Original Paper

Fetal Diagnosis and Therapy

Fetal Diagn Ther 2018;44:173–178
DOI: 10.1159/000479926

Received: June 6, 2017
Accepted after revision: July 25, 2017
Published online: December 20, 2017

In utero Plastic Surgery in Zurich: Successful Use of Distally Pedicled Random Pattern Transposition Flaps for Definitive Skin Closure during Open Fetal Spina Bifida Repair

Martin Meuli[a, b] Claudia Meuli-Simmen[c] Luca Mazzone[a, b]
Sasha J. Tharakan[a, b] Roland Zimmermann[a, d] Nicole Ochsenbein[a, d]
Ueli Moehrlen[a, b]

[a]Zurich Center for Fetal Diagnosis and Therapy and [b]Department of Pediatric Surgery, University Children's Hospital Zurich, Zurich, [c]Department of Plastic, Reconstructive, Aesthetic, and Hand Surgery, County Hospital Aarau, Aarau, and [d]Department of Obstetrics, University Hospital Zurich, Zurich, Switzerland

Keywords
Transposition flaps · Spina bifida · Myelomeningocele · Myeloschisis · Fetus · Fetal surgery · Fetal repair · In utero surgery

Abstract
Background: One of the intraoperative challenges of fetal spina bifida repair is skin closure when there is an extended skin defect. Thus, we examined whether distally pedicled random pattern transposition flaps (TFs) are a valid option to overcome this problem. ***Subjects and Methods:*** All patients undergoing in utero repair of spina bifida with application of a TF for back skin closure were analyzed focusing on intraoperative flap characteristics and postoperative flap performance. ***Results:*** In 30 (70%) of the 43 fetuses a primary skin closure was achieved, in 5 (12%) a skin substitute was used, and in 8 (18%) a TF was applied. Flap raising and insertion was uneventful and perfusion was sufficient in all 8 fetuses (100%). In 3 fetuses (37%) the donor sites were closed primarily, and in 5 (63%) a skin substitute was used for coverage. At birth, 7 flaps were viable and provided robust skin coverage over the center of the former lesion. Complications included a small skin defect with CSF leakage in 1 patient (13%). ***Conclusion:*** During open fetal spina bifida repair, TFs can be safely and efficaciously used to obtain solid and durable skin coverage over lesions too large to allow conventional primary skin closure.

Introduction

Worldwide, open fetal surgery for spina bifida is a reality in a few centers after the milestone publication of the MOMS Trial has generated robust evidence that fetal surgery is a viable option for select fetuses suffering from the most devastating survivable neural tube defect, i.e., spina bifida aperta [1, 2].

Generally speaking, spina bifida closure, postnatal and prenatal, is variable, and the best closure technique is not clear. Basically, the operative technique used for fetal back reconstruction is the same as the one we and others apply for postnatal surgery [2]. The goal is to reconstruct

E-Mail karger@karger.com
www.karger.com/fdt

Martin Meuli, MD
Department of Pediatric Surgery
University Children's Hospital Zurich
Steinwiesstrasse 75, CH–8032 Zurich (Switzerland)
E-Mail martin.meuli@kispi.uzh.ch

Table 1. Patient and flap characteristics

Patient No.	GA at fetal surgery	Type of lesion	Donor site coverage	Dura closure	Flap shape	Location of donor site
1	24+4	MS	Alloderm®	direct	3:1	right
2	24+3	MMC	Alloderm®	direct	2:1	right
3	24+4	MS	direct closure	Durapatch	1:1	right
4	26+0	MS	Permacol™	direct	4:1	left
5	25+2	MS	direct closure	Durapatch	2:1	left
6	25+6	MS	Epiflex®	Durapatch	2:1	right
7	25+2	MS	Epiflex®	direct	2:1	right
8	25+0	MMC	direct closure	Durapatch	2:1	right

Patient No.	GA at birth	Repair site	Donor site	Further course	Complications
1	34+5	healed	Alloderm® in situ	spontaneous healing	
2	35+4	healed	healed		
3	36+5	healed	two small skin defects	spontaneous healing	
4	36+6	healed	skin defect	secondary skin closure	
5	33+0	healed	healed		
6	31+6	skin defect	skin defect	surgical revision with duraplasty and skin flap	CSF leakage with temporary external CSF drainage
7	36+2	healed	skin defect	secondary skin closure	
8	37+0	healed	healed		

MMC, myelomeningocele; MS, myeloschisis.

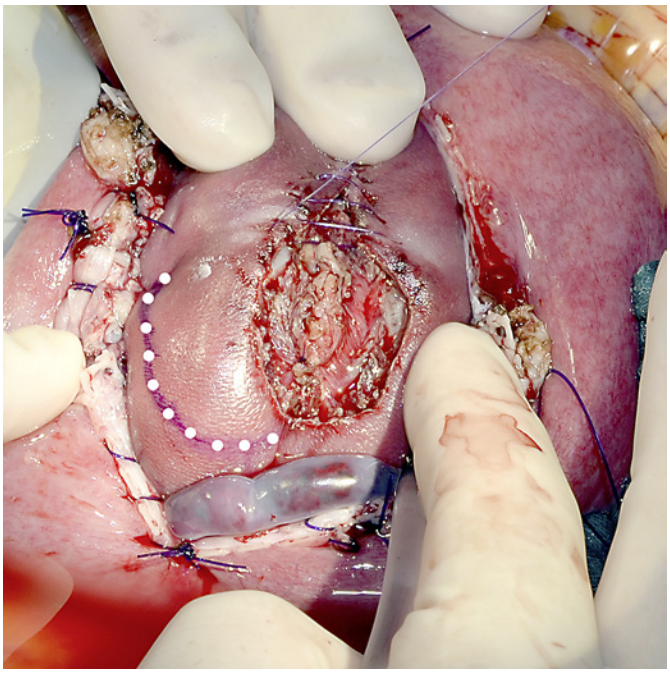
the anatomy as well as possible in order to restore a near normal anatomical architecture at the site of the lesion. The main steps of the intervention include resecting the cystic sac in case of a myelomeningocele (MMC), while in case of myeloschisis (MS), the noncystic variant of this malformation, this step is not necessary. If present, the filum terminale or other pathological attachments and adhesions causing the spinal cord to be tethered must be severed. Thereafter, the open pia (if possible) and dura mater are dissected free and tubularized, completely enclosing the exposed nonneurulated part of the spinal cord in a watertight manner. In order to add one more robust tissue layer over the potentially vulnerable and at least partly functional spinal cord, bilateral paraspinal myofascial flaps are prepared and swung over the open spinal canal where they are sutured together. The last step of the fetal operation is skin closure. In a majority of cases, a primary skin suture with acceptable tension is achievable after extensive skin mobilization. Yet, in about 1 out of 4 cases, primary skin closure is not feasible, mandating alternatives.

There are only few experimental studies where flaps were used in the context of fetal surgery for spina bifida. Meuli-Simmen et al. [3] reported on the successful use of latissimus dorsi flaps to close experimental MMC lesions in fetal sheep.

We hypothesized that distally pedicled random pattern transposition flaps (TFs) might be a valid option for an effective, safe, and definitive skin defect coverage. This article reports on our experience with this technique.

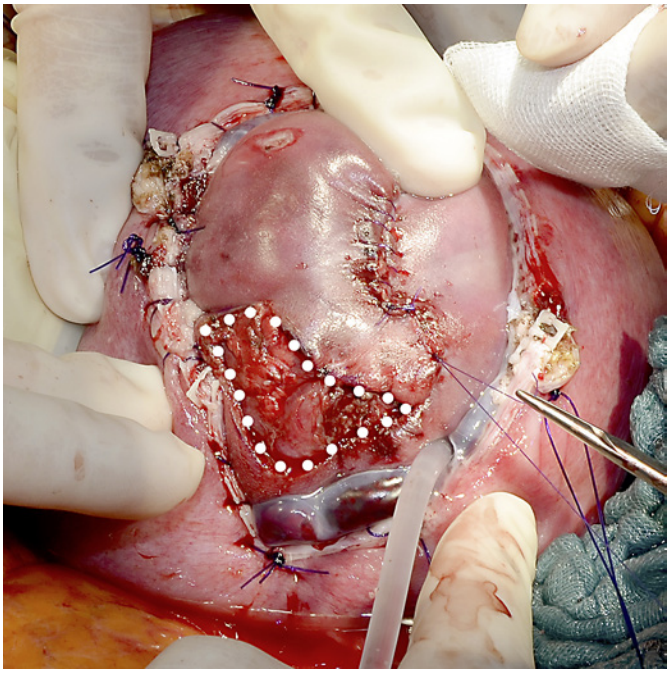
Subjects and Methods

For prospective and comprehensive data collection, we had created a registry for all fetal surgery patients from the start of our program. This registry served as a data repository for the present study that was approved by our local ethics committee (KEK-ZH No. 2015-0172). Between December 2010 and August 2016, a total of 43 patients underwent fetal surgery for spina bifida aperta at the Zurich Center for Fetal Diagnosis and Therapy (www.swissfetus.ch). We used our database to analyze the type and size of spina bifida (MMC or MS) as well as the type of back skin closure (direct, implantation of a skin substitute [1], or TF). Regarding TFs, we analyzed the flap length/flap base ratio, whether the flap could be correctly positioned and sutured over the lesion, whether the flap was viable intraoperatively and at birth, and what the postnatal follow-up was in terms of local flap performance and complications.



Color version available online

Fig. 1. Intraoperative picture during fetal repair showing a large skin defect requiring a transposition flap (white dotted line) for closure. Flap design is done with a tissue marker. As a rule, flap length should not exceed twice the flap base. Technically, cutting the flap is perpendicular to the surface to ensure that a tissue "plate" is raised that consists of the same amount of skin and subcutaneous fat. Flap preparation occurs between fascia and subcutaneous fat, taking care to spare perforating vessels at the flap base.



Color version available online

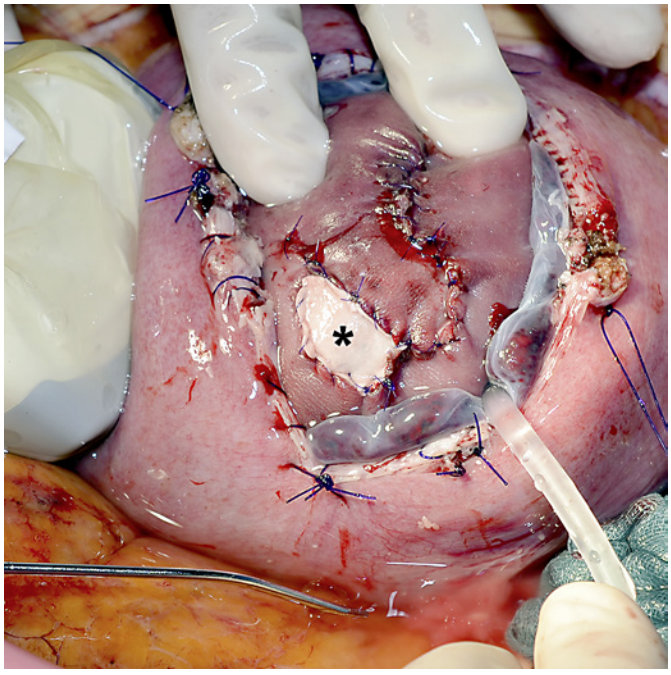
Fig. 2. Transposition and insertion of the transposition flap over the lesion. At the end of this step, all manipulated tissues must be sufficiently perfused for flap survival. The donor site is marked with a white dotted line (same patient as in Fig. 1).

Results

From the total of 43 fetal patients, 28 (65%) had an MMC and 15 (35%) an MS. In 30 fetuses (70%) there was a direct back skin closure, in 5 (12%) a skin substitute was implanted for coverage (four times Integra™, Integra LifeScience, Plainsboro, NJ, USA, and once Alloderm®, LifeCell™, Bridgewater, NJ, USA), and in 8 fetuses (18%) TFs were used for skin closure. The key results regarding TFs are listed in Table 1. In all these 8 patients (100%) flap raising, inserting, and suturing was straightforward. All flaps covered the central area of the repair site and were clinically viable and well perfused at the end of the fetal part of the operation. In 3 cases (37% of all flaps) the donor site could be closed primarily after additional skin mobilization, and in 5 cases (63%) a skin substitute was used for coverage. An illustrative series of pictures is shown in Figures 1–6. In all 8 cases (100%), the postoperative course was uneventful, and all patients were delivered by elective cesarean section. At birth, all 8 flaps (100%) were viable and 7 (87%) were perfectly healed. One baby (13%) demonstrated a significant complication (case 6). There was a central wound dehiscence (diameter 1 cm), probably due to tension, that was closed with a small TF. Removal of the sutures resulted in CSF oozing from one stitch canal. After inserting a temporary local CSF drainage, the leak dried out and remained dry after drainage removal. The flap donor sites in the remaining 7 cases were healed at birth in 3 (38%) cases; they were partly open and healed spontaneously over the ensuing days in 2 (25%) and required a secondary surgical skin closure in 2 (25%).

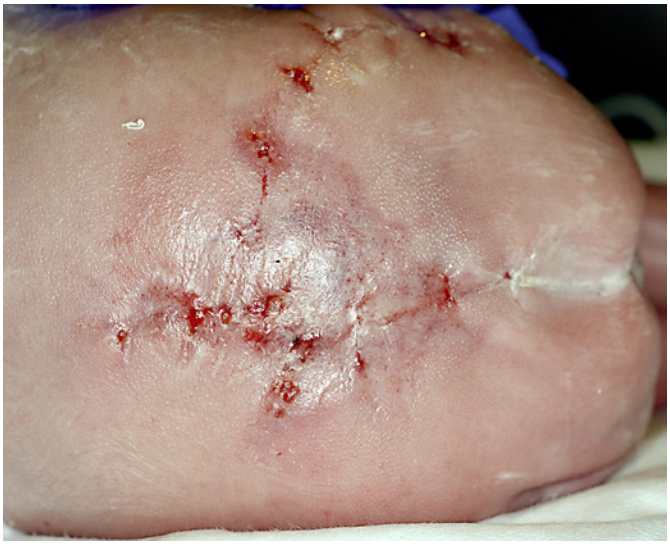
Discussion

This is, to the best of our knowledge, the first article reporting on successful in utero application of TFs for definitive closure of large skin defects during fetal spina bifida repair. TFs are commonly used in adult and also



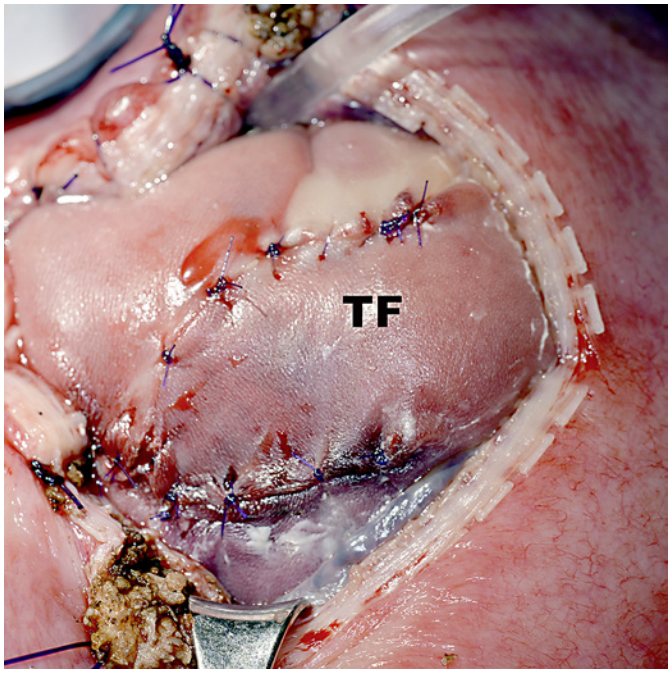
Color version available online

Fig. 3. The donor site is covered with Alloderm (asterisk) (same patient as in Fig. 1 and 2).



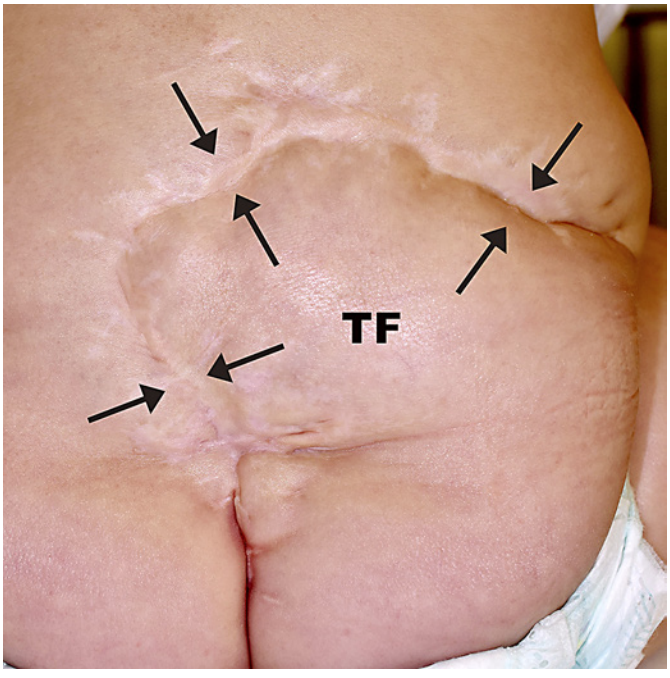
Color version available online

Fig. 5. Picture showing the healed transposition flap immediately post partum (same patient as in Fig. 4). There is only mild scarring.



Color version available online

Fig. 4. Intraoperative picture during fetal repair. A large transposition flap (TF) covering the lesion is seen. Direct closure of the donor site.



Color version available online

Fig. 6. Result at 1 year of age after intrauterine transposition flap (TF) application. Note the perfect flap appearance; however, there is marked scarring (arrows) possibly resulting from former healing that was not tension free.

pediatric plastic and reconstructive surgery [4]. The flap type chosen here is distally pedicled and draws its perfusion from a random pattern vascular supply present at its base. The general rule suggests that the flap length to flap width (pedicle base) should not exceed the 2:1 ratio to ensure sufficient perfusion of the entire flap, in particular the tip. This was the case in 6 cases; in 1 it was 3:1 and in another 4:1. Since these potentially "risky" flaps were well perfused after being sutured in, we assumed that they would survive and saw our hypothesis confirmed at birth. Possibly, fetal skin elasticity, perfusion dynamics, and wound healing capabilities in a midgestational and physiologically hypoxic fetus raise the threshold for tissue necrosis to occur [5, 6]. Also, it is conceivable that perforator vessels, although not formally identified and preserved as in a classical "perforator flap" [4], additionally enhance flap perfusion. While our observations suggest that the 2:1 rule may sometimes be disobeyed, we would suggest that this be done only if absolutely necessary. A number of other issues call for a comment. Empirically, raising and inserting this type of TF did not significantly prolong operating time. The entire maneuver takes about 5 min. Straightforward suturing as well as alternative ways to close the central skin defect, including lateral skin incisions [1, 7, 8] or implantation of skin substitutes (e.g., Alloderm™ [1, 9], Epiflex® (German Institute for Cell and Tissue Replacement GmbH, Berlin, Germany) (we used it twice, not published), or Integra Artificial Skin™ [10]), require similar amounts of time.

Of note, using a skin substitute prenatally appears safe since we did not encounter any complications during the remainder of pregnancy or at birth. Yet, it usually implies a several-week-long spontaneous healing period postnatally (many dressing changes, risk of wound infection, ugly scar) or a secondary postnatal operation for definitive skin reconstruction soon after birth. The latter typically includes a secondary suture (defect size permitting), a simple skin graft (that may scar and shrink and that requires 6–10 days for graft and donor site healing), or a local flap (e.g., rotational, transposition, or Limberg flap [4]). These procedures prolong hospitalization since they interfere with the baseline investigations (repeat ultrasound, craniospinal MRI, bladder manometry, etc.) these neonates require postnatally. Of course, we must admit that in 5 patients (63%) the flap donor site could not be closed in utero, leading to the same undesirable consequence of a secondary intervention. Yet, here the area of interest is not the center of the lesion. We trust that a refined operative technique, e.g., closing the flap donor site with yet another, smaller TF or rotational flap, will lower the incidence of postnatal interventions.

Finally, there are patients (likely those with low MS, see Table 1) that require a dura substitute during fetal repair, particularly in the distal, i.e., sacral, part of the defect. Here, coverage with a well-perfused TF instead of a nonviable material is certainly preferable, especially in cases where there is scarce or no paraspinal tissue to construct myofascial flaps that can be placed over the spinal cord for protection.

In the literature, there is only one article [8] dealing with the problem of in utero skin reconstruction, although it is a frequent and clinically relevant topic. The authors reported using bipedicular bilateral advancement flaps for MS repair. The possible advantages of this approach include a low donor site morbidity and a rather low suture tension. However, for several reasons, we favor TFs. Our approach encompasses one flap instead of two, one donor site instead of two, one or no skin defect instead of two, and no median suture line overlying two deeper median suture lines (one from myofascial flaps, one from dura closure).

Conclusion

Distally pedicled random pattern TF can be safely and successfully used for definitive back skin reconstruction in cases of open fetal spina bifida repair when there is insufficient skin for simple primary back skin closure. It seems possible to "stretch" the 2:1 rule and construct longer than usual flaps with sufficient perfusion.

Disclosure Statement

The authors do not have any commercial interest or other association that might cause a conflict of interest, and they are independent from funders and sponsors.

References

1 Adzick NS, Thom EA, Spong CY, Brock JW III, Burrows PK, Johnson MP, Howell LJ, Farrell JA, Dabrowiak ME, Sutton LN, Gupta N, Tulipan NB, D'Alton ME, Farmer DL: A randomized trial of prenatal versus postnatal repair of myelomeningocele. N Engl J Med 2011;364:993–1004.

2 Meuli M, Moehrlen U: Fetal surgery for myelomeningocele: a critical appraisal. Eur J Pediatr Surg 2013;23:103–109.

3 Meuli-Simmen C, Meuli M, Hutchins GM, Harrison MR, Buncke HJ, Sullivan KM, Adzick NS: Fetal reconstructive surgery: experimental use of the latissimus dorsi flap to correct myelomeningocele in utero. Plast Reconstr Surg 1995;96:1007–1011.

4 Berger A, Hierner R (eds): Plastische Chirurgie. Berlin, Springer, 2003.

5 Adzick NS, Lorenz HP: Cells, matrix, growth factors, and the surgeon. The biology of scarless fetal wound repair. Ann Surg 1994;220:10–18.

6 Bullard KM, Longaker MT, Lorenz HP: Fetal wound healing: current biology. World J Surg 2003;27:54–61.

7 Bruner JP, Tulipan N: Intrauterine repair of spina bifida. Clin Obstet Gynecol 2005;48:942–955.

8 Mangels KJ, Tulipan N, Bruner JP, Nickolaus D: Use of bipedicular advancement flaps for intrauterine closure of myeloschisis. Pediatr Neurosurg 2000;32:52–56.

9 Adzick NS: Fetal surgery for myelomeningocele: trials and tribulations. J Pediatr Surg 2012;47:273–281.

10 Meuli M, Meuli-Simmen C, Flake AW, Zimmermann R, Ochsenbein N, Scheer I, Mazzone L, Moehrlen U: Premiere use of Integra™ artificial skin to close an extensive fetal skin defect during open in utero repair of myelomeningocele. Pediatr Surg Int 2013;29:1321–1326.