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Keystone Flap for Coverage of Myelomeningocele Defects: A Simple Solution to Complex Reconstructive Problem

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Abstract

Background: The management of myelomeningocele (MMC) defects poses significant reconstructive challenges. Numerous methods of reconstruction have been described. Nevertheless, the choice of the ideal reconstructive technique is still controversial. The keystone perforator island flap (KPIF), an emerging reconstructive option that is gaining popularity among reconstructive surgeons in many parts of the body. Herein, we aimed to evaluate the efficacy, safety, and surgical outcomes of using KPIF for the coverage of MMC defects. **Methods:** From April 2022 to April 2024, 16 consecutive newborns (10 male, 6 female) at a mean age of 4.4 days (1 - 15 days) with MMC defects were included in this prospective study. They underwent reconstruction using KPIF. The mean defect size was 5.6 × 5.1 cm (range, 4 × 4.5 cm to 8 × 6 cm). The defects were lumbar in 2 patients, thoracolumbar in 5 patients and lumbosacral in 9 patients. Operative time and early post-operative complications were recorded. Objective assessment regarding the durability of flaps and scar quality was done. Parents' satisfaction with esthetic outcome was assessed. **Results:** Surgery time lasted between 60 and 110 minutes (mean: 83.8 minutes), excluding the neurosurgical repair. We used KPIF type III in 12 patients, and KPIF type IV -Omega variant in 4 patients. In all patients, tension-free closure was achieved, and healing was successful without any complications, except for 1 small wound dehiscence (<2 cm), which healed with conservative wound management. There was no case with a late breakdown of the wound or associated CSF fistula formation for a mean of 10.6 months (range, 6 - 18 months). 14 patients had thin, narrow scars, while 2 patients had wide, flat scars, and 87.5% subjective cosmetic satisfaction was noted. **Conclusions:** The keystone

flap is an ideal reconstructive option for MMC defect closure as it is simple, reliable, versatile, and cost-effective treatment modality, offering advantages over traditional methods by improving patient outcomes and minimizing complications.

Keywords

Myelomeningocele, Keystone Flap, Perforator Flap, Island Flap

1. Introduction

Myelomeningocele (MMC), a severe form of spina bifida [1], is the commonest congenital neural tube defect that results from fusion failure in the spinal part of the neural tube with subsequent protrusion of the meninges and often dysplastic spinal cord through a vertebral defect, most commonly in the lumbosacral region [2] [3]. The incidence of MMC is approximately 4 per 10.000 births worldwide [4]. Although compatible with life, this disease poses significant challenges due to its associated life-long morbidity, including paraplegia, bowel and bladder dysfunction, hydrocephalus, increased risk of CNS infection, and severe mental retardation in up to 15% [5].

Ideally, early surgical intervention within the first 48-72 hours of life is critical to reduce the risk of cerebrospinal fluid (CSF) leakage, CNS infection, and further neurological damage [6]. Surgical management involves not only watertight neurosurgical repair of the dural defect but also well-vascularized, reliable, and durable soft tissue coverage to protect the repair and prevent infection [7]. While small MMC defects can often be closed primarily, larger defects can preclude 1ry closure and present a reconstructive challenge due to insufficient adjacent soft tissue and the need for tension-free closure [8]. Traditional reconstructive techniques, including skin grafts, local fasciocutaneous flaps, musculocutaneous flaps and muscle flaps have been employed to treat large defects [9]-[11]. Although effective in many cases, some of these methods can be complex, time-consuming, and associated with complications such as CSF leakage, wound dehiscence, flap loss and tip necrosis [12].

The keystone perforator island flap (KPIF) was 1st described by Behan in 2003 [13]. The keystone is the apical curvilinear stone of Roman architecture [14]. The KPIF is an island fasciocutaneous flap based on musculocutaneous perforators and neurocutaneous connections that provide reliable vascularity [4]. Its design allows for en bloc movement of well-vascularized skin, cutis, and fascia, making it particularly suitable for complex defects [15]. Moreover, it distributes wound tension across a large area, reducing the risk of tissue breakdown and scar stretching [16]. Its application in locoregional reconstruction in various parts of the body has demonstrated promising outcomes with low morbidity [17] [18]; nevertheless, its use in covering MMC defects remains relatively novel and underreported. The

aim of the present study was to evaluate the efficacy, safety, and surgical outcomes of using the KPIF as a reconstructive technique for the coverage of MMC defects.

2. Materials and Methods

This prospective study was approved by our University Ethical Committee (36264PR1360/9/1) and all the patients' parents or legal guardians provided written informed consent regarding the treatment, photography and publication. The study was adhered to the principles of the Declaration of Helsinki. Between April 2022 and April 2024, sixteen patients (10 male and 6 female) with myelomeningocele underwent KPIF reconstruction at our plastic surgery department after neurosurgical repair. The mean age was 4.4 days (range, 1 - 15 days). The location of myelomeningocele was lumbar in 2 patients, thoracolumbar in 5 patients and lumbosacral in 9 patients. The dimensions of MMC soft tissue defects range from 4 × 4.5 cm to 8 × 6 cm with a mean of 5.6 × 5.1 cm. Inclusion criteria were as follows: Neonatal patients with confirmed diagnosis of MMC via obstetric ultrasound or clinical examination at birth and with large skin defects that are inappropriate for 1ry closure by a multidisciplinary team of neurosurgeons and plastic surgeons (≥3 cm in width). All cases had flaccid paralysis of both lower limbs and associated hydrocephalus. Patients with life-threatening congenital anomalies, sever kyphosis, small defects amenable to 1ry closure and those who had previous surgeries were excluded from the study. Patient data are summarized in Table 1.

Table 1. Patient data.

No.	Sex/Age, day.	Defect site	Defect size (length × width), cm.	Type of KPIF	Operative time*, min.	Blood transfusion	Complications	Length of hospital stay, day.	Scar quality	Esthetic outcome	Follow up, mon.
1	M/4	Thoracolumbar	6.5 × 5.5	Type **III**	110	No	No	18	Thin narrow	Satisfactory	10
2	F/3	Lumbosacral	4.5 × 5	Type **III**	70	No	No	10	Thin narrow	Satisfactory	12
3	M/3	Lumbosacral	5 × 5	Type **III**	75	No	No	12	Thin narrow	Satisfactory	18
4	M/4	Lumbosacral	4 × 4.5	Unilateral type **IV (Omega variant)**	70	No	No	18	Flat wide	Unsatisfactory	10
5	M/15	Thoracolumbar	6 × 5.5	Type **III**	90	No	No	15	Thin narrow	Satisfactory	12
6	M/1	Lumbosacral	5.5 × 5	Type **III**	75	No	No	12	Thin narrow	Satisfactory	10
7	F/4	Lumbar	5 × 5	Type **III**	85	No	No	18	Thin narrow	Satisfactory	8
8	M/5	Lumbosacral	6 × 5	Type **III**	90	No	No	12	Thin narrow	Satisfactory	5
9	M/7	Lumbosacral	6.5 × 5	Type **III**	80	No	No	10	Thin narrow	Satisfactory	18
10	F/3	Thoracolumbar	7 × 6	Bilateral type **IV (Omega variant)**	100	No	Small wound dehiscence	25	Flat wide	Unsatisfactory	6
11	M/1	Lumbosacral	6 × 5	Type **III**	90	No	No	14	Thin narrow	Satisfactory	14
12	F/2	Lumbosacral	5 × 5	Type **III**	85	No	No	14	Thin narrow	Satisfactory	6
13	F/5	Thoracolumbar	6 × 5.5	Type **III**	80	No	No	12	Thin narrow	Satisfactory	8
14	M/11	Thoracolumbar	8 × 6	Type **III**	110	No	No	14	Thin narrow	Satisfactory	5

Continued

15	M/2	Lumbar	5 × 4	Unilateral type **IV (Omega variant)**	60	No	No	20	Thin narrow	Satisfactory	12
16	F/1	Lumbosacral	4 × 4.5	Unilateral type **IV (Omega variant)**	70	No	No	10	Thin narrow	Satisfactory	5

M, male; F, female; KPIF, keystone perforator island flap, *Operative time excluding the neurosurgical repair.

2.1. Surgical Technique

All procedures were performed under general anesthesia with endotracheal intubation. Patients were placed prone on the operating table with all pressure points padded. Intravenous prophylactic antibiotics were administrated and routinely maintained for a week after surgery (cefuroxime 50 mg/kg every 12 hours). The neurosurgical team performed standard neural tube and meningeal closure. The surgery was then continued by our plastic surgery team. Following debridement of the macerated skin to healthy margins and meticulous hemostasis, the size of the soft tissue defect was measured, and the laxity of the surrounding tissues were assessed.

The keystone perforator island flap (KPIF) was used for reconstruction. By using a handheld doppler, we identified the location of the surrounding perforators. The KPIF was drawn around the MMC defect with the perforators included in the middle of the flap. The keystone flap is curvilinear trapezoidal in shape with two conjoined V-Y advancement flaps. The width of the flap was marked equal to 1:1 to 1.5 ratio the defect width, and the length of the flap was determined by drawing two right angled lines, each equal to the width of the defect, from the vertices of the defect. Then a curved line was drawn to join the two right angled lines to create the keystone design. A unilateral keystone flap was used for defects ≤ 4.5 cm in width, while double keystone flaps were used for defects > 4.5 cm in width. For transversely oriented defects or those with limited lateral tissue laxity, a modified rotation-advancement flap was used (KPIF type IV-Omega variant).

After the flap was outlined, skin incisions were made and continued through the subcutaneous fat to lumbar fascia, while the muscles were left intact. The lateral and medial edges were undermined by vertical spreading of the fibrous septa using blunt-tip scissors that improved flap advancement whilst preserving their perforator vascularity. Attempt to close the MMC defect was started centrally by direct apposition of the defect edges. Then, the V-Y advancement of both the proximal and distal ends of the flap in the longitudinal axis is performed to create more skin laxity in the central portion of the flap and to decrease the horizontal tension (**Figure 1**). If complete defect closure wasn't obtained, the upper and lower portions of the flap were fully elevated in the subfascial plane while keeping the central "hot spot", including the lumbar and intercostal arteries perforators. Then, the two limbs and margins were advanced, rotated and conjoined to close the defect, creating a fortune cookie shape (**Figure 2**). Simple inverted absorbable sutures were used for subcutaneous tissue closure. The skin was closed by simple interrupted nonabsorbable monofilament sutures. All secondary defects were closed primarily.

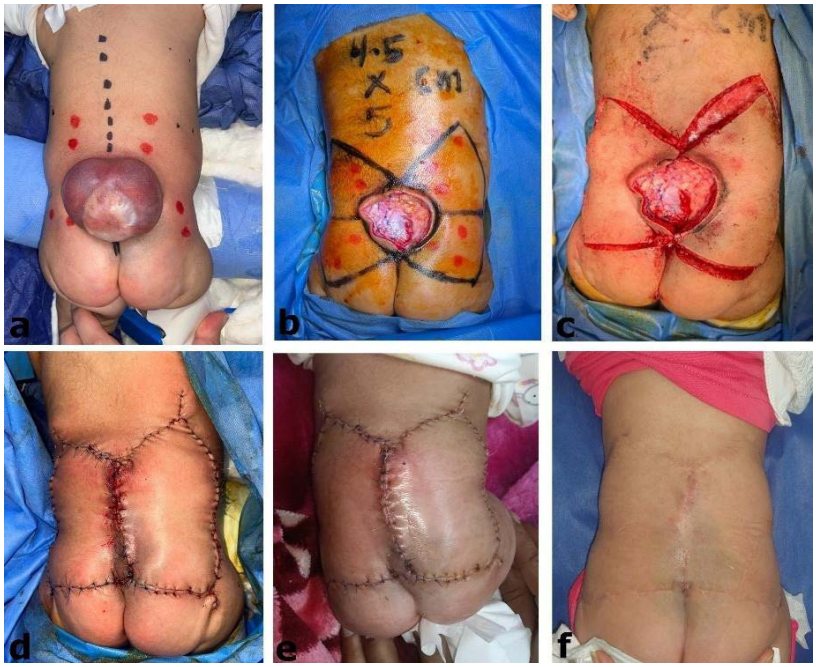

Figure 1. A 3-day-old female newborn with a lumbosacral myelomeningocele was reconstructed with type III keystone flap. (a) Preoperative photo. (b) Intraoperative photo after dural repair and marking of flaps. (c) Intraoperative photo after dissection of flaps. (d) Immediate intraoperative photo after reconstruction of the defect. (e) Patient at 7 days postoperative. (f) Patient at 11 months postoperative.

Figure 2. A 2-day-old male newborn with a lumbar myelomeningocele was reconstructed with Omega modification of a type IV keystone flap. (a) Intraoperative photo after dural repair and marking of flaps. (b) Immediate intraoperative photo after reconstruction of the defect. (c) Patient at 6 days postoperative. (d) Patient at 3 months postoperative.

2.2. Postoperative Care and Follow Up

At the end of the operation, light dressing was applied with an open window to allow flap monitoring. The patients were kept on prone and lateral positions strictly for three weeks, after which they were allowed to be positioned on their backs. The sutures were removed after 10 - 15 days. Patients were followed postoperatively once monthly for six months, then every three months for two years. All cases were examined for the occurrence of early postoperative complications including venous congestion, seroma, hematoma, partial or total flap necrosis, wound dehiscence, wound infection, and CSF leak. A routine long-term follow-up was conducted regarding the cosmesis of scars and durability of flaps. Scar quality was reported as thin narrow, flat wide, and thick raised. The parents' subjective satisfaction regarding the esthetic outcome was also documented.

SPSS for Windows (Version 18.0, SPSS Inc., Chicago, IL, USA) was used for statistical analysis. Quantitative data were expressed as range and mean ± standard deviation (SD), whereas qualitative data were expressed as number and percentage.

3. Results

All cases had uneventful operations. The operating time lasted between 60 and 110 minutes (mean: 83.8 minutes), excluding the neurosurgical repair. No perioperative or intraoperative blood transfusion was required. Twelve patients (75%) were reconstructed by two double opposing keystone flaps (KPIF type III) (**Figure 3**), and four patients (25%) were reconstructed by the omega-shaped modification (KPIF type IV-Omega variant) (3 unilateral and 1 bilateral) (**Figure 4**). Immediate venous congestion and noticeable increase in flap volume were noted in all cases, which were completely resolved spontaneously within a few days. Only one (6.3%) postoperative small wound dehiscence (<2 cm) was encountered in case 10, which was completely healed with conservative wound care procedures. Otherwise, all wounds healed without any evidence of early complications such as seroma, hematoma, partial or total flap necrosis, wound infection, and CSF leak. All cases were discharged within 10 to 25 days (mean: 14.6 days).

After a mean follow-up period of 10.6 months (range, 6 - 18 months), all cases showed no evidence of late complications such as CSF fistula or wound breakdown with durable soft tissue coverage. Fourteen patients (87.5%) had thin, narrow scars, while two patients (12.5%) had wide, flat scars. The final scars on the back of the patients were usually inconspicuous after six months. Fourteen parents (87.5%) were satisfied with the cosmetic results, while two parents (12.5%) were not pleased by the wide scars, which necessitated three to four sessions of fractional CO_2 laser to ameliorate.

4. Discussion

Surgical management of large MMC defects has always been a challenging reconstructive problem. Many surgical techniques are reported for the closure of such

defects. Nevertheless, each reconstructive modality has its own advantages and limitations [2]. Skin grafts, while simple and straightforward procedure, carry risks of ulceration, infection, and poor durability, particularly in weight-bearing areas like the lumbosacral region [19]. Local random fasciocutaneous flaps, including simple rotation, transposition, bipedicle and Limberg flaps are simple and provide pliable coverage with reduced morbidity, but they are limited by the length/width ratio and the reduced mobility necessitating extensive undermining and dissection to deliver further flap mobility that may potentially compromise vascularity and increase the risk of flap necrosis [20]. Gluteus maximus or latissimus dorsi myocutaneous flaps provide robust coverage. Nevertheless, they have long operating time and involve significant donor site morbidity, which are less desirable in neonates [21]. Transposed muscle flaps like using latissimus dorsi with split skin graft, while effective, it is recommended to preserve this muscle in prospective paraplegic patient who will largely depend on it for ambulation [22]. With advancement in reconstructive surgery, perforator flaps namely dorsal intercostal artery perforator and superior gluteal artery perforator flaps for resurfacing the MMC defects, but the need for tedious small perforators skeletonization in neonates in such time-consuming and complex procedures is the main drawback [23].

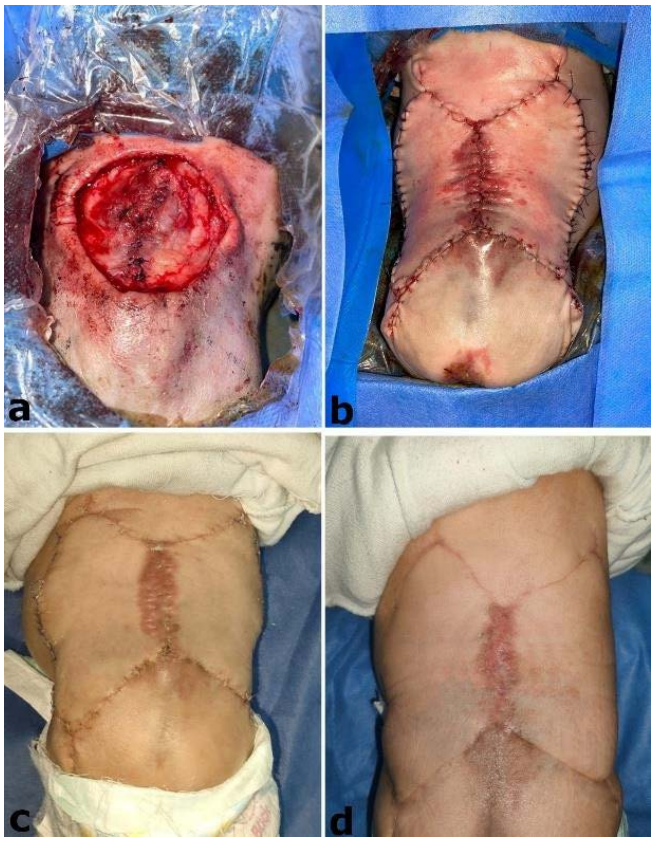

Figure 3. A 4-day-old female newborn with a lumbar myelomeningocele was reconstructed with type III keystone flap. (a) Intraoperative photo after dural repair. (b) Immediate intraoperative photo after reconstruction of the defect. (c) Patient at 8 days postoperative. (d) Patient at 6 months postoperative.

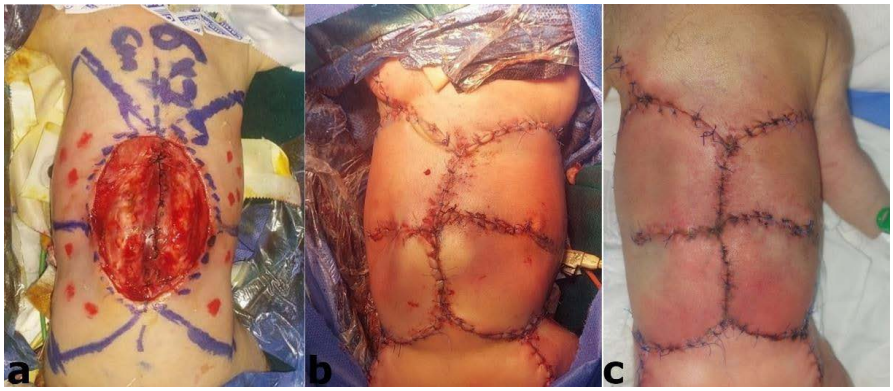

Figure 4. A 3-day-old female newborn with a thoracolumbar myelomeningocele was reconstructed with bilateral Omega modification of a type IV keystone flap. (a) Intraoperative photo after dural repair and marking of flaps. (b) Immediate intraoperative photo after reconstruction of the defect. (c) Patient at 6 days postoperative.

In the present study, we report a case series of 16 patients in which KPIF was used for covering MMC defects. The KPIF flap is a multiperforator island fasciocutaneous flap that was originally described and classified by Behan in 2003. Four main types have been reported in the literature: Type I (unilateral keystone flap, the deep fascia is left intact), Type IIA (as in type I with additional division of the deep fascia along the outer curvilinear line to facilitate tissue mobilization), Type IIB (with closure of the secondary defect using a skin graft), Type III (2 identical opposing flaps are utilized to create double keystone flaps), and Type IV (raised up to 50% of the flap subfascially and rotated with closure of the secondary defect using a skin graft) [3] [7]. Several modifications have been made to the original keystone flaps to effectively resurface soft-tissue defects of different sizes and shapes. These modifications include partial undermining, maintaining a skin bridge, and folding the double arms of the keystone flap on themselves, creating a horseshoe appearance "Omega variant keystone flap" [24].

In our series, we used type III keystone flaps in twelve patients, and type IV-Omega variant keystone flaps in four patients (3 unilateral and 1 bilateral). However, many studies [8] [12] [25] mainly used type III keystone flaps for closure of MMC defects. Hifny *et al.* [24] safely and effectively demonstrated the use of type IV-Omega variant keystone flaps to close MMC defects in seven patients. The decision of which type of keystone flap to use was based on the defect size, the laxity of the skin, and the quality of adjacent tissue (the infant's size). In the present series, we used unilateral keystone flap for defects ≤ 4.5 cm in width, and double keystone flaps for defects > 4.5 cm in width. The omega variant was particularly useful in patients where unilateral coverage was sufficient or for transversely oriented defects or those with limited lateral tissue laxity. In the study of Putri *et al.* [3], small defects (<15 cm^2) tended to be covered with type II keystone flaps, intermediate defects (15 - 30 cm^2) were resurfaced by Type III or unilateral type IV keystone flaps and larger defects (>30 cm^2) were mostly covered with bilateral type IV keystone flaps. This demonstrates the versatility of the keystone flap and high-

lights the adaptability of this technique to the anatomical and defect-specific requirements of MMC repair.

The optimal time for closure of MMC defects and reconstruction of the adjacent soft tissues is immediately after birth. In our study, the mean age at surgery was 4.4 days (range, 1 - 15 days). Similarly, Kushida and Gaxiola [25] operated between 1 and 8 days (mean 2.5 days). In another study [3], only 2 cases underwent surgery within the 1st 24 hours of life. However, 3 cases underwent surgery latter on (75, 113, and 369 days), which could be attributed to late referrals from 1ry and 2ry care hospitals that prevent timely surgery for soft tissue repair. Importantly, tissue laxity in older children may be diminished due to the presence of fibrosis and scarring due to previous infections; therefore, tissue mobilization is limited when compared to younger cases. So, when designing the keystone flap, a double keystone flap or a wider flap may be needed to resurface a relatively small defect.

Although we routinely use a handheld doppler to localize the location of the perforators. It is unnecessary to identify specific perforators within skin paddle of the flap using the handheld doppler. Many previous studies have demonstrated dual flap vascularization, which is reliable because of the subcutaneous vascular network and constant random musculocutaneous perforators of the latissimus dorsi, lumbosacral arteries, dorsal intercostal artery, and superior gluteal arteries perforators [8] [26]. Moreover, in the Omega variant keystone flap, the vascularity is maintained through several constant lumbar and intercostal musculocutaneous perforators, which attach to the central non-undermined part of the flap. This robust vascular supply provides better tissue bulk, and a wider geometrical versatility than the traditional random 1:1 flap. It has also been postulated that the blunt dissection used to raise the flap causes a local sympathectomy resulting in paradoxical vasodilatation, thus increasing blood supply to the flap [12].

In terms of operative time, ours lasted between 60 and 110 minutes (mean: 83.8 minutes), excluding the neurosurgical repair. In similar studies, Hifny and Hamadan [2], Putri *et al*. [3], Kushida and Gaxiola [25] reported average operative times of 85, 89.6, and 61 minutes, respectively. In another studies, Gutman *et al*. [12], and Park *et al*. [4] reported average operative times of 223, and 225 minutes (including the neurosurgical repair), respectively. Accordingly, our study showed comparable results to the previous series on MMC defect closure, indicating that KPIF is a time-efficient method compared with more complex reconstructive options. Importantly, no intraoperative or perioperative blood transfusions were required, further highlighting the low blood loss and minimal surgical trauma associated with the procedure.

A notable finding was the universal occurrence of immediate postoperative venous congestion and transient flap swelling, which resolved spontaneously within a few days without intervention. This phenomenon has been documented in previous reports [8] [12] and is thought to be related to the temporary impedance of venous outflow following flap mobilization. The spontaneous resolution of this

congestion and absence of progression to flap necrosis underscore the reliability of the flap's vascularity and confirm the robust perfusion of KPIF due to its rich subdermal and perforator-based vascular supply.

The low complication rate in this series is particularly encouraging. Only one patient (6.3%) had a minor wound dehiscence (<2 cm), which healed with conservative management. The absence of major complications such as flap necrosis, infection, or CSF leakage is significant. This low complication rate compares favorably with historical data on other reconstructive techniques, such as latissimus dorsi or gluteal muscle flaps, which have reported higher rates of wound breakdown or infection in similar patient populations. Moreover, this low complication rate is in accordance with many other studies [5] [7] [8] [14]. Gomez and Barrera [5], Mrad *et al*. [7], and Jamjoom *et al*. [14] demonstrated satisfactory results in using KPIF for MMC closure and reported no complications after defect closure using keystone flaps. Formentin *et al*. [8] noticed that 1 patient out of 7 had complications. The absence of late complications, such as CSF fistula or wound breakdown, after a mean follow-up of 10.6 months further confirms the durability of the keystone flap for long-term soft tissue coverage. Donaldson *et al*. [27] in their expanded case series for long-term follow-up of KPIF in reconstructed MMC defects found that the KPIF remains a robust, durable and effective alternative for closure of large MMC defects. Bas and Goker [28] noticed that in two patients, minimal wound dehiscence was detected in the distal part of the flap. No wound infections, hematomas, donor site complications, or seromas were observed in any patient. Leach *et al*. [29] in a systematic review of the literature to evaluate all reconstructions for MMC, found that the perforator flaps demonstrated the lowest rate of major wound complications (8.1%).

In the present study, the cosmetic outcomes were also favorable, with 87.5% of cases achieving thin, narrow scars and most parents were satisfied with the aesthetic results. The two patients with wide scars (12.5%) required fractional CO_2 laser treatment, which is a relatively minor intervention to improve scar appearance. Similarly, Gutman *et al*. [12] noticed that the aesthetic results were excellent in all cases, and they recommended the use of KPIF closure as an alternative for larger and more complex MMC defects. Park *et al*. [4] demonstrated that all keystone flaps were sensate above the level of spinal lesion with non-tender, cosmetically acceptable scar and preservation of the natural curvature of the lumbosacral spine. In a recent study, Kelly and Leong [1] assessed the long-term follow-up of KPIF in MMC defects closure and demonstrated that the use of KPIF is considered a robust, non-itchy, non-painful, and aesthetically acceptable surgical technique that can facilitate the closure of MMC defects. The high satisfaction rate among parents suggests that the keystone flap not only provides functional coverage but also meets aesthetic expectations, an important consideration in pediatric reconstructive surgery where long-term psychosocial impact is a concern.

Limitations of this study include the relatively small sample size (16 patients), which may limit the generalizability of the findings. Additionally, the follow-up

period, while adequate to assess early and intermediate outcomes, may not capture complications that could arise over a longer term, such as scar contracture or issues related to growth in pediatric patients. Future larger-scale, and multicenter studies with extended follow-up are needed to further validate these findings and to refine patient selection and flap design strategies.

5. Conclusion

The keystone flap is simple, reliable, versatile, and cost-effective treatment modality. It provides robust reconstructive option that is sensate and aesthetically acceptable and follows the principle of replacing "like with like" with negligible donor site morbidity. These results support its consideration as the first option for MMC defect closure, offering advantages over traditional methods by improving patient outcomes, and minimizing complications.

Conflicts of Interest

There is no conflict of interest.

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