Journal of Pediatric Surgery (2010) 45, E17–E20

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Journal of Pediatric Surgery

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Management of a giant omphalocele with an external skin closure system

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Received 25 December 2009; revised 30 April 2010; accepted 3 May 2010

Key words:
Giant omphalocele;
Abdominal closure device

Abstract

Background/Purpose: The management of neonates with giant omphalocele remains challenging and multiple strategies have been described. We present the case of a 34-week-old neonate with isolated giant omphalocele managed with an external surgical skin closure system as a component of a staged closure strategy.

Case Presentation: An Inuit boy of 34 weeks gestation was born by urgent Caesarean delivery at an affiliated obstetrical hospital with a giant ruptured omphalocele and loss of abdominal domain. He was transferred to our institution and a silastic silo was fashioned and placed in the operating room. He returned to the operating room several times and was treated by placement of a combined Gore-Tex (WL Gore and Associates, Flagstaff, Ariz)/silastic inlay mesh. An eschar formed over this temporary closure, and we elected to place a dynamic skin closure device to continue gradual bedside reduction. The initial abdominal wall defect was 8.5 cm in transverse diameter and was reduced to 4.5 cm over 3 weeks. Complete closure was subsequently achieved without the need for skin grafting.

Discussion: The use of a dynamic reduction skin closure device has not been documented previously in the pediatric population or in the context of a congenital defect. We describe the use of an external surgical skin closure device in the context of the staged closure of a giant neonatal omphalocele and postulate that such a device may prove useful in the treatment of other congenital tissue defects.

The optimal management of neonates with giant omphalocele remains controversial. The resultant visceroabdominal disproportion often excludes the possibility of a primary repair, and multiple, staged closure strategies have been described. The use of skin flaps was originally described by Gross [1], whereas newer publications favor the use of absorbable [2] or nonabsorbable [3] mesh to achieve closure. Adult surgeons have been increasingly using external closure devices to aid in abdominal closure after damage control laparotomy [4,5]; a phase IV trial of this strategy was due to begin on July 2009 (ClinicalTrials.gov Identifier: NCT00754156). We present the case of a 34-week-old neonate with isolated giant omphalocele managed with an external surgical skin closure system as a component of a staged closure strategy.

1. Case presentation

An Inuit boy of 34 weeks gestation was born to an 18-year-old mother at an affiliated obstetrical hospital with a

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doi:10.1016/j.jpedsurg.2010.05.004

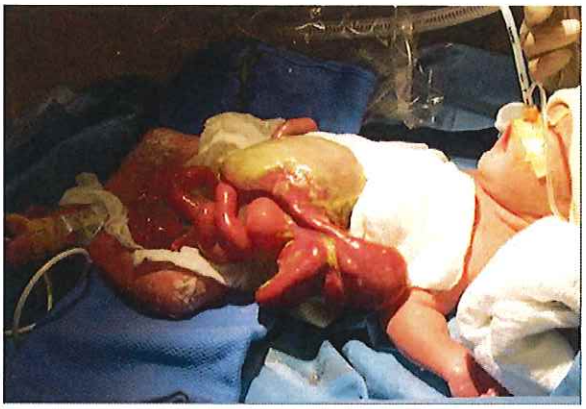

Fig. 1 Day of life 1 after transfer to our institution. Note complete evisceration of liver and small intestine.

Fig. 3 External skin closure device (ABRA device, Canica) with underlying eschar over a Gore-Tex/Vicryl neofascia.

giant ruptured omphalocele (defect >6 cm) and loss of abdominal domain. The diagnosis of omphalocele had been established at 25-week antenatal ultrasound, at which time a cardiac echo and karyotype were performed, both of which were normal. The mother underwent urgent Caesarean delivery for a nonreassuring nonstress test. Upon delivery, Apgar scores were 1^1, 4^5, 7^{10} and most of the small intestine and liver were noted to be extraabdominal with no visible sac (Fig. 1). After placing the lower extremities and trunk of the child in a plastic bag, the patient was transferred to our nursery. At operation, a 7.5-cm silastic sac (Bentec Medical Company, Woodland, Calif) was fashioned to size and sewn to the fascial edges to attain initial coverage. Investigations in the neonatal intensive care unit revealed no other associated congenital abnormality and the child was managed with parenteral nutrition, prophylactic antibiotics, and gradual plication of the silo with plastic clamps.

Fig. 2 Day of life 16 after insertion of Gore-Tex "neofascia" (covered by gauze). Note the central area of silastic undergoing gradual plication (solid arrow) and the circumferential vertical traction sutures (unfilled arrows) on the "neofascial" edges.

He returned to the operating room on day of life (DOL) 14 as the silastic had begun tearing away from the fascial edges, and the visceroabdominal disproportion precluded further reduction. At this time, the defect was enlarged inferiorly, the skin and fascia were formally separated, and a Gore-Tex (WL Gore and Associates, Flagstaff, Ariz) "neofascia" ring was sewn in place. Complete reduction was not attained at this time, however, and the center of the incomplete Gore-Tex ring was again covered with a silastic sheet sewn to the neofascial edges. Continued plication of the silastic was undertaken, and traction was applied to this neofascia by securing it to an arch within the isolette, although the resultant force vectors were vertical as opposed to horizontal (Fig. 2). With gradual plication of the silastic sheet, viscera were brought to the level of the abdominal wall. Because the silastic bag continued to tear, it was replaced in the OR on DOL 27 with a Vicryl mesh within the neofascial ring.

An eschar formed over this temporary closure, and we elected to place a surgical skin closure apparatus (ABRA device, Canica, Ontario, Canada) to aide in gradual bedside reduction (Fig. 3). The force vectors of this device were horizontal across the wound, optimizing skin stretch and ingrowth. The initial abdominal wall defect was 8.5 cm transverse diameter and was reduced to 4.5 cm over 3 weeks, with improved skin coverage. Definitive closure was attained in the OR via a modified component separation technique on DOL 86. Asymmetry in the integrity of the fascial layers meant incomplete fascial coverage in the right hemiabdomen. However, skin coverage was obtained without grafting, reinforced with retention sutures. These were gradually replaced by steri-strips over the following weeks, and the patient was discharged from hospital on DOL 98. The patient began enteral feeding on DOL 23, mechanical ventilation was stopped on DOL 38, and parenteral nutrition was ceased on DOL 52. After discharge, the baby was on the 10th percentile growth curve for both height and weight with an

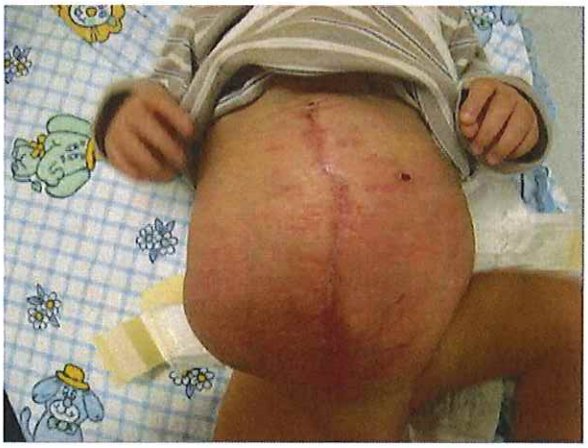

Fig. 4 Post-discharge follow-up visit at 4.5 months of age. Note the acceptable cosmesis of midline wound and right-sided ventral hernia.

acceptable cosmetic result. A ventral hernia persists in the lateral aspect of the abdominal wall (Fig. 4).

2. Discussion

The myriad of treatment options described in the management of giant omphalocele is a testament to the challenge of caring for these neonates. There remains no consensus regarding the strict definition of "giant" omphalocele; many publications simply fail to define the condition while others vary in size cutoffs and omphalocele content [3,6-11]. The overarching definition of "giant" omphalocele, therefore, relies less on absolute measurements and more on the disproportion between abdominal cavity and herniated contents [12]. Such a scenario tests the creative limits of each clinician and has resulted in the evolution of several different treatment strategies. The historical treatment of neonates with abdominal loss-of-domain has been to paint the sac or exposed viscera with topical agents designed to promote granulation and eventual epithelialization [13]. Although this method held initial promise, several reports of mercury toxicity after mercurochrome application hastened the search for surgical alternatives [14-16].

A noninvasive technique is the use of the amniotic sac to aid in gradual visceral reduction, although this strategy is only applicable in the context of an intact sac or one with a tear that can be easily repaired [17,18]. In the absence of an intact sac, several options exist. Absorbable mesh has been used to cover the defect followed by either mobilizing skin flaps or using skin grafts to achieve complete coverage [3,19]. Intestinal fistula remains a risk with this technique. Vacuum-assisted closure devices have also been described as an adjunct to a mesh closure strategy as a means of accelerating tissue in-growth and managing enterocutaneous fistulas [20]. Both of these methods often necessitate subsequent skin grafting, which may be a challenge in neonates lacking substantial donor sites. Although recent reports have suggested several alternatives to skin grafting, including acellular human dermis (Alloderm, Lifecell, Branchburg, NJ) and porcine small intestinal submucosa [21,22], the use of autologous native skin is always preferred.

The use of a dynamic wound closure system has been investigated in adults in the context of the traumatic "open abdomen." These systems have been shown to significantly decrease abdominal procedures and days to abdominal closure when compared to other closure techniques. They have also resulted in a reduction in the need for skin grafting and the incidence of incisional hernias [4,5]. Dynamic wound closure appears to exert its benefits through multiple mechanisms at both the cellular and tissue levels. Indeed, connective tissue growth has been found to be up-regulated in cells exposed to mechanical stress [23]. The mechanism most responsible for the success of dynamic wound closure is the resultant "mechanical creep"—a biomechanical property of skin that allows it to gradually stretch beyond the limits of its inherent extensibility [24]. This increase in length occurs within minutes and is explained by the straightening of collagen fibers in the direction of the stretching force, until the fibers are parallel and resist further extension [25,26].

Based on our experience with this patient, the authors endorse a 2-phase approach to the management of ruptured omphalocele. The initial priority includes the establishment of a reasonable antimicrobial barrier followed by gradual visceral reduction without comprising cardiorespiratory stability. Once this initial phase has been achieved, complete tissue coverage is required. The authors feel that the ABRA system provides an additional tool in the armamentarium of clinicians once the patient has progressed to this second phase of care.

We report a case of giant ruptured omphalocele managed with a staged closure strategy. A dynamic wound closure device was used as an adjunct to attain complete skin coverage and likely mitigated the need for grafting. The authors would now consider earlier use of this device but only once visceroabdominal disproportion had been reduced and skin closure was being considered. We postulate that such a device may prove useful in the treatment of other congenital tissue defects.

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