The use of Alloderm for a giant omphalocele with a ruptured sac and inadequate skin coverage

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Giant omphaloceles often present with a loss of abdominal domain. An intact sac may permit simple daily dressings to promote epithelialization for a delayed operative closure; a ruptured sac presents challenges. There may be insufficient native tissue for adequate coverage; tissue expanders or grafts may be needed. We present a case of a ruptured omphalocele sac in which non-operative management was not possible – silo placement and reduction was unsuccessful due to a lack of abdominal domain. We used an acellular human dermal matrix (Alloderm, Life Cell, Branchburg, NJ), sutured to the patient’s fascia to provide coverage of the omphalocele contents. There was insufficient skin to cover the majority of the graft. Tissue expanders had to be removed due to infection. With regular dressing changes, the tissue underneath the graft granulated and epithelialized and the graft lifted off. Skin grafting was not required. Alloderm’s biologic properties render it less prone to infection than a synthetic graft; there was no evidence of graft infection in this patient. This ruptured omphalocele was managed with Alloderm coverage and minimal native skin.

Omphaloceles occur in 1 in 4000–6000 live births [1]. Improvements in antenatal, neonatal, surgical and anesthetic care of neonates with abdominal wall defects have resulted in increased survival rates [2,3]. Prognostic indicators include the size of the defect, antenatal rupture of the sac, low birth weight, gestational age, associated anomalies, and perinatal respiratory distress [1,4,6]; the morbidity and mortality are dramatically higher with giant omphaloceles, i.e., the diameter of the defect is greater than 5 cm and the sac contains liver [3,4,7–9].

Small- to medium-sized defects should be repaired. Giant omphaloceles are challenging to manage – there is often a large degree of visceroadominal disproportion such that primary reduction and closure of the abdominal wall is prohibited [10,11]. This challenge is compounded by the presence of associated anomalies [6,12]. A forced closure can result in increased intra-abdominal pressure and/or kinking of visceral blood vessels leading to renal failure, pulmonary failure and superior mesenteric artery occlusion [5,7,13–15].

1. Case report

A 1.7 kg girl was delivered by elective cesarean section for intrauterine growth restriction at 35 weeks of gestation and an antenatally diagnosed large omphalocele. There was a maternal history of chronic hypertension, carpal tunnel syndrome, and marijuana use. The abdomen was initially wrapped in plastic sheeting and the baby was transferred to the neonatal intensive care unit. Examination revealed bowed and shortened lower extremities, absence of the right great toe and oligodactyly of the left foot. As well, there was a giant omphalocele involving the stomach, liver, spleen, small and large bowel. There was a large rent in the sac, and given the exposed liver, a primary sac repair was not performed (Fig. 1). A 7.5 cm spring-loaded silo (Bentec Medical Inc., Woodland, CA) was placed around the sac and secured. Despite attempts at serial reduction, there had been minimal progress, and 10 days later the silo was replaced another one with the ring portion removed — this was sewn directly to the abdominal skin (after trimming the sac from the skin edges). We received approval for the use of Alloderm — 10 days later the second silo was removed. We noted a fibrinous peel on the small bowel but felt it insufficient to simply proceed with topical management. Skin flaps were raised from the underlying fascia and the Alloderm was sewn circumferentially to the fascia under moderate tension. The Alloderm was...
then sewn to itself across the top of the defect (two sheets of 8 × 16 × 0.78 mm each). The skin edges were tacked up the sides of the Alloderm with interrupted sutures.

There was insufficient skin to cover the Alloderm — dressings were applied with non-adherent, moisturizing, gel and dry gauze layers, and were changed two to three times per week.

Subcutaneous tissue expanders were placed in each flank but had to be removed on post-operative day 2 due to concerns about infection (overlying cellulitis and purulent discharge). Intra-abdominal expansion was not performed as the patient required significant ongoing respiratory support. Overtime, granulation tissue appeared at the skin-Alloderm interface and the Alloderm retracted to the most ventral portion of the omphalocele (Figs. 2 and 3). The remaining Alloderm was ultimately removed at 15 months of age. The baby had a tracheostomy placed at 2 months of age for pulmonary complications (pneumonitis secondary to reflux of feeds) and bronchomalacia, and was on continuous nasojejunal feeds. She was discharged using a home-ventilator at 8 months of age with her mother performing dressing changes.

2. Discussion

Ladd and Gross originally used skin flaps [11,16], but other methods have been applied to achieve closure of giant omphaloceles. Schuster introduced the staged silo closure [17], and then others used tissue expanders [18,19], serial sac clamping [20,21], topical management [22,23], component separation [24], or grafts [23,25–27]. Others have recently employed negative-pressure therapy to reduce the viscera, followed by grafting with an acellular dermal matrix [25,28]. An intraperitoneal tissue expander has been used to create peritoneal domain followed by primary closure [29].

Non-operative management can be applied to omphaloceles with an intact sac by topical treatment of the omphalocele membrane to promote epithelialization followed by secondary repair of the ventral hernia [3,22,30]. Non-operative management may result in lower complication rates than staged reductions with a silo [2,3], and removes the need for major surgery in the newborn period [2]. Our patient, however, presented with a ruptured sac so topical treatment was not possible.

The management of ruptured omphalocele sacs can be more complicated. Some may respond to simple silo placement and serial reduction [2], whereas others may require the use of grafts to achieve interim organ coverage [31]. Alloderm is an acellular human dermal matrix with a tensile strength similar to human fascia; its properties make it less susceptible to infection or to an inflammatory reaction, including the formation of adhesions, than synthetic grafts [32–36]. Its structure supports tissue ingrowth to
initiate angiogenesis [34,37]. Alloderm has typically been used with subsequent coverage by skin or skin grafts [31,32,38]. Our patient is unique in that there was insufficiency of skin to cover the graft, and the infant was too small to provide enough skin for allografting. Additionally, the surgically placed tissue expanders had to be removed due to infection. We used serial dressings to achieve granulation and epithelialization of the tissue exposed as the Alloderm lifted off over time. Given its native properties, it was unusual that the Alloderm lifted off. Ultimately, we plan for serial reductions and removal of excess epithelialized tissue.

3. Conclusion

As giant omphaloceles are uncommon, it is difficult to carry out a prospective evaluation of the different treatment modalities available [39]. There are many ways to attempt closure, depending on the patient comorbidities, relative size of the defect, and extent of native tissue available. For patients with giant omphaloceles and a ruptured sac, Alloderm, or other biologic grafts may provide an option for visceral coverage even in the absence of adequate skin coverage.

The patient in this case is doing well at home. She is almost 2 years of age, continues on nasojejunal feeds, and is slowly weaning from her home-ventilator.

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References


