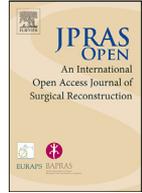Contents lists available at ScienceDirect

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Experience with various reconstructive techniques for meningomyelocele defect closure in India

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A R T I C L E   I N F O

A B S T R A C T

Background: The estimated incidence of spina bifida is 1–2 cases per 1000 population. In earlier literature, the global prevalence of meningomyelocele (MMC) is reported as 0.8–1.0 per 1000 live births. This retrospective study analyses the outcome of various surgical procedures performed for the closure of MMC defects.

Method: A total of 22 patients with MMC defects who underwent repair at our institute from July 2016 to August 2018 were included in the study. A retrospective review of all the cases operated was completed to analyse patient demography including defect size, defect location, surgical procedures, complications and the final outcome.

Results: Out of 22 cases, wherein the neurosurgery department sought help from the plastic surgery department, 11 defects were closed using the Limberg flap technique, 4 defects were closed with either primary closure or the double flap rotation flaps, one defect was closed using the triple rotation flap and 2 defects were closed using the local transposition flap cover technique. Complications were noted in only three cases. One patient had a local wound infection, while in two other cases, wound dehiscence was observed. All 3 cases were managed conservatively. On average, it takes approximately 70 days in India to close such defects.

Conclusion: MMC defects can be effectively managed with local flap options such as Limberg flap, local transposition flap or rotation flaps. Various reasons for the delay in closure were reported in patients late to our centre, when the first point of contact was with other departments.

Introduction

Meningomyelocele (MMC) is one of the two most common types of neural tube defects (NTD) along with meningocele. The estimated incidence of spina bifida is 1–2 cases per 1000 population.[1] In previous literature, the global prevalence of MMC is reported to be 0.8–1.0 per 1000 live births.[5] Embryologically, at approximately four weeks of gestation, the lateral edge of the neural plates fuses to form a tube called the neural tube. Failure of this process will lead to the development of a NTD.[2] Spina bifida occurs when there is failure in posterior fusion.[3]

The aetiology of NTD may include genetics, geography, low socioeconomic status and folic acid deficiency.[4] The main objectives of MMC defect closure are to preserve the neural tissue function and to prevent secondary infection.[7] Closure can be performed either by direct repair or by using several other reconstructive options such as local and regional flaps, musculocutaneous flaps and skin grafting.[9,11] Closure of such defects is a challenging task, as the amount of tissue available in an infant is less, and there are high chances of wound dehiscence.

Methodology

A retrospective observational study was conducted, which included a total number of 22 patients with MMC defects who underwent repair by plastic surgeons from July 2016 to August 2018. Out of the 22 patients, there were 14 males and 8 females. Four patients (20%) had undergone direct repair, 11 patients (55%) had undergone Limberg flap repair and 5 patients (25%) had undergone the rotation flap procedure to cover the MMC defect. Out of 5 rotation flaps, 4 patients underwent double rotation flaps, whereas one patient underwent triple rotation flap to cover the MMC defect. Two patients underwent local transposition flap coverage. In all patients, repairs of the neural tube and dura mater were performed by the neurosurgeon. All MMC defects were closed by plastic surgeons. A review of all the cases operated was completed, and patient demography including defect size, defect location, surgical procedures, complications, and the final results were analysed (Tables 1 and 2).

All 22 patients had lumbosacral defects. The flap was chosen depending on the defect size and shape. All the defects were closed without any undue tension at the suture margins. The mean follow-up period was 6 months. The mean age of the 21 patients operated was 68 days, while one male patient was operated at the age of 16 years. The mean defect size was 6.25 cm^2 in the direct repair cases, 36.5 cm^2 in the double rotation flap cases, 40 cm^2 in the triple rotation flap cases, 49 cm^2 in the Limberg flap cases and 27 cm^2 in the local transposition flap cases. The average operative time was 43.25 min for direct repair, 70 min for the rotation flaps, 59.55 min for the Limberg flaps and 57.5 min for the local transposition flap.

Surgical technique

Direct repair

After the direct repair of the dural defect with non-absorbable sutures, an adjacent skin flap above the muscular layer was undermined to release tension. At first, a few stay sutures were taken; if there was no undue tension, then the wound was closed primarily.

Table 1
Demographic data of patients with meningomyelocele.

S. No.	Age	Sex	Defect size (cm^2)	Type of repair	Total operative time (min)	Post-op complications	Secondary repair	Final outcome
1	1 month	M	5 × 6	Limberg flap	60	None	None	Complete healing
2	3 months	F	3 × 4	Limberg flap	65	None	None	Complete healing
3	10 months	F	6 × 6	Double rotation	70	None	None	Complete healing
4	2 months	M	7 × 6	Double rotation	50	None	None	Complete healing
5	37 days	M	2 × 2	Primary repair	40	None	None	Complete healing
6	4 months	M	11 × 5	Limberg flap	60	Wound Dehiscence	Conservative management	Complete healing
7	16 years	M	8 × 7	Limberg flap	50	None	None	Complete healing
8	45 days	F	5 × 4	Limberg flap	45	None	None	Complete healing
9	61 days	F	8 × 6	Double rotation	80	None	None	Complete healing
10	29 days	M	10 × 4	Triple rotation	90	Infection and Necrosis	Debridement & direct repair	Complete healing
11	48 days	M	3 × 2	Primary repair	40	None	None	Complete healing
12	50 days	F	6 × 4	Local transposition Flap	45	None	None	Complete healing
13	38 days	M	3 × 2	Primary repair	48	None	None	Complete healing
14	42 days	F	9 × 8	Limberg flap	90	None	None	Complete healing
15	43 days	M	4 × 2	Limberg flap	40	None	None	Complete healing
16	63 days	F	3 × 3	Primary repair	45	None	None	Complete healing
17	40 days	M	8 × 8	Limberg flap	60	None	None	Complete healing
18	50 days	M	5 × 4	Double rotation	60	None	None	Complete healing
19	4 months	F	10 × 8	Limberg flap	55	None	None	Complete healing
20	2 months	M	11 × 7	Limberg flap	65	None	None	Complete healing
21	42 days	M	6 × 5	Local transposition Flap	70	Wound Dehiscence	Conservative management	Complete healing
22	2 months	M	8 × 8	Limberg flap	65	None	None	Complete healing

M – Male, F - Female.

Figure 1. (A) Meningomyelocele in a 2-month-old child, (B) Limberg flap planned to cover the defect, (C) Final inset of the Limberg flap into the defect.
Figure (1′) Line diagram of the Limberg flap cover.

Limberg flap

Limberg flap is a type of transposition flap. The Limberg flap is a parallelogram with two 120° angles and two 60° angles (Figure 1). Once the dural defect was closed by the neurosurgeon, the defect shape and size were assessed and marking was done for the Limberg flap. The margin of the defect was then trimmed into a parallelogram to act as the Limberg flap. A line (equal in length to the short diagonal of the parallelogram) was extended from the parallelogram as a continuation of the short diagonal. Drawing a second line from the distal end of the first line forms an apex of the flap. The second line was equal in length and parallel to one of the adjacent defect sides. A skin incision was made to the muscle fascia first, and then the Limberg flap was dissected above the muscle fascia.[10] After the dissection, the Limberg flap was transposed to the MMC defect and suturing was done by the standard surgical technique (Figure 1 and Figure 1′).

Double rotation flap

In the double rotation flap, pedicles were split in the opposite direction to the vertical of the defect. These flaps were marked from the upper and bottom edges of the circular defect and needed a basis that was 1.5 times bigger than the largest diameter. Again, the flaps were dissected in the subfascial plane and transposed to close the defect (Figure 2 and Figure 2′).

Triple rotation flap

Around the primary defect, three flaps were raised by marking an arc around the defect. The length of the arc was 1.5 times the diameter of the defect and at an angle of 30°–50°. With good undermining, three arcs were rotated into the defect and sutured first at the centre and then laterally (Figure 3 and Figure 3′).

Local transposition flap

In the planning of the local transposition flap, the defect was triangulated first. The transposition flap was designed as a rectangle in a ratio of not more than 3:1 immediately adjoining the defect and was moved laterally to cover the defect. The base of the flap was alongside the apex of the triangle (Figure 4 and Figure 4′).
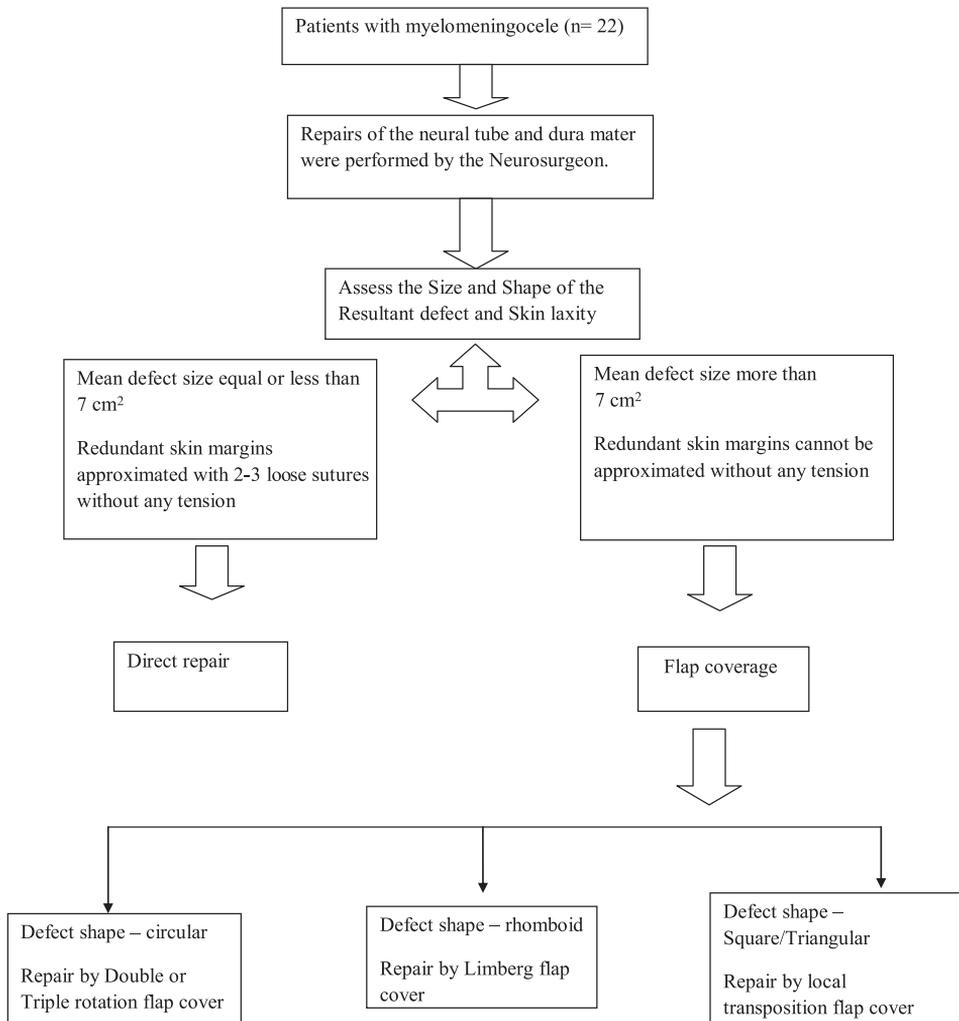
Decision-making algorithm

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                    ┌─────────────────────────────────────┐
                    │  Patients with myelomeningocele (n= 22)  │
                    └─────────────────────────────────────┘
                                    │
                    ┌─────────────────────────────────────┐
                    │  Repairs of the neural tube and dura mater  │
                    │  were performed by the Neurosurgeon.  │
                    └─────────────────────────────────────┘
                                    │
                    ┌─────────────────────────────────────┐
                    │  Assess the Size and Shape of the     │
                    │  Resultant defect and Skin laxity     │
                    └─────────────────────────────────────┘
                                    │
        ┌──────────────────────────┐   ┌──────────────────────────┐
        │ Mean defect size equal or less than │   │ Mean defect size more than │
        │ 7 cm²                     │   │ 7 cm²                     │
        │                           │   │                           │
        │ Redundant skin margins    │   │ Redundant skin margins cannot be │
        │ approximated with 2-3 loose sutures │   │ approximated without any tension │
        │ without any tension       │   │                           │
        └──────────────────────────┘   └──────────────────────────┘
                    │                               │
            ┌──────────────┐                ┌──────────────┐
            │ Direct repair │                │ Flap coverage │
            └──────────────┘                └──────────────┘
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Mean defect size equal or less than 7 cm^2

Redundant skin margins approximated with 2-3 loose sutures without any tension

Mean defect size more than 7 cm^2

Redundant skin margins cannot be approximated without any tension

Defect shape – circular

Repair by Double or Triple rotation flap cover

Defect shape – rhomboid

Repair by Limberg flap cover

Defect shape – Square/Triangular

Repair by local transposition flap cover

Table 2

Result summary.

Groups	Direct repair	Limberg flap	Double rotation flap	Triple rotation flap	Local transposition flap
No. of patients	4	11	4	1	2
Mean defects (cm^2)	6.25	49	36.5	40	27
Mean operative Time (min)	43.25	59.55	65	90	57.5
Complications	None	One	None	One	One

Results

A total of 22 patients underwent surgical closure of MMC defects. All patients had lumbosacral defects. All defects were successfully closed either primarily or with local flaps. The mean follow-up period was 6 months. The mean age of the 21 patients operated was 68 days, while one male patient was operated at the age of 16 years. The mean defect size was 6.25 cm^2 in the direct repair cases, 36.5 cm^2 in the double rotation flap cases, 40 cm^2 in the triple rotation flap cases, 49 cm^2 in the Limberg flap cases and 27 cm^2 in the local transposition flap cases. The average operative time was
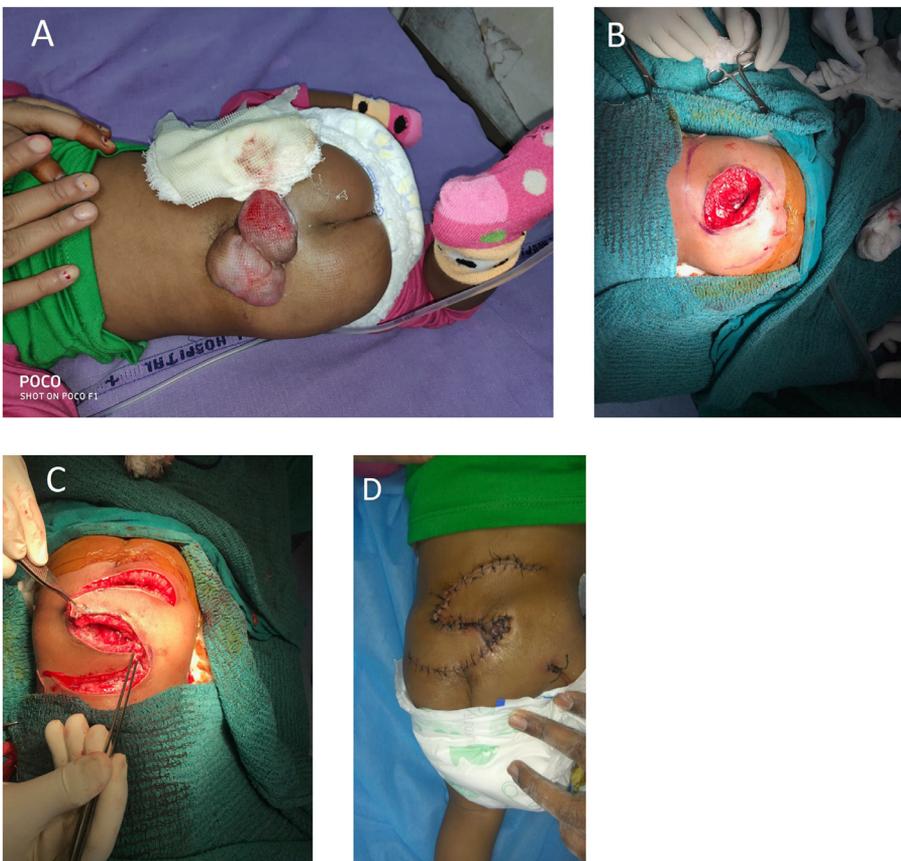
Figure 2. (A) Meningomyelocele in a 61-day-old child**,** (B) Defect, (C) Double rotation flap coverage of the defect, (D) Post-op image after 5 days.
Figure. (2′) Line diagram of the double rotation flap.
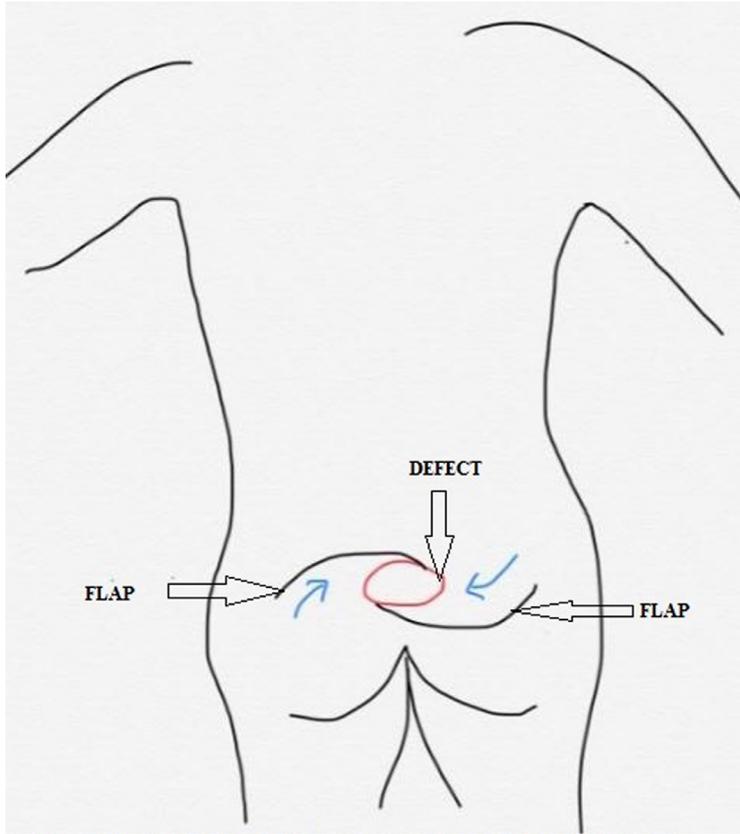
Figure 2. Continued

43.25 min for the direct repair, 70 min for the rotation flaps, 59.55 min for the Limberg flaps and 57.5 min for the local transposition flap.

Complications were noted in only three cases. The patient who underwent the triple rotation flap (for a defect size of 40 cm^2) had developed flap necrosis, and a local wound infection at the tips of the rotation flaps was seen during the immediate postoperative period. The patient was managed with further wound debridement and the direct repair of the wound. Another patient who underwent defect closure (defect size of 55 cm^2) with the Limberg flap had developed wound dehiscence in the post-op period, which was managed conservatively with regular dressings and healed secondarily. In the third case, wound dehiscence was noted in a patient who underwent the local transposition flap for closure of the defect (defect size of 30 cm^2). Subsequently, it was also managed conservatively with regular moist dressings and healed secondarily. All other remaining wounds were healed successfully with no further complications (Tables 1 and 2).

Discussion

Primary healing of small defects can be attained by a wide undermining of wound edges and tensionless primary repair. In a case series of 130 patients, Patterson and Till mentioned that most of the MMC defects were closed by direct repair, while other reconstructive options were required in the remaining cases, that is, only 25%.[8] To close the larger defects has always been challenging. Various options ranging from a simple skin graft to the local flaps to various regional flaps or even the perforator flaps have been mentioned in the literature.
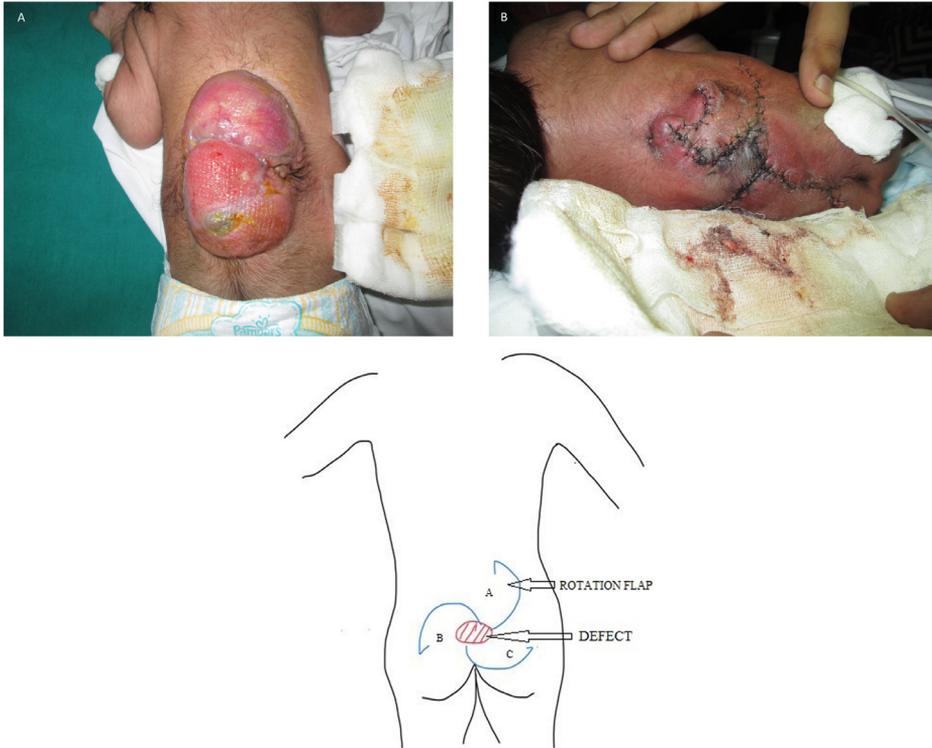
Figure 3. **(A) A** large meningomyelocele in a 29-day-old child, (B) Triple rotation flap coverage of the defect with dehiscence of wound during the post-op period, which was then managed with debridement and direct closure.
Figure (3′) Line diagram of a triple rotation flap.

Musculocutaneous flaps are an option for the closure of extremely wide MMC defects as described by Mustarde in 1968,[12] as shown in 1978, when McCraw et al.[13] had used bilateral latissimus dorsi musculocutaneous flaps for the closure of MMC. McDevitt et al. in 1982 had shown the use of bilateral latissimus dorsi musculocutaneous flaps with extended gluteal fasciocutaneous flaps for the closure of thoracic and lumbar dysgraphic defects.[14] Other options mentioned in the literature were distally based latissimus dorsi flaps,[3] Limberg latissimus dorsi musculocutaneous flaps[15] and reverse latissimus dorsi musculocutaneous flaps.[16] Ramirez et al. had shown in 1987 that the combination of latissimus dorsi and gluteus maximus musculocutaneous flaps could be used for lower sacral defects.[17] These flaps did not gain much popularity owing to extensive flap dissection, increased blood loss and prolonged operative time, thereby limiting their use.

The utility of the perforator flaps by El-Sabbagh in 2011 and Duffy et al. in 2004 had also been previously described.[19,20] The limitation of these perforator flaps is that these flaps require expertise with microsurgical instrumentation and the microscope.

More recently, in 2012, Patel et al.[18] had described the local turnover fascial flaps and the midline linear skin closure as methods of repair for the extradural MMC defects.

We had operated in a total of 22 cases, out of which 11 defects were closed using the Limberg flap technique, 4 each with primary closure and the double flap rotation flap closure technique, one with the triple rotation flap closure and 2 with the local transposition flap cover technique.

In previous literature, it had been emphasised that the use of local flaps for moderate to large lumbosacral MMC defects can be considered as a viable option. Other options included were the double Z-rhomboid flaps, V-Y advancement flaps, bilobed flaps, rotation flaps and Limberg flaps.[6,21–23] |The first mentioned use of the Limberg flap for closing MMC defects was stated by Ohtsuka et al.[23] in
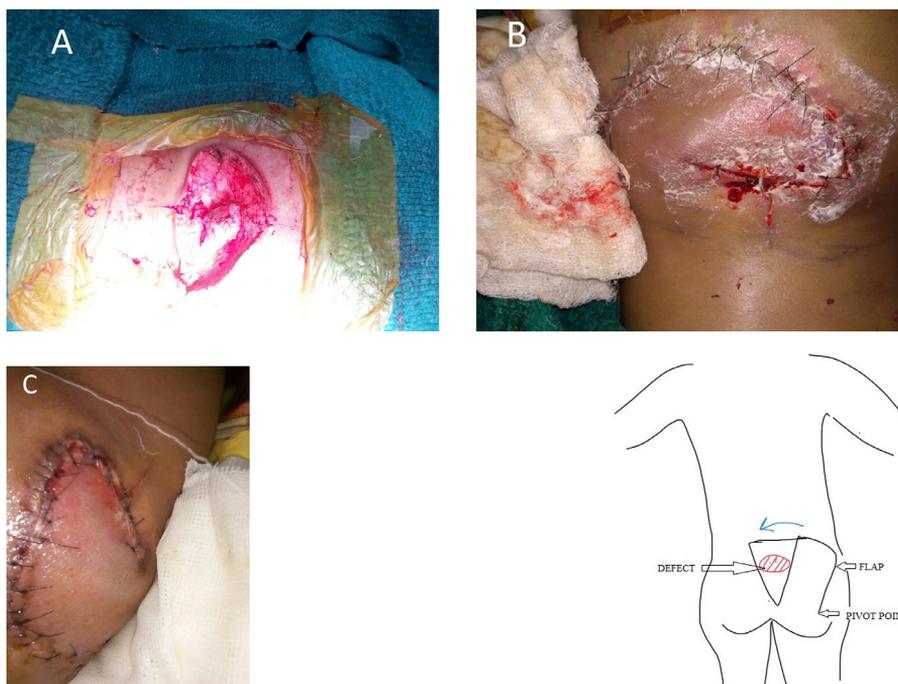
Figure 4. (A) Defect in a 42-day-old child after excising the meningomyelocele, (B) Local transposition flap coverage for the defect triangulated before, (C) Wound dehiscence at the upper end managed conservatively.
Figure (4′) Line diagram of the transposition flap.

1979. Cruz et al.[21] also described the use of the Limberg flap for closure and lately, Shim et al.[24] had shown the utility of the Limberg flaps for the closure of MMC. In the mentioned study by Shim et al., the mean age of the patients was 32 days when compared with our study, which was 68 days, while one male patient was operated at 16 years of age; there was a significant difference in terms of average operative times, that is, 190.9 min and 220 min compared to 43.25 min and 59.55 min in the direct and the Limberg flap closures, respectively, of our study. One patient was operated with the double Limberg flap in the study by Shim et al. whereas none of the patients were operated with the double Limberg flap. Similarly, the local transposition and the triple rotation techniques were used in our study, which were not seen in the study by Shim et al.

We selected these flaps because the Limberg and the rotational flaps make use of the nearby skin laxity, anywhere from the upper back up to the gluteal region. These flaps have an additional advantage of the preservation of back muscles unlike the musculocutaneous flaps so that the correct posture development of the trunk may not be hampered in the future. The mean operative time of the Limberg flap procedure was not significantly more when than the primary closure technique. Most of the flaps survived without any significant post-operative complications except two of the cases. In one case, where the defect was closed using the triple rotation, wound infection and flap tip necrosis were managed with debridement and direct closure. In the second case, where a large defect was closed using the Limberg flap technique, wound dehiscence in the immediate post-operative period occurred due to the closure in tension; this case was managed conservatively.

One of the uncommon complications is that of dural leaks. In case of dural leaks, we coordinate with the neurosurgery team for its management. If these dural leaks act as low-output fistulas, they are managed conservatively. But if the fistula has high output, as decided by the neurosurgery unit, then dural repair is warranted and carried out either with the tight re-suturing of the dura with fibrin sealants or with the dural patch or the fascial graft repair. None of our patients had encountered

any dural leaks in the post-operative period. According to the protocol, which is followed in the neurosurgery department of our institute, the indications for the use of the VP shunt are the rapidly evolving hydrocephalus with increased intracranial pressure and high output CSF leakage through a ruptured MMC sac.

It was observed that most of the children were referred to us at an older age, with the mean age of 68 days in 21 of the patients and one patient was operated at 16 years of age. The mentioned mean age is still higher than that in a previous study conducted by Shim et al. in 2013.[24]

According to earlier literature, the best time for MMC defect closure is within 48 h of birth. Early surgical intervention is associated with reduced mortality, lesser incidence of meningitis and improved muscle function. In addition, the technical difficulties are much less during the first 48 h of life than they are at a later age.[25] In a developing country like India, most of the deliveries occur in remote areas where superspecialty services may not be available, and it might take a long period of time before the patient reaches a tertiary care centre. One of our patients presented at the age of 16 years, a much-delayed presentation, only related to the cosmetic deformity, can be seen if it is not associated with any functional problem. The first point of contact for these patients is the paediatric department, who refers them to the neurosurgery and plastic surgery teams for surgical intervention. In high-volume centres like ours, sometimes it might take days to weeks to arrange the OT if the condition is not life threatening. It is advisable for those who first see the patient at birth to consider the MMC as a surgical emergency requiring immediate closure and refer them to an appropriate centre.

When we compared the outcomes of delayed repair with those of immediate repair, we drew the conclusion that immediate repair of MMC is always preferable to avoid complications such as meningitis and to protect the exposed neural placode. The patients who presented late often have the tethering of the spinal cord to the placode. The sac is epithelialised in late presentation; therefore, delayed repair involves the MMC closure to prevent sac rupture, correct the deformity and remove the skin from the placode. Hence the chances of skin/flap necrosis are higher in the delayed repair. Other delayed outcomes are cerebrospinal fluid leak, retethering and increased chances of injuries to the neural structures while dissecting the large disfiguring sacs, leading to paralysis or other deformity.

Limitation of the study

First, the cases were not followed up for a longer duration. Most of the patients were not brought up for follow-up by their parents once the wound healed. Second, sufficient images were not available of the defect that was closed using triple rotation flap technique.

Conclusion

We have seen with this study that there was a long delay in the repair. Various reasons were attributed to the delay, which involved the patients reporting late to our centre and the first point of contact was departments such as neurosurgery, paediatric or emergency, which then referred these patients to our department if required.

With this study, we concluded that for larger defects in MMC, the Limberg flap and the rotational flaps proved to be an effective armamentarium for the repair, whereas direct closure can still be considered for smaller defects.

Declaration of Competing Interest

None.

Funding source

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Declaration

The study is in accordance with the ethical standards of the responsible committee on human experimentation (Institutional or Regional) and with the Helsinki Declaration of 1975, as revised in 2000.

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