



Contents lists available at ScienceDirect

Journal of Pediatric Surgery CASE REPORTS

journal homepage: www.jpascasereports.com

Repair of giant omphalocele in a premature neonate with non-cross-linked porcine acellular dermal matrix (Strattice Tissue Matrix)



Helene Engstrand Lilja^{a,*}, Daisy Schulten^b

^a Institution of Women's and Children's Health, Uppsala University, Uppsala, Sweden

^b University Hospital of Cologne, Kerpener Straße 62, 50937, Cologne, Germany

ARTICLE INFO

Article history:

Received 31 May 2016

Received in revised form

26 June 2016

Accepted 28 June 2016

Key words:

Giant omphalocele

Acellular dermal matrix

Strattice

ABSTRACT

The management of giant omphalocele (GO) is a major challenge in pediatric surgery and there are many different surgical strategies described. Here we report a complicated case in which the abdominal wall in a premature neonate (gestational age 33 + 2 weeks and 1700 g) with GO was reconstructed with a non-cross-linked acellular porcine dermal matrix (Strattice™) combined with vacuum therapy. This strategy can be an alternative method in the repair of GO in premature neonates with high risk of infection, underdeveloped abdominal cavity and insufficient native tissue.

© 2016 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Omphalocele is a congenital abdominal wall defect with herniation of intraabdominal organs covered by a sac [1]. The prevalence is estimated to 1 in 2500–6000 live births in Western countries [2]. A giant omphalocele (GO) has by several authors been defined as an omphalocele with a defect larger than 5–6 cm in diameter and the sac contains liver [1,3,4]. Other authors have defined GO as a defect with >50% of the liver in the omphalocele sac [5]. In neonates with GO the abdominal cavity is underdeveloped and the abdominal wall muscles hypoplastic and laterally displaced [6]. The thoracic cavity may also be underdeveloped and pulmonary hypoplasia is often present [7,8]. The outcome is dependent on associated anomalies and genetic disorders, size of the defect, prematurity, pulmonary hypoplasia and intact sac [8–10]. Closure of GO is a major challenge to the pediatric surgeon and many different techniques have been described. A forced closure can result in abdominal compartment syndrome and respiratory insufficiency.

In this case report we describe the use of a non-cross-linked acellular porcine dermal matrix (Strattice™, Life Cell Corp., Branchburg, New Jersey, United States) in combination with vacuum therapy in a premature baby with a very thin sac and insufficient abdominal wall and skin to cover the defect.

1. Case report

The diagnosis of giant omphalocele was detected prenatally at the routine ultrasound at 18 weeks of gestation. The mother suffered from severe pre-eclampsia and therefore a cesarean was performed at the gestational age of 33 + 2 weeks. The birth weight was 1700 g, length 42 cm and Apgar Scores were 7, 6, 10. The child needed ventilator support with continuous positive air pressure with 25% O₂. Preoperative echocardiography found a dextrocardia. Intravenous antibiotics were initiated after birth as prophylaxis against infection. The first GO repair was scheduled to the following day. The omphalocele was found to contain the whole liver, small bowel, stomach and spleen. The sac was extremely thin, fragile and not intact (Fig. 1A) and subsequently conservative treatment was not an alternative.

The abdominal cavity was very small and there was not enough skin available to cover the defect (Fig. 1B).

At surgery it was not possible to close the abdomen. After partial closure of the upper part of the abdominal wall, the stomach and spleen could be repositioned into the abdomen. A silo was constructed of 10 × 16 cm Strattice™ Pliable with single 3:0 Prolene sutures (Ethicon, Somerville, New Jersey, United States). The company recommended Pliable, due to the size of the child, and use of permanent sutures (Fig. 2). A vacuum dressing (V.A.C., KCI-Medical, Mölndal, Sweden) was fashioned over the silo. A continuous

* Corresponding author. Section of Pediatric Surgery, University Children's Hospital, SE-751 85 Uppsala, Sweden. Tel.: +46 186115901; fax: +46 186115905.

E-mail address: helene.lilja@kbh.uu.se (H.E. Lilja).

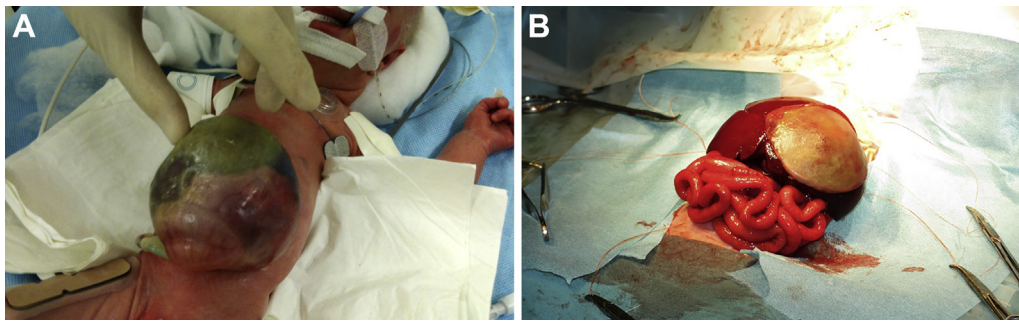


Fig. 1. A) The giant omphalocele was found to contain all of the liver, small bowel, stomach and spleen and the sac was extremely thin. B) After removal of the sac, during the first GO repair.

pressure of 50 mm Hg was applied (Fig. 3). The vacuum dressing was changed 10 times over a period of 5 weeks before the vacuum therapy could be discontinued. Enteral feeding was gradually introduced 10 days after the primary surgery and the patient was tolerating full enteral nutrition 4.5 weeks later. Eight weeks post-operatively the central part of the Strattice™ (4 × 4 cm) became dry and there was a local infection at the skin margins. A surgical revision was performed, the dry Strattice™ was removed and the patient was treated with antibiotics. A Jelonet Paraffin gauze dressing was applied on the defect and covered by a sterile Hydrocolloid dressing. After one week a 4 × 2 skin transplant was applied. The healing was successful and the patient was discharged home at the age of 3 months. A second surgery was scheduled at about 23 months of age (Fig. 4). The plan was to close the fascia without any foreign material.

At the second GO repair it was found that the Strattice Pliable™ was incorporated to the surrounding fascia but too thin to be used to close the defect. It was also adherent to the liver but not to the intestines. It was left in place. The fascia was identified in the flanks and mobilized. A 16 × 20 cm Strattice™ Firm was divided into two halves and sutured to the fascia with interrupted 2:0 Prolene sutures. The defect was reduced as much as possible and the two

halves of Strattice™ were sutured together in the midline with interrupted 2:0 Prolene sutures. The thin skin in the midline was excised and the skin was closed in the midline with intracutaneous 5:0 Monocryl (Ethicon, Somerville, New Jersey, United States). Four months after the second surgery some of the Prolene sutures penetrated the thin skin. A minor surgery was performed to shorten the sutures. A third GO repair was scheduled at 4 years of age. Several Prolene sutures again penetrated the skin and there was a local skin infection. Surgery was therefore scheduled at 13 months after the second GO repair when the girl was 3 years old.

At this surgery the Strattice™ was found to be of a fascialike quality, it could be reduced in the middle resulting in a midline diastasis of 3–4 cm (Fig. 5). A sample of Strattice™ was sent for histological examination (Fig. 6). All Prolene sutures were removed and the closure was made with running 3:0 Ethibond (Ethicon, Somerville, New Jersey, United States). The patient was discharged home 5 days after surgery. The girl was doing well, living a normal active life with her family at the latest control at the age of 3 years and 3 months (Fig. 7). A future operation on cosmetic indication is probably needed to further tighten the abdominal wall.

2. Discussion

Multiple techniques have been described to achieve closure of GO such as conservative treatment with epithelialization with or without topical agents and later repair of the ventral hernia [11], staged silo closure [12], grafts [13,14], use of intraperitoneal tissue expanders [15,16], or vacuum assisted closure [17]. Other pediatric surgeons have used AlloDerm (an acellular human dermal matrix) covered by skin and skin grafts in the management of GO [18–21]. We decided to use Strattice as it is stiffer and thicker than AlloDerm and is usually less expensive.

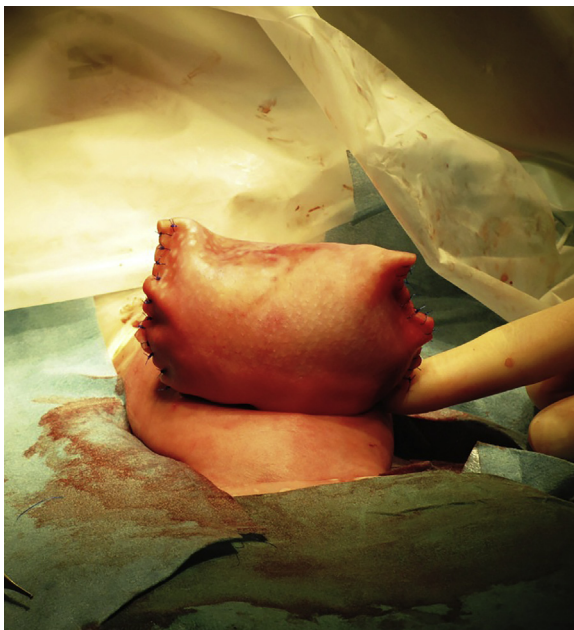


Fig. 2. A silo was constructed of 10 × 16 cm Strattice™ Pliable with interrupted 3:0 Prolene sutures.



Fig. 3. A vacuum dressing was fashioned over the silo.



Fig. 4. Before the second surgery at about 23 months of age.

In this premature girl a silo placement and reduction was not an alternative due to a very small abdominal cavity and a large defect with the entire liver in the sac. There was not enough skin to be closed over the defect. Conservative treatment was not an alternative due to a very thin and fragile sac.

Strattice Reconstructive Tissue Matrix is a non-cross-linked, acellular dermal matrix derived from porcine origin. It is supplied in two different types: *Pliable*, which is thinner and softer, and has been used in breast reconstruction, and *Firm* which is thicker and has been used in complex abdominal wall hernia reconstructions in adults. The graft is treated to remove all porcine cells and still retain the 3D-structure of the extracellular matrix. This matrix acts like a scaffold for the ingrowth of the patient's own tissue by promoting

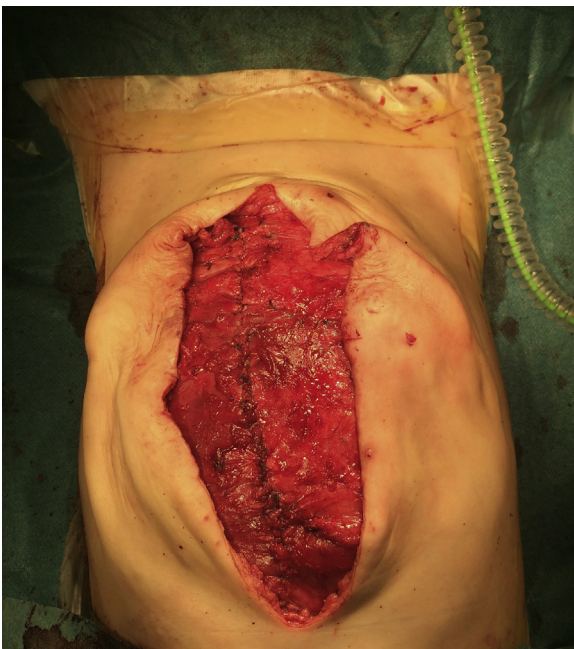


Fig. 5. The Strattice™ *Firm* was found to be of a fascialike quality at the third surgery (13 months after it was applied).

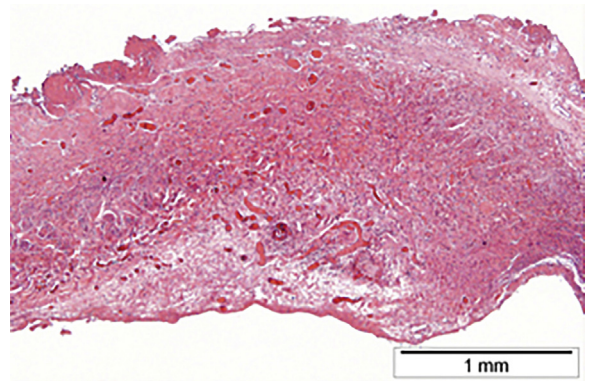


Fig. 6. Microscopically, there are highly vascularized collagen rich connective tissue containing several small foci of inflammation. The inflammatory infiltrate varies and dominates partially by neutrophils granulocytes and partially by foreign body giant cells. In addition, there are scattered macrophages and lymphocytes (Hematoxylin and Eosin staining).

rapid revascularization, white cell migration and cell repopulation. Its properties make it less susceptible to infection or to an inflammatory reaction, including the formation of adhesions, than synthetic grafts [22–24]. Recently, the use of Strattice™ was reported in a pediatric population including two children with omphalocele reconstructed at the age of 4.25 and 5.8 years [25]. A full term neonate with GO was reconstructed with Strattice™ in combination with vacuum therapy on the 14th day after birth [26]. To our knowledge this is the first report on the use of Strattice™ in a premature neonate with GO. In this case, Strattice was a suitable



Fig. 7. The child three months after the third surgery.

graft as it is less susceptible to infection and it facilitates the skin growth as it becomes highly vascularized in contrast to Gore-Tex (W. L. Gore & Associates, Inc., Medical Products Division, Flagstaff, AZ). In contrast to adults with a seroma formation of 29%, there was no seroma formation in our patient [27]. The lesson we learned from this case is to avoid the Strattice™ to dry which easily occurs when the VAC therapy is discontinued. We recommend a skin transplant to cover the Strattice™ at this stage. In future cases we will use Strattice™ Firm at the first surgery and Ethibond instead of Prolene as it easily penetrates the skin.

3. Conclusion

Repair of GO in a premature neonate with a large fascia and skin defect was well tolerated and successful with the use of Strattice™ in combination with vacuum therapy. This strategy can be an alternative method in the repair of GO in premature neonates with increased risk of infection, underdeveloped abdominal cavity and insufficient native tissue.

Conflicts of interest

The authors declare that they have no conflict of interest.

Informed consent

Informed consent was obtained from the parents to the patient in this report.

References

- [1] Gamba P, Midrio P. Abdominal wall defects: prenatal diagnosis, newborn management, and long-term outcomes. *Semin Pediatr Surg* 2014;23(5):283–90.
- [2] Coran AG, Caldamone A, Adzick A, Krummel TM, Laberge JM, Shamberger R. *Pediatric surgery*. 7th ed. St. Louis, MO: Mosby; 2012.
- [3] Foroutan H, Jahromi BJ, Dastgheyb N, Najafi S. A new method of repairing giant omphaloceles with bilateral mesh grafts lateral to the rectus abdominis muscles. *Ann Colorectal Res* 2013;1(1).
- [4] Pacilli M, Spitz L, Kiely EM, Curry J, Pierro A. Staged repair of giant omphalocele in the neonatal period. *J Pediatr Surg* 2005;40(5):785–8.
- [5] Akinkuotu AC, Sheikh F, Olutoye OO, Lee TC, Fernandes CJ, Welty SE, et al. Giant omphaloceles: surgical management and perinatal outcomes. *J Surg Res* 2015;198(2):388–92.
- [6] Islam S. Advances in surgery for abdominal wall defects: gastroschisis and omphalocele. *Clin Perinatol* 2012;39(2):375–86.
- [7] Kamata S, Usui N, Sawai T, Nose K, Fukuzawa M. Prenatal detection of pulmonary hypoplasia in giant omphalocele. *Pediatr Surg Int* 2008;24(1):107–11.
- [8] Danzer E, Hedrick HL, Rintoul NE, Siegle J, Adzick NS, Panitch HB. Assessment of early pulmonary function abnormalities in giant omphalocele survivors. *J Pediatr Surg* 2012;47(10):1811–20.
- [9] Mann S, Bliinman TA, Douglas Wilson R. Prenatal and postnatal management of omphalocele. *Prenat Diagn* 2008;28(7):626–32.
- [10] Corey KM, Hornik CP, Laughon MM, McHutchison K, Clark RH, Smith PB. Frequency of anomalies and hospital outcomes in infants with gastroschisis and omphalocele. *Early Hum Dev* 2014;90(8):421–4.
- [11] 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.
- [12] Schuster SR. A new method for staged repair of large omphaloceles. *Surg Gynecol Obstet* 1967;837–50.
- [13] Ein SH, Shandling B. A new nonoperative treatment of large omphaloceles with a polymer membrane. *J Pediatr Surg* 1978;13:255–7.
- [14] Drewa T, Galazka P, Prokurat A, Wolski Z, Sir J, Wysocka K, et al. Abdominal wall repair using a biodegradable scaffold seeded with cells. *J Pediatr Surg* 2005;40:317–21.
- [15] Martin AE, Khan A, Kim DS, Muratore CS, Luks FI. The use of intraabdominal tissue expanders as a primary strategy for closure of giant omphaloceles. *J Pediatr Surg* 2009;44:178–82.
- [16] De Ugarte DA, Asch MJ, Hedrick MH, Atkinson JB. The use of tissue expanders in the closure of a giant omphalocele. *J Pediatr Surg* 2004;39:613–5.
- [17] Kilbride KE, Cooney DR, Custer MD. Vacuum-assisted closure: a new method for treating patients with giant omphalocele. *J Pediatr Surg* 2006;41:212–5.
- [18] Alaish SM, Strauch ED. The use of Alloderm in the closure of a giant omphalocele. *J Pediatr Surg* 2006 Mar;41(3):e37–9.
- [19] Almond SL, Goyal A, Jesudason EC, Graham KE, Richard B, Selby A, et al. Novel use of skin substitute as rescue therapy in complicated giant exomphalos. *J Pediatr Surg* 2006;41:e1–2.
- [20] Ladd AP, Rescorla FJ, Eppley BL. Novel use of acellular dermal matrix in the formation of a bioprosthetic silo for giant omphalocele coverage. *J Pediatr Surg* 2004;39:1291–3.
- [21] Kapfer SA, Keshen TH. The use of human acellular dermis in the operative management of giant omphalocele. *J Pediatr Surg* 2006;41:216–20.
- [22] Connor J, McQuillan D, Sandor M, Wan H, Lombardi J, Bachrach N, et al. Retention of structural and biochemical integrity in a biological mesh supports tissue remodeling in a primate abdominal wall model. *Regen Med* 2009;4(2):185–95.
- [23] Sandor M, Xu H, Connor J, Lombardi J, Harper JR, Silverman RP, et al. Host response to implanted porcine-derived biological materials in a primate model of abdominal wall repair. *Tissue Eng Part A* 2008;14(12):2021–31.
- [24] Campbell KT, Bums NK, Rios CN, Mathur AB, Butler CE. Human versus non-cross-linked porcine acellular dermal matrix used for ventral hernia repair: comparison of in vivo fibrovascular remodeling and mechanical repair strength. *Plast Reconstr Surg* 2011;127(6):2321–32.
- [25] Begum T, Farrelly PJ, Craigie RJ. Non-cross-linked porcine acellular dermal matrix (Strattice Tissue Matrix) in pediatric reconstructive surgery. *J Pediatr Surg* 2016;51(3):461–4.
- [26] Travassos DV, van Eerde AM, Kramer WL. Management of a giant omphalocele with non-cross-linked intact Porcien-derived acellular dermal matrix (Strattice) combined with vacuum therapy. *Pediatr Surg Rep* 2015;3(2):61–3.
- [27] Itani MF, Rosen M, Vargo D, Awad SS, Denoto 3rd G, Butler CE, et al. Prospective study of single-stage repair of contaminated hernias using a biologic porcine tissue matrix: the RICH Study. *Surgery* 2012;152:498–505.