ, Rao R, Keswani S, Mathur AM. Outcomes of exomphalos: an institutional experience. Pediatr Surg Int. 2009: 25:139-46.
20. Bunker CB. Topical gentian violet in dermatology. J Am Acad Dermatol 2009; 60: 247-8.
21. Balabanova M, Popova L, Tchipeva R. Dyes in dermatology. Clin Dermatol 2003; 21: 2-6.

Address for correspondence

Md. Ashrarur Rahman Mitul,

Department of Pediatric Surgery, Dhaka Shishu (Children) Hospital Dhaka, Bangladesh.

E mail: drmitul@dhaka.net

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