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Repair of spina bifida cystica: an institutional experience

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Abstract

Objective Although most cases with spina bifida cystica could be closed by primary skin closure with or without undermining its edges, about 25% of patients have large defects not amenable for closure by these simple methods. We conducted this study to review our techniques in closing spina bifida cystica defects.

Methods We retrospectively reviewed the data of consecutive 21 patients diagnosed with spina bifida cystica in our setting. According to the surface area of the defect, the approach was decided; primary closure for small defects (11 cases) and flap-based approach for large defects (10 patients).

Results The age of the included pediatric patients ranged between 3 and 75 days. For the primary closure cases, a vertical incision was done in seven cases. For the flap-based group, bilateral rotation transposition flap was done for circular defects (7 cases) while elliptical ones were repaired via bilateral V–Y flap. Complete skin healing was achieved after two weeks (range 12–18 days) in most cases. Complications were as follows; for the primary closure group, cerebrospinal fluid leakage (18.18%%), partial wound dehiscence (18.18%) and superficial surgical site infection (9.09%). In the flap-based group, CSF leakage (20%), superficial surgical site infection (10%), and distal flap necrosis (30%) were detected. Reoperation for wound complication was needed only in one case in the flap-based group.

Conclusions Flap-based procedures are recommended for patients with large spina bifida cystica defects because of less tissue dissection and low complication rates. Primary closure should be kept for small defects.

Keywords Spina bifida cystica, Large defects, Closure technique

Background

Spina bifida is a common congenital malformation affecting the spinal column. It occurs secondary to failure in the posterior fusion of lateral neural tube edges [1]. The incidence of spina bifida is not uncommon, as it is encountered in less than 1–7 per 1000 live births [2, 3]. Egypt is one of the countries with the highest rates, along with China, Ireland, Pakistan and India [4]. Multiple etiologies have been proposed for this pathology, including chromosomal disorders, environmental exposure to toxins, or folic acid deficiency [5].

As it is a grossly visible lesion, most cases with spina bifida cystica are noticed at birth. The neural plate appears as a raw, red, fleshy plaque seen through a defect in the vertebral column (spina bifida) and the skin. A protruding membranous sac containing meninges, cerebrospinal fluid (CSF), and nerve roots are under the dysplastic spinal cord, which protrudes through the defect. The exposed neural tissue may be flat or elevated by a CSF sac below [6, 7].

Spina bifida has three forms; spina bifida occulta (the mildest form), meningocele (the least common form),

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and meningomyelocele, which contains meninges and nerves and it is associated with the most severe complications [8]. Spina bifida with myeloschisis is the most severe form of meningomyelocele, as the involved region is represented by a flattened, plate-like mass of nervous tissue with no overlying membrane, making the baby more liable for infectious complications (meningitis) [9]

Early surgical intervention is crucial for such cases, as any delay is associated with an increased risk of meningitis and progressive neurological deficits. Most surgical interventions are based on two main concepts; neural tissue protection and reliable closure of the dura [10].

In most patients, the detected defects are usually small and amenable for primary skin closure. Nevertheless, about 25% of patients have large defects which cannot be closed with such simple methods [11–13]. Any forceful attempts to close the skin increase the risk of wound dehiscence, skin necrosis and subsequent infection [14, 15].

These cases may require extensive subcutaneous dissection, skin grafts, local skin flaps, or myocutaneous flaps [16]. In fact, reconstruction of these large defects is still a challenging problem for both neurosurgeons and plastic surgeons [16], as the amount of neonatal tissue available for reconstruction is limited compared to the adult [1].

As there is no general management guidelines or global consensus on how to manage these lesions, we report our experience regarding the management of paediatrics diagnosed with spina bifida cystica in a tertiary care setting (Figs. 1, 2, 3).

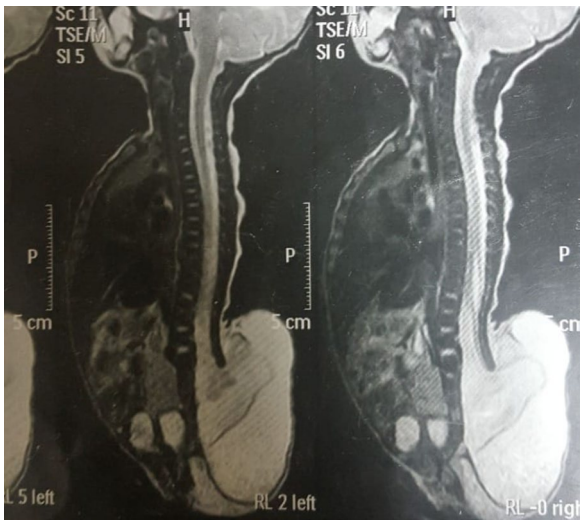

Fig. 1 MRI whole spine (sagittal T2) showing large myelomeningocele

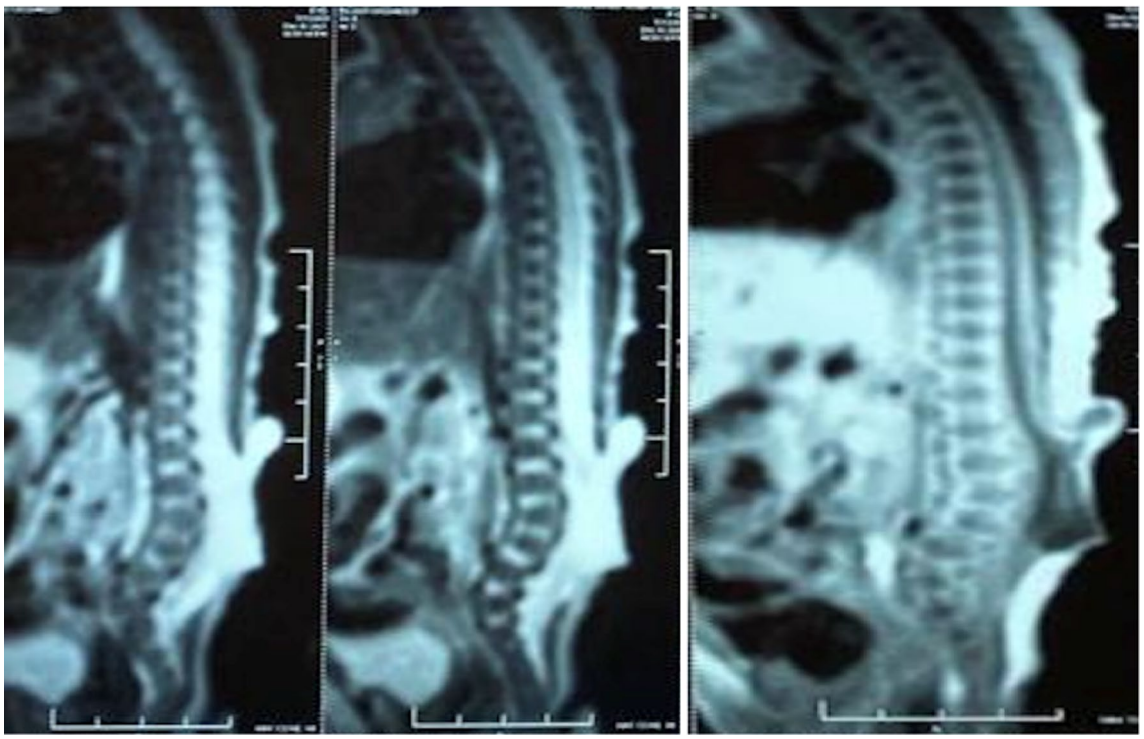

Fig. 2 MRI whole spine (sagittal view) showing myelomeningocele

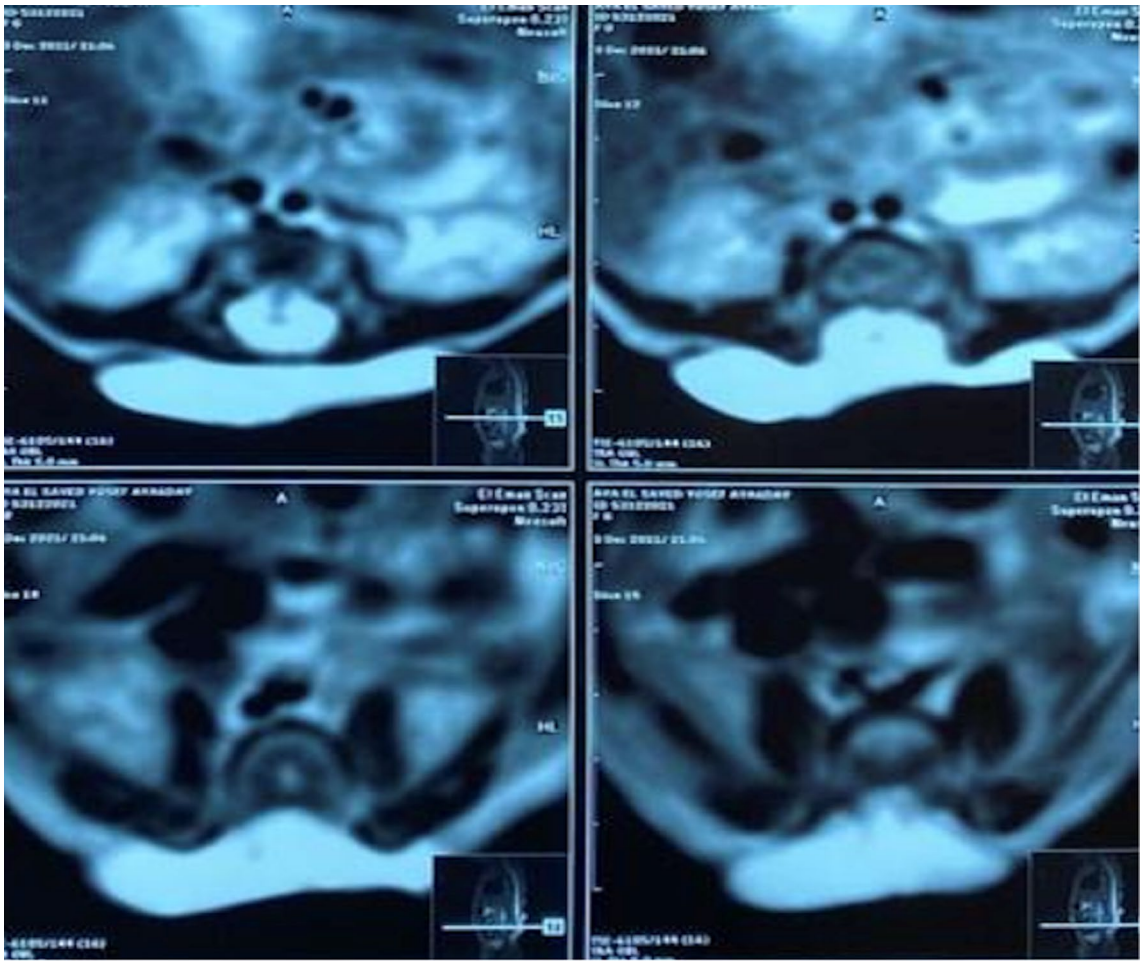

Fig. 3 MRI axial view showing myelomeningocele

Patients and methods

This retrospective study was conducted at Mansoura University Pediatric Neurosurgery Unit after obtaining approval from the local ethical committee and Institutional Review Board of our medical school. We retrospectively reviewed the data of spina bifida cystica patients managed by surgery during the period between January 2020 till January 2021. During that period, 21 pediatric cases with spina bifida cystica were operated on, and 10 of them had large defects (47.62%). These 21 patients were included in the current study. The large defect was established when the maximum defect diameter was more than 50% in relation to the width of the child back, as reported by Nejat and his associates [17]. When measuring this diameter, it was started and ended with the initial area of healthy skin surrounding the lesion.

All patients were subjected to standard history taking, general and detailed neurological examination. In addition, routine laboratory and radiological investigations (including magnetic resonance imaging of the lumbosacral spine and brain computed tomography) were ordered. After deciding on surgical intervention, it was completely explained to the patients' guardians with its indications and complications and following that, informed written consent was obtained.

All procedures were performed under general anaesthesia when the baby was in the prone position. The surgical incision was started at the neural placode margin, which was separated from the overlying peripheral tissues, to be positioned back into the spinal canal. After that, we performed trimming of the overlying granulation tissue and skin. Electrocautery was used with caution to avoid neural tissue injury. A new neural tube was constructed via both arachnoid and pia maters, which were drawn to the midline, and sutured using Vicryl 5/0 sutures without spinal cord compression. After that, we separated the dura circumferentially from the deep facial layers, brought the two sides of the dura together in the midline, and closed them with an absorbable suture in a continuous manner.

When it came to skin closure, we aimed to create tension-free skin closure to decrease the incidence of skin necrosis or wound dehiscence, especially with these large defects. Generally, we used the hairy skin covering the defect for closure, and most skin edges were undermined to decrease wound tension. In cases with small defects, primary skin closure was performed (Fig. 4). If the central wound edges appeared pale or if we felt some tension on wound edges, a vertical incision parallel to the midline was performed in one or two flanks to decrease wound tension.

In cases with large defects, in which no healthy skin for adequate closure was present, we used the help of the plastic surgery team. They used fasciocutaneous flaps based on the shape of the defect. For circular defects, bilateral rotation-transposition flaps were used (Fig. 5); one flap was based on the midline superiorly, and the opposite flap was based on the inferior midline. For elliptical defects, bilateral V–Y advancement flaps were performed (Fig. 6), where the V-flap base was designed nearby the midline defect and their apex reaching the midaxillary lines. All flaps were elevated through the supramuscular plane, and we ensured preserving the perforators at their bases. Finally, closure was done in layers via Vicryl 3/0 sutures for the subcutaneous layers and prolene 4/0 thread for the skin. A drain was left under the flap and was removed within five days following surgery.

Concomitant ventriculoperitoneal shunt (VPS) was performed in patients with significant hydrocephalus (increased head circumference and ventriculomegaly), while patients with normal or mild ventriculomegaly were followed by serial computed tomography, and subsequent VPS was scheduled if the patient developed latent hydrocephalus.

All patients received standard post-operative care, with frequent assessment throughout the day. Any post-operative complications were noted and then recorded. Regular follow up visits were scheduled for these cases. Patients with hydrocephalus who had no surgery were closely monitored at follow up if they developed any hydrocephalic manifestations requiring surgery.

Statistical analysis

Data entered and analyzed using Microsoft Excel software. Data were then imported into Statistical Package for the Social Sciences (SPSS 27, IBM/SPSS Inc., Chicago, IL) software for analysis. Baseline characteristics of the study population were presented as frequencies and percentages (%) or mean values and standard deviations (SD) or median and range (after testing of normality by Kolmogorov–Smirnov and Shapiro–Wilk's tests).

Results

The age of the included pediatric patients ranged between 3 and 75 days (median = 34 days). We included a total of 12 boys in addition to nine girls. These spina bifida cystica cases were diagnosed as follows; meningomyelocele (76.19%) and myeloschisis (23.81%). The age of the patients diagnosed with former lesions ranged between six and 75 days, while patients with the latter had an

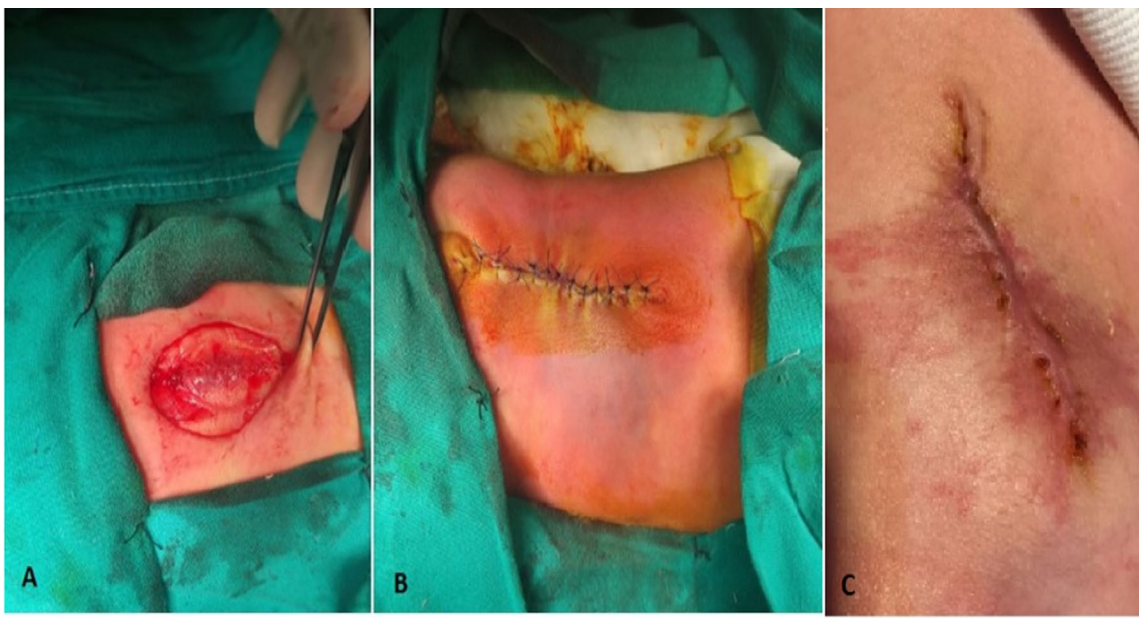

Fig. 4 **A** Meningomyelocele 3.5 × 4.5 cm. **B** After primary repair. **C** After stitches removal

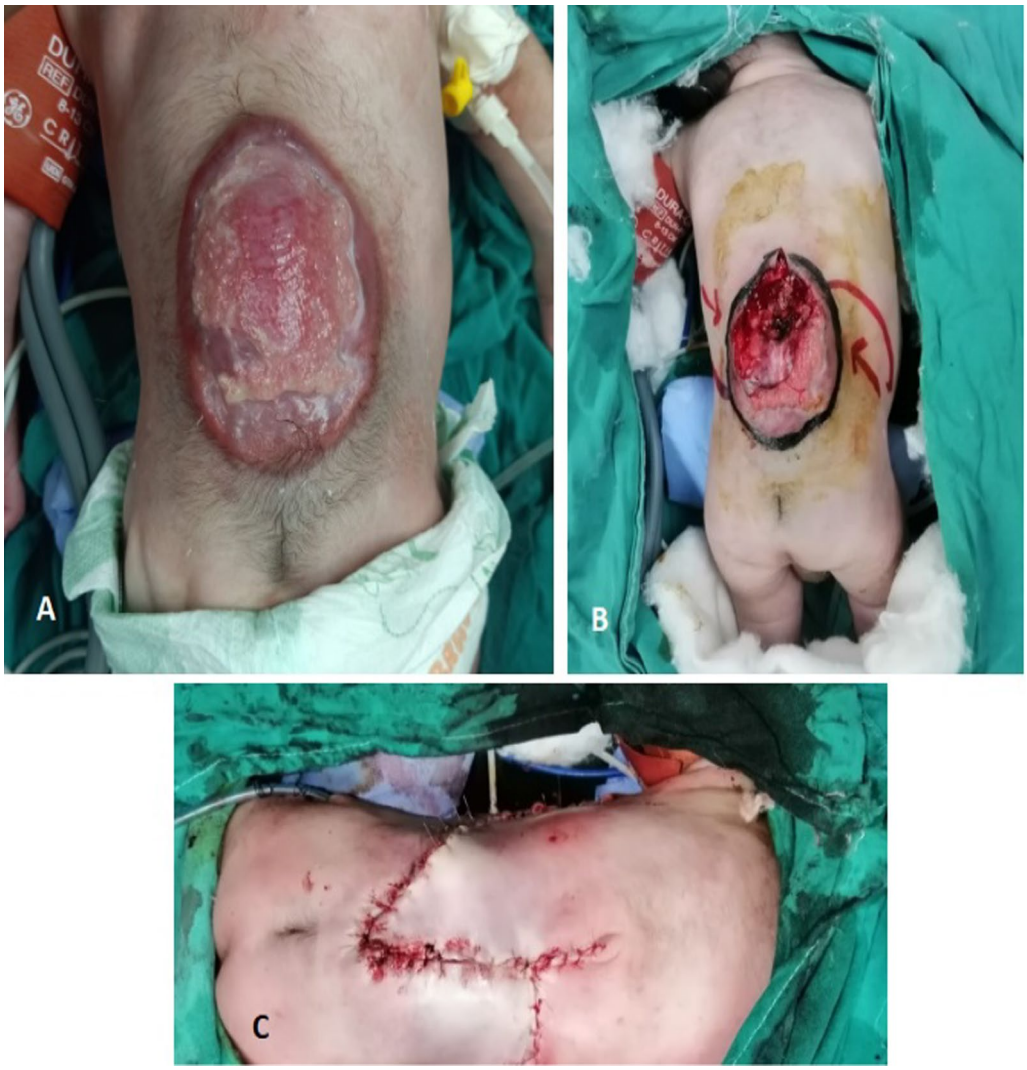

Fig. 5 **A** Large myelomenineocele. **B** Skin defect after dura repair. **C** Double rotational fascio cutaneous flap

age ranging between three and 72 days. Thoracolumbar lesions (61.9%) were more common than lumbosacral ones (38.1%). Clinical assessment revealed the following findings; bladder sphincter dysfunction (95.24%), lower limb weakness (71.19%), foot deformity (47.62%) and kyphotic spine (9.52%). Associated significant hydrocephalus was detected in only two cases (9.52%), while the remaining cases had normal or mild dilated ventricles with normal head circumference. The former two cases were managed by concomitant VPS during the same setting. The previous data are summarized in Table 1.

According to defect size, the management plan was decided (Table 2). For the eleven cases with small defect, primary closure was performed, and a vertical incision was performed in seven of them (63.64%). For large defects, seven patients (70%) with circular defects were managed by rotation or transposition flaps, while the remaining three patients with elliptical defects were managed by bilateral V–Y flaps (30%).

In most cases, complete skin healing (defined as 100% epithelization [18]) was achieved after two weeks (range 12–18 days), apart from two cases. The first one was in the primary group who reached complete healing after 23 days, whereas the second one was in the flap group. That case had a kyphotic deformity, and it took about three months for complete wound healing.

For the 19 cases who had normal or mild dilated ventricles prior to defect closure, 11 of them (57.89%) developed latent significant hydrocephalus which was managed by VPS.

Regarding the encountered complications (Table 2); for the eleven cases with small defects, complications were as follows; cerebrospinal fluid leakage (2 cases—18.18%), partial wound dehiscence (two cases—18.18%) and

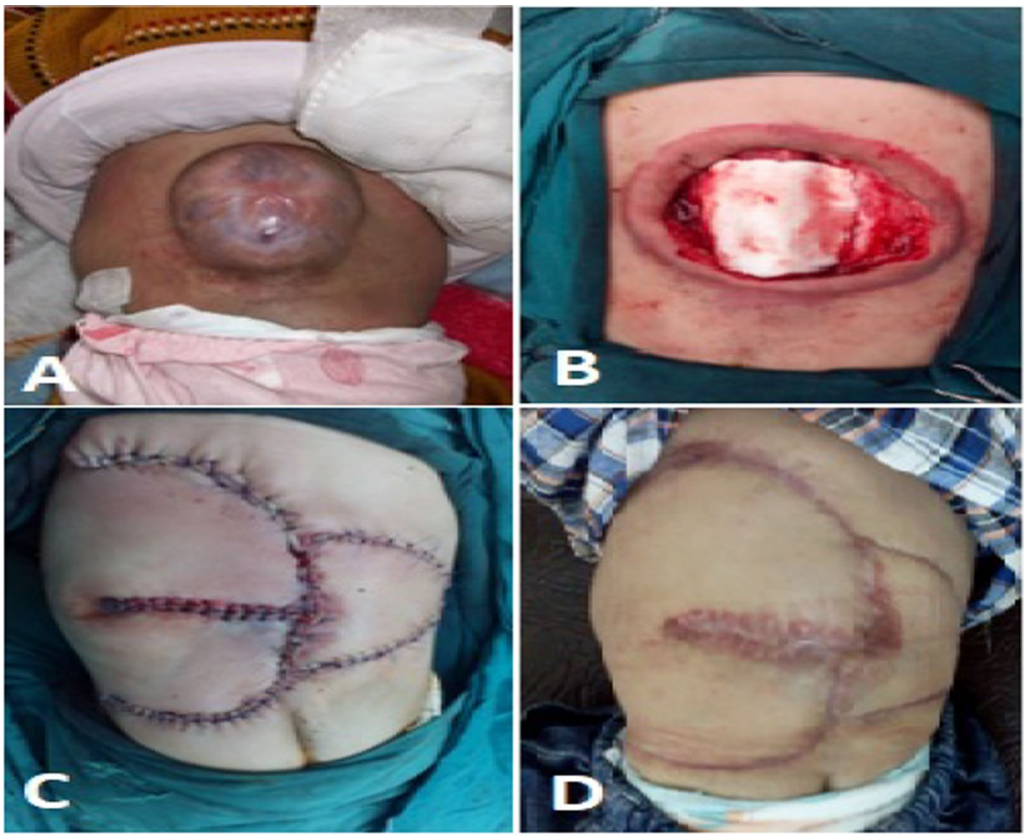

Fig. 6 **A** Large meningomyelocele. **B** Defect 9 × 7 cm after dural repair. **C** Reconstruction by bilateral V–Y flap. **D** Follow up picture after wound healing

superficial surgical site infection (one case—9.09%). CSF leakage was conservatively managed in two cases, while the remaining patient required a VPS. Wound dehiscence was managed conservatively in one case, while the other one was managed by vertical release incision and resuturing.

In patients with large defects, CSF leakage was encountered in two cases (20%) who required VPS insertion for managing this complication. One patient developed superficial surgical site infection (10%), while three cases had distal flap necrosis (30%); two of them were managed via frequent dressing, while the third one had kyphosis with necrosis of about half of the flap area. Thus, he required debridement and installation of a skin substitute (Integra) to cover the exposed dural repair, followed by a skin graft (Fig. 7).

Discussion

The current study was conducted to report our experience regarding the management of spina bifida cystica in our tertiary care setting. The ideal soft tissue reconstructive approach for large spina bifida defects would allow for a primary, one-stage cover of the restored neural tissue with viable, vascularized soft tissue while maintaining little or no wound tension [19, 20]. Two crucial factors should affect the surgical approach of these cases, including both defect size and location [10, 21].

In our study, meningomyelocele was the commonest form of spina bifida in the included cases (76.19%), followed by myeloschisis (23.81%). This agrees with a previously published review, which reported that meningomyelocele is the most common form of spina bifida [22].

Our findings showed that the thoracolumbar area was the most commonly affected region by the studies pathology (61.9%), whereas the remaining cases had lumbosacral defects. This also agrees with Nejat et al., who reported that the thoracolumbar area was affected by spina bifida cystica in 27 out of the included 40 cases with large defects (67.5%). Other sites included the lumbar, lumbosacral and sacral areas [17].

Bladder dysfunction was present in the majority of our cases (95.23%), and that coincides with multiple reports reporting that nearly all patients with similar neural defects have bladder dysfunction [23, 24].

Table 1 Demographic, clinical and operative data of the study cases

Variable	Data ($n=21$)
Age (days)	34 (3–75)
Gender	
Boy	12 (57.14%)
Girl	9 (42.86%)
Classification	
Meningomyelocele	16 (76.19%)
Myelocele	5 (23.81%)
Location	
Thoracolumbar	13 (61.9%)
Lumbosacral	8 (38.1%)
Clinical examination findings	
Bladder sphincter dysfunction	20 (95.24%)
Lower limb weakness	16 (71.19%)
Foot deformity	10 (47.62%)
Kyphotic spine	2 (9.52%)
Associated hydrocephalus	
Significant	2 (9.52%)
Normal or mild dilated ventricles	19 (90.48%)
Defect size	
Small	11 (52.38%)
Large	10 (47.62%)

In the current study, concomitant significant hydrocephalus was encountered in 9.52% of the included cases, while the remaining cases had normal or mild dilated ventricles. This is confirmed by most of many studies in the literature, which stated that hydrocephalus is present in about 75–90% of patients with congenital spinal malformations, and that item is unrelated to the surgery correcting the existing spinal defect [25, 26].

In our unit, we prefer to operate on the spina bifida cystica defect prior to the shunting procedure, especially when the defect is so large with difficult supine positioning. Also, we keep strict and adequate follow up for such cases, which allows us for early intervention via shunting or endoscopic methods, before CSF leakage or wound complications occur. That is why we performed only VPS for two patients (9.52%), who had clinical and radiological findings of increased intracranial pressure. Other 11 cases were managed in another setting after the spina bifida cystica correction due to latent significant hydrocephalus.

Our incidence of hydrocephalus agrees with a previous report which stated that about 10% of those babies will have apparent hydrocephalus at birth [27].

Nejat et al. reported that all of the forty patients included had hydrocephalus, from whom 30 cases underwent simultaneous shunting during spina bifida cystica repair. These authors reported a higher shunting rate during the primary procedure based on the fact that spina bifida cystica repair could increase CSF pressure. Therefore, it would be wise to perform the shunting procedure to decrease post-operative CSF leakage rates [17]. According to some researchers, this concomitant procedure is not associated with increased complication rates [28].

Herein, we decided the operative technique mainly based on defect surface area. If the maximum defect diameter was less than 50% in relation to the width of the child back, the wound was primarily closed. If larger defects were encountered, consultation of the plastic surgery team was performed to create the appropriate coverage flap technique. The current literature has variable criteria upon which the surgical decision was made. For example, Ozveren and his coworkers invented another approach for managing large spina bifida lesions. They classified the included patients according to the percentage of the defect to the affected body region (e.g. percentage of the defect to the thoracolumbar region). Defects less than 0.08 were managed by primary closure, while patients with higher ratios were managed by myocutaneous flaps [29].

Luce and Walsh reported the application of primary closure and split-thickness skin graft for defects with a mean surface area of 22.7 and 37.3 cm^2, respectively.

Table 2 Management and complication of the included cases according to defect size

Variable	Small defects ($n=11$)	Large defects ($n=10$)
Defect size (cm^2)	14.26 ± 2.68	25.34 ± 2.05
Management plan	Primary closure 11 (100%)	Rotation transposition flap 7 (70%)
	Vertical incision 7 (63.64%)	Bilateral V–Y flap 3 (30%)
CSF leakage	2 (18.18%)	2 (20%)
SSSI	1 (9.09%)	1 (10%)
Partial wound dehiscence	2 (18.18%)	0 (0%)
Flap necrosis	0 (0%)	3 (30%)

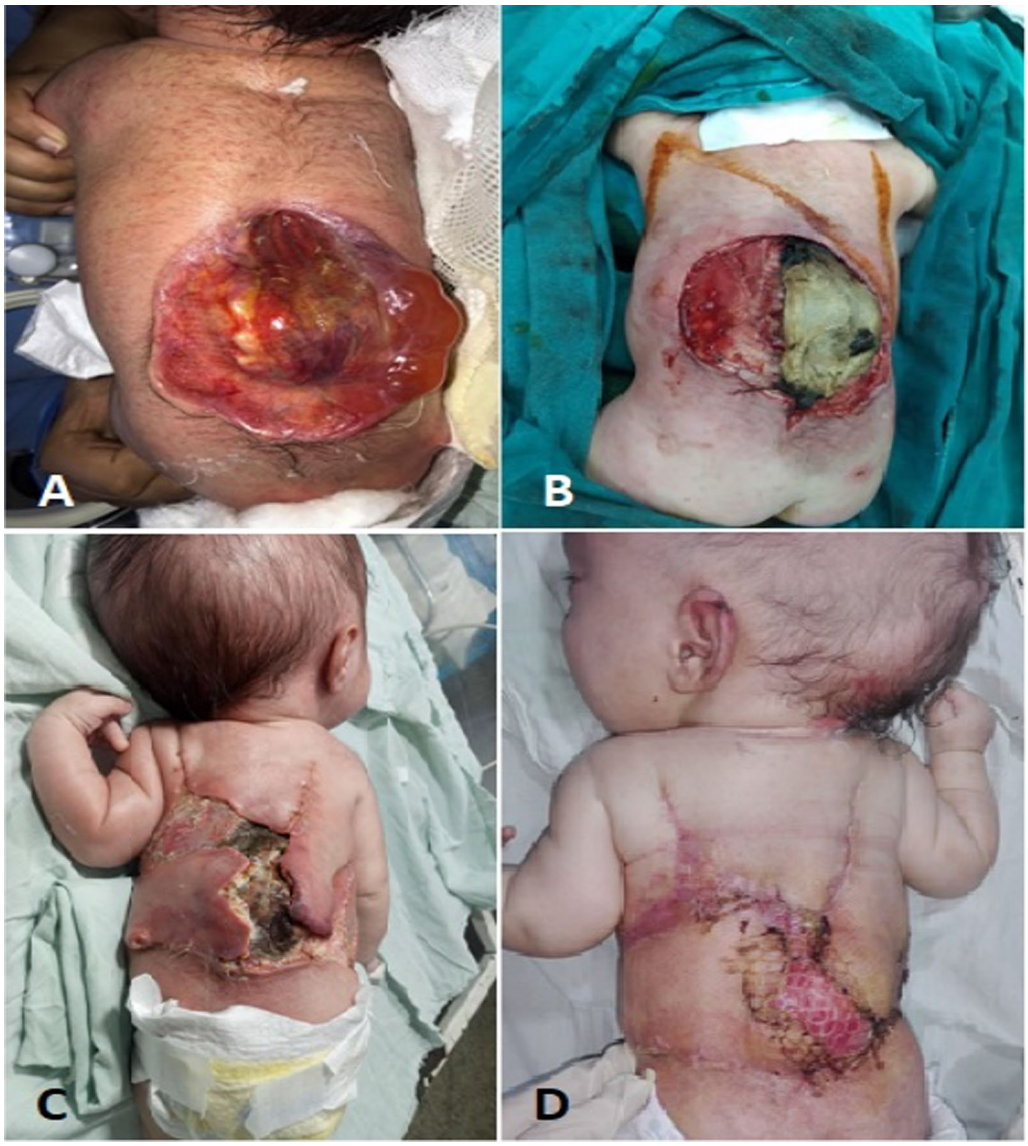

Fig. 7 **A** Large meningomyelocele. **B** Defect 10 × 15 cm after dural repair. **C** Debridement after flap necrosis. **D** Healing after split thickness skin graft

They noticed a significant rise in necrotic wound complications in association with primary closure ($p < 0.01$), which was encountered in 41% and 6.3% of the same two groups, respectively [29]. Of course, one could not exclude primary repair from the management options for large spina bifida cystica defects based on the previous results, as authors reported higher mean surface area compared to ours, which explains decreased complication rates reported in our study.

In a more recent study, Sharma et al. recommended direct primary repair for defects less than 7 cm^2 area, while they recommended flap coverage techniques for larger defects (>7 cm). The defect size had a mean value of 6.25 cm^2 for the primary cases, while it had mean values of 49, 36.5, 40, and 27 cm^2 for Limberg, double rotation, triple rotation and local transposition flaps, respectively. Only three cases developed wound complications in the previous study; one in the triple rotation flap group, and it was managed by surgical debridement followed by secondary sutures. The other two cases were in the Limberg and transposition groups; they had partial wound dehiscence, which was conservatively managed [1].

According to the previously mentioned methods, one could see that all authors used the primary closure technique for smaller lesions, while a more complex

procedure, including flap or graft, was used for extremely larger lesions. This is in line with our concept applied in the current study.

In our cases with small defects, we applied the same technique reported by Nejat et al., which utilized all hairy skin surrounding the defect for closure. Even if tension was encountered, a vertical incision was done in one or two flanks [17]. Although skin undermining is a good approach to enhance wound closure without tension, this would increase the risk of wound complications in paediatrics, as they have a restricted dermal circulation [17]. In these situations, where wound tension could affect wound healing, more complex approaches should be tried, including skin grafts, local flaps, musculocutaneous and fasciocutaneous flaps [1].

Mustardé was the first to suggest split-thickness skin grafts as a method of large defect closure [30]. With a low rate of problems, Luce et al. used delayed split-thickness skin grafting or simultaneous dural closure and skin grafting [31]. Later reports, on the other hand, noted a significant rate of graft ulceration due to gibbus deformity and severe kyphosis, necessitating further surgeries [13].

The application of flaps in managing large spina bifida lesions has been widely discussed in the literature. It was supposed that flaps originating from the paravertebral region provide adequate and reliable vascular support when used for constructing large spina bifida defects [32].

Musculocutaneous flaps have been also reported, including bilateral latissimus dorsi flap applied for closing large meningomyelocele defect [33]. In 1982, others used the same flap with extended gluteal fasciocutaneous flap for the closure of lumbar defect [34]. Other reported options included Limberg latissimus dorsi musculocutaneous flaps [35], distally based latissimus dorsi flaps [20], and reverse latissimus dorsi flaps [36]. Although musculocutaneous flaps have the advantages of high vascularity, better tissue coverage and more rapid healing, it has some disadvantages, including more operative time, blood loss and muscle sacrifice [10].

Others evaluated the application of perforator flaps for such lesions. However, more surgical expertise is needed, along with the need for specific ergonomics like microscopic instruments for such operations [37, 38]. Therefore, their application is more limited. In 2012, Patel and his associates reported closure of a large spina bifida defect using a local turnover fascial flap [39]. Other reported options included rotation flaps, V–Y advancement flaps, double Z-rhomboid flaps, bilobed flaps, and Limberg flaps [40–43]. Fasciocutaneous flaps have some advantages over myocutaneous flaps, including less operative time, less blood loss, and decreased functional consequences of muscle damage [19].

In the current study, complications were as follows for the primary closure group; cerebrospinal fluid leakage (18.18%), partial wound dehiscence (18.18%) and superficial surgical site infection (9.09%). In the flap-based group, CSF leakage was encountered in two cases (20%). One patient developed superficial surgical site infection (10%), while three cases had distal flap necrosis (30%).

Our CSF leakage rate lies within the reported ranges in the literature after such operations which is up to 30% [44, 45].

Another study reported that partial wound dehiscence was encountered in three out of the 20 cases subjected to primary closure (15%), and they were all managed by frequent dressings and left to heal with secondary intention. In the twelve cases managed by flaps, suture detachment occurred only in one case (8.33%), while another one died of sepsis secondary to CSF leak and secondary infection by faecal E. coli (8.33%) [29].

In another study evaluating rotation-transposition flaps for the same pathology, authors did notice any wound complications in patients undergoing a single flap procedure, while one case out of the eight undergoing double flap procedure developed distal necrosis of the superior flap (12.5%) [10]. Moreover, Akan et al. reported the application of V–Y flaps in such cases, without any post-operative complications [46]. However, another study, which evaluated the same V–Y flap technique in 27 neonates, reported wound complications in seven cases (25.92%) as follows; flap necrosis (2 cases), fat necrosis (2 cases), hematoma, cerebrospinal fluid leak, and wound infection (one case for each) [47].

Kemaloğlu pointed to the importance of proper patient selection; they applied primary closure for smaller defects, while larger defects were managed by different types of flaps. Wound healing was uneventful in all cases, apart from 4 patients (out of 20) who had partial flap necrosis, which was conservatively managed [48]. Another study evaluated the use of faciocutaneous flaps in closing large spina bifida cystica defects, and complications were as follows; partial flap necrosis (6.3%), wound seroma (4.2%), meningitis (4.2%), CSF leakage (2%) [19].

It is accepted to find some differences regarding the incidence of these complications in the literature. This could be explained by different lesion characteristics, surgical expertise, and perioperative care.

In our study, reconstruction of extremely large defects in ten cases with Fasciocutaneous flap had only one case with early reoperation for wound complication (10%), in which there was severe kyphosis of the spine, and we supposed that it adds extra tension on the flap. Therefore, we managed with skin substitute dressing followed by a split-thickness skin graft. This patient will require ongoing and careful monitoring, as there is a risk of progression of the

kyphotic deformity with growth. Neonatal kyphectomy and posterior fixation should be a good treatment option for this patient in case of progressing kyphosis later on.

Selçuk et al. pointed to the importance of kyphosis severity on spina bifida cystica management, especially in patients with large defects. It is supposed that more kyphosis would increase tension on the wound. Therefore, these authors recommended double flaps for patients with marked kyphosis compared to only a single flap for patients without that problem [26].

Our study has some limitations; It is a single-centre study that included a relatively small sample size. It also lacks long-term follow up for the included patients. These drawbacks must be handled in the upcoming studies.

Conclusions

Based on the previous findings, although we included large spina bifida cystica defects, primary closure was associated with good results in small defects. Additionally, flap-based procedures yielded satisfactory results in large defects. Hence, the appropriate procedure should be chosen according to patient criteria and surgeon experience. More large studies including more cases should be conducted to establish a worldwide consensus when managing such cases.

Abbreviations

CSF	Cerebrospinal fluid
SD	Standard deviation
SPSS	Statistical Package for the Social Sciences
VPS	Ventriculoperitoneal shunt
SSSI	Superficial surgical site infection

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Author contributions

All authors contributed to the study conception and design, read and approved the final manuscript for submission and publication. Contribution to the study was organized according to: MB, AFK contributed to Conceptualization. AFK, HIB, AZ, MFE, AME, MB, AEZ, MMA, MMA contributed to Methodology. AME, MB, AEZ, MMA, MMA contributed to Formal analysis and investigation. AFK, HIB contributed to Writing—original draft preparation. AFK contributed to writing—review and editing. MB, HB, AZ, MMA contributed to Resources. AME, MB, AEZ, MMA, AFK, HIB, AZ contributed to Supervision. All authors read and approved the final manuscript.

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Availability of data and materials

All data related for this study are available for sharing upon request.

Declarations

Ethics approval and consent to participate

This project was approved by the IRB of Mansoura University Faculty of Medicine. This article does not contain any studies with human participants performed by any of the authors.

Consent for publications

All data and records of patients were approved for publications by authors and involved patients.

Competing interests

All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

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