Journal of Plastic, Reconstructive & Aesthetic Surgery (2014) 67, 1070—1075

JPRAS
An International Journal of Surgical Reconstruction
www.JPRASurg.com

Assessment of long-term donor-site morbidity after harvesting the latissimus dorsi flap for neonatal myelomeningocele repair

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Received 12 November 2013; accepted 22 April 2014

KEYWORDS
Latissimus dorsi flap;
Myelomeningocele;
Spina bifida;
Functional outcome;
Long-term follow-up

Summary *Background and aim:* The latissimus dorsi flap (LDF) has been employed very successfully over decades to cover large soft-tissue defects. Its donor-site morbidity has been extensively investigated in adults — but not in children — and is considered to be nonrestrictive. The aim of this long-term study was to assess donor-site morbidity with the modified Constant score more than 8 years after coverage of large myelomeningocele (MMC) defects with a reverse latissimus dorsi flap.
Methods: Within the first days after birth, the reverse latissimus dorsi muscle flap was used uni- or bilaterally in three neonates to cover a large MMC defect. Bilateral shoulder function was tested more than 8 years postoperatively according to the modified Constant score.
Results: The mean age at follow-up was 11.7 years. None of the patients experienced any pain or shoulder restrictions during normal daily activities. They all managed to position both of their arms comfortably above the head. Forward flexion was normal in all patients as was abduction and external rotation. Dorsal extension was minimally reduced on the operated side. Internal rotation was symmetric in all patients; the extent of active movement varied from excellent to poor.
Conclusions: Our long-term data suggest that there is no specific and significant impairment of shoulder function after using the distally pedicled reverse LDF for neonatal MMC repair.

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http://dx.doi.org/10.1016/j.bjps.2014.04.018

Introduction

The latissimus dorsi flap (LDF) has been employed over decades to cover large soft-tissue defects in many body locations.[1–5] In neonates, the LDF has been used to close large soft-tissue defects caused by spina bifida.[6–8] Myelomeningocele (MMC) is the most frequent form of spina bifida and typically characterized by an extruded spinal cord onto a sac filled with cerebrospinal fluid. Lower extremity paralysis as well as neuropathic bowel and bladder dysfunction are major disabilities long-term survivors are suffering from. Frequently, an associated hydrocephalus needs to be treated with a ventriculo-peritoneal shunt.[9] Of all congenital lesions of the central nervous system compatible with life, spina bifida is the most complex one. There is a wide geographic and racial difference in prevalence,[10] and despite periconceptional folic acid supplementation, the incidence remains at 3.4 per 10,000 live births in the United States.[11] Early and sufficient closure of the MMC is essential to reconstruct the back architecture and to minimize complications. Therefore, surgical closure is usually performed within the first 24 h of life. Since a few years, there has been even evidence that prenatal surgery improves motor outcome and reduces the need for shunting in select fetuses with MMC.[12] (Of note, there is a Fetal Surgery Program for MMC at the Swiss Center for Fetal Diagnosis and Therapy at the University of Zurich, Switzerland).[8,13] In some postnatal patients, the surgeon faces the challenge that the MMC lesion is too large to allow for the correct multilayer wound closure. In such cases, coverage with a reverse LDF has shown to provide adequate soft-tissue coverage, a technique that has been used by ourselves and others.[6–8] We have published a feasibility study in aborted human fetuses of different gestational ages showing that almost the entire spine can be reached with a proximally or distally pedicled LDF.[14] In addition, the LDF has successfully been used to repair experimental MMC in utero in a fetal sheep model.[15,16] Very recently, we have, for the first time, employed the LDF in a neonate with a history of successful fetal surgery for MMC in order to strengthen the repair site and better protect the underlying spinal cord rescued in utero.[8]

Although the LDF is an excellent option to cover large spina bifida lesions, the literature only provides little evidence regarding donor-site morbidity after unilateral or bilateral harvesting of an LDF in neonates. The only work published in this regard was by Zakaria et al. presenting 11 neonates (operated on day 2–5 of life) with only minor complications after a short follow-up time of <12 months.[7] The aim of our study was to assess donor-site long-term sequelae after more than 8 years with help of the modified Constant score.[17]

Patients and methods

Patients

Three patients born at the University Hospital of Zürich in 1996, 2001, and 2005 received either a unilateral or a bilateral reverse LDF to cover a large MMC defect 2–5 days post birth. Skin closure was achieved by thoracic release incisions or split-thickness skin graft over the muscle flap, or by a combination of both. In all three patients, the entire operative procedure including dura repair was performed by the same team of pediatric and plastic surgeons. Multiple other operations followed over the ongoing years due to concomitant disabilities, and all patients underwent spondylodesis. However, none of these operations affected the shoulder region. Patient data are summarized in Table 1.

Patient 1

One day after cesarean delivery, the thoracolumbar (Th4–L5) MMC repair of the female patient was performed including removal of an intraspinal lipoma. The defect was covered with a reverse latissimus dorsi muscle flap raised from the left side through a separate incision. Preserving the lower lumbar perforating vessels, the flap was mobilized far enough to cover the entire defect after subcutaneous tunneling with sufficient space in order to avoid entrapment between the separate incision and the defect. Bilateral thoracic release incisions allowed the skin to be mobilized enough to achieve direct wound closure over the flap. The incision sites were covered with synthetic wound dressing. One week later, a ventriculo-peritoneal shunt was implanted to drain the hydrocephalus. Four weeks later, the synthetic wound dressing was removed and the former release incisions were closed. All wounds showed uneventful healing.

Table 1 Summarized patient data with reference to surgery.

	Age at surgery	Covering technique	Wound closure	Age at follow-up
Patient 1	2 days	Left reverse latissimus dorsi flap	Bilateral thoracic release incisions and coverage of the release sites with epigard	8 years
	28 days		Epigard removal and direct wound closure	
Patient 2	3 days	Left reverse latissimus dorsi flap	Right thoracic release incision and coverage of the release site with epigard Coverage of the central part of the latissimus muscle with split-thickness skin graft from the left buttock	16 years
	27 days		Epigard removal and direct wound closure	
Patient 3	5 days	Bilateral reverse latissimus dorsi flaps	Coverage of the latissimus muscles with split-thickness skin graft from the scalp	11 years

Patient 2

On the day of cesarean delivery, the male patient was transferred to the Department of Pediatric Surgery with a thoracolumbar (Th11—L5) MMC. Two days later, MMC repair followed and the soft-tissue defect was covered with a reverse latissimus dorsi muscle flap raised from the left side through a separate incision. Dissection included the entire upper part of the muscle and after verification of the lumbar perforating vessels the thoracodorsal pedicle was severed. The muscle was then turned over along the oblique line where the perforating vessels enter the deep surface of the muscle. It was put in place through a subcutaneous tunnel large enough to avoid entrapment, so as to cover the entire defect. Because direct closure of the skin was not possible, a thoracic release incision on the right side was performed, allowing the skin to be approximated much more. The release incision site was covered with synthetic wound dressing. A split-thickness skin graft was taken from the left buttock to cover the remaining defect of 2 × 2 cm over the latissimus muscle. The incision to raise the muscle was closed tension-free. Two weeks later, a ventriculo-peritoneal shunt was installed to drain the hydrocephalus; 4 weeks later, the release incision was closed with minor tension after removal of the synthetic wound dressing. All wounds healed uneventfully.

Patient 3

On the day of cesarean delivery, this female patient with a thoracolumbar (Th10—L5) MMC and an acutely symptomatic hydrocephalus was transferred to the Department of Pediatric Surgery. A ventriculo-peritoneal shunt was installed 1 day later. After 3 days, the large MMC was repaired. Because of the size of the defect, the latissimus dorsi muscle was raised bilaterally as described in the above two cases. The most proximal lumbar perforators were sacrificed on both sides in order to achieve maximum length to place the proximal part of the latissimus muscle as distally as possible. The flap was covered with split-thickness skin graft from the scalp, and the separate incisions to raise the flaps were directly closed. Wound healing was uneventful.

Methods

Assessment of shoulder functional status

To assess and quantify shoulder function, all patients were evaluated according to the modified Constant score.[17] The first, subjective part of this score consists of a questionnaire followed by the second, objective part which assesses shoulder function. The protocol demands the questionnaire to be filled in before the assessment of shoulder function and all movements have to be painless and active. At the assessment of activities of daily living, we replaced the question "How much of your normal work does your shoulder allow?" with "How much of a normal day at school does your shoulder allow?" as our patients are not adults. Active range of motion was measured in degrees with a goniometer. Muscle strength was measured with the IsoForceControl V1.1 device, an improved version of the Isobex apparatus (Medical Device Solutions, Bern, Switzerland) introduced by Gerber.[18] Measurement was performed with the patients sitting in their wheelchairs, as all suffer from lumbar instability. The pronated arm was positioned horizontally in the scapular plane; then patients were instructed to pull the extended arm upwards with the measuring device being attached between the wrist and the floor. The opposite hand was placed on the thigh to prevent it from clinging to the wheelchair creating additional force. Measurements were repeated three times for both sides and the mean value of strength was calculated in kilograms. Finally, scores were calculated for both shoulders of each patient. Per shoulder, a maximum score of 100 points can be achieved, whereof 35 points can be contributed by the subjective assessment and 65 points by the objective measurement. The higher a patient score, the less discomfort he experiences and the better shoulder function is.

Going beyond the assessment of Constant, we additionally measured dorsal extension and adduction strength. These two composite movements are strongly influenced by the latissimus dorsi muscle,[19] but do not have any impact on the Constant score. To measure the strength of adduction, the attachment of the IsoForceControl device was changed, now being spanned between the ceiling and the wrist of the patient. Finally, handedness was determined by asking which hand was used for writing.

Written permission to publish photographic work was obtained from all patients and their parents.

Results

The age at follow-up was 8 (patient 1), 16 (patient 2), and 11 (patient 3) years, respectively (mean age 11.7 years). All patients are right handed. None of the patients experienced any pain during ordinary activities over a 24-h period (15 points). They all managed to position both of their arms comfortably above the head (10 points); subjectively no shoulder limitation was experienced in activities of daily living including sleeping (10 points). Additionally, our patients — all wheelchair bound — did not report any impairment in the use of the mobility aid. Forward flexion was good in all patients (10 points), as was abduction (10 points) and external rotation (10 points). Dorsal extension was good, although slightly reduced on the side of operation (patients 1 and 2) by 6° and 7°, respectively. Results for internal rotation varied among our patients: Patient 2 showed excellent movement skills by easily reaching between the shoulder blades (10 points) (Figure 1), patient 3 reached the 12th thoracic vertebra with the left (8 points) and the waist level with the right hand (6 points). Patient 1 could merely reach behind the buttocks (2 points). Measurement of the abduction strength revealed the following mean values for patient 1: Left 0.97 kg (2 points), right 0.46 kg (1 point); patient 2: Left 2.50 kg (5 points), Right 2.55 kg (5 points), patient 3: Left 1.41 kg (3 points), Right 1.40 kg (3 points). Measurement of the adduction strength revealed the following mean values for patient 1: Left 1.70 kg, right 1.88 kg; patient 2: Left 3.97 kg, right 4.22 kg; patient 3: Left 4.06 kg, right 4.04 kg.

The following Constant scores were achieved per shoulder: Patient 1 scored 69 points for the left and 68 points for the right shoulder. Patient 2 scored 80 points bilaterally. Patient 3 scored 76 points for the left and 74 points for the right shoulder. Table 2 gives an overview of all functional results including the final Constant scores.

Discussion

The latissimus dorsi muscle is the largest of the 26 muscles involved in shoulder joint function.[20] It rotates the arm internally, extends it dorsally, adducts and depresses the raised arm, and rotates the scapula downwards.[19] The first description to use it as a flap dates back to 1906,[21] and it was not before 1976 until Olivari rediscovered it.[22] It has since then become one of the workhorses to cover soft-tissue defects all over the body. Early publications investigating donor-site morbidity stated subjectively unchanged shoulder function.[22–25] Later studies found only minor impairments when examining the full extent of shoulder function after latissimus dorsi transfer.[26–28] The main deficiency was not, as one might expect, a loss of power, but a more rapid onset of fatigue after prolonged activities in which the arms are extended over the head, such as ladder climbing and swimming. No decrease in range of shoulder motion was found.[19] However, all patients included in the studies mentioned were adults. Newborns or children were never assessed.

Figure 1 Full internal rotation of patient 2 on the left, operated side 16 years postoperatively.

Three pediatric patients were included in this study, two receiving a unilateral, one a bilateral reverse LDF to cover a large MMC defect just after birth. Shoulder function was assessed more than 8 years later with help of the modified Constant score,[17] an increasingly used shoulder outcome instrument for adults that has been endorsed by the European Society of Shoulder and Elbow Surgery since 1991.[29] The score is easy to perform, shows good inter-observer reliability, and has been validated as a measurement tool to determine shoulder function.[30] In the same study, normative age- and sex-specific Constant scores including strength values for a large population sample have been reported using the Isobex isometric dynamometer.

Although values for patients <20 years of age are missing, we used the Constant score. To the authors' knowledge, an alternative validated score for teenage patients does not exist.

Movement

All patients show good and symmetric movement of the upper extremities. Dorsal extension was minimally reduced on the operated side (patients 1 and 2). These results are absolutely congruent with long-term results in adults.[19] Internal rotation was the sole function where the maximum score could not be reached by all three patients. Patient 2 showed excellent results (10 points), patient 3 showed good results (6 and 8 points, respectively), and patient 1 showed clearly limited bilateral internal rotation skills (2 points), although only the left side was operated on. Therefore, the harvest of the LDF cannot be made responsible for the limited internal rotation in this case.

Strength

The strength of abduction of the operated side in comparison to the other side was not diminished in patients 1 and 2. This is not surprising with the latissimus dorsi muscle acting on the humerus in internal rotation, dorsal extension, adduction, and depression of the raised arm, but not in abduction. In patient 3, the strength of abduction is difficult to verify despite symmetry, as both sides are operated and normative values in children for comparison are not available.

The strength of adduction, however, was slightly diminished on the harvest side compared to the opposite side in patients 1 and 2. This might be caused by the missing action of the LDF, but the low number of patients does not allow to draw a definitive conclusion. The slight difference might also be related to the handedness of the patients. The extent of strength loss can be assumed in patients 1 and 2 comparing the operated to the not operated side.

Constant score

We did not find a difference in Constant scores between operated and non-operated shoulders in patients 1 and 2. The absolute scores reached by our patients cannot be compared to any previously published scores because of

Table 2 Patient data summary: Range of motion in degrees for forward flexion, abduction, dorsal extension. Rating according to Constant for internal and external rotation. Abduction and adduction strength in kilograms, Constant score. Left = Left shoulder, Right = Right shoulder.

	Forward flexion [°]	Abduction [°]	Dorsal extension [°]	External rotation [points]	Internal rotation [points]	Mean abduction strength [kg]	Mean adduction strength [kg]	Constant score [points]
Patient 1								
Left	159	160	43	10	2	0.97	1.70	69
Right	157	152	49	10	2	0.46	1.88	68
Patient 2								
Left	180	180	37	10	10	2.50	3.97	80
Right	182	177	44	10	10	2.55	4.22	80
Patient 3								
Left	172	173	53	10	8	1.41	4.06	76
Right	182	171	48	10	6	1.40	4.04	74

lack of reference values. Even the largest trial including more than 1700 patients does not provide a subgroup with patients being <20 years of age.[30]

The literature is scarce on reports after harvesting the LDF in children. The only follow-up study included 11 MMC patients and had a mean follow-up of 8.2 months[7]: All patients were reported to have an uneventful course without any complications except for one patient whose skin graft was lost and that had to be re-grafted. Motor function of patients without hydrocephalus was reported to be normal, while patients with hydrocephalus showed variable degrees of motor and sensory deficits. It is understood that functional shoulder examination was not part of the investigation with patients being merely 1 year of age.

Here, we present the first real long-term results more than 8 years after surgery. The only difference found in unilaterally operated patients, when comparing bilateral shoulder function, was both a slightly reduced dorsal extension capacity and a reduced adduction strength on the operated side. Due to the small patient number however, no statistical analysis of these variations could be performed. But since the differences were minimal, we state that harvesting the latissimus muscle in newborn patients seems not to affect the future strength, nor range of motion of the corresponding shoulder in both uni- and bilaterally operated patients. This is of great importance as most patients are wheelchair bound and thus rely upon a normal shoulder function. All our patients were able to independently use their wheelchairs manually without any restriction. They could also reach for the back pockets to grab a comic book, for example. The extent of loss of adduction power cannot be quantified because there are no values to compare our findings to. Therefore, further studies are needed to establish normative age- and sex-specific Constant scores including strength values for comparison.

Conclusion

Our long-term data demonstrate that there is no specific and significant impairment of shoulder function after using the distally pedicled reverse LDF for neonatal MMC repair.

Conflict of interest

None. Additionally, there has not been any kind of funding.

Acknowledgments

We would like to thank Gábor J. Puskás, MD, Department of Orthopaedics, University of Zurich, Balgrist University Hospital, for providing us with the IsoForceControl device to measure muscle strength.

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