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KYPHOSIS CORRECTION AS AN OPTION FOR SURGICAL TREATMENT OF MENINGOMYELOCELE IN A NEWBORN: IMMEDIATE RESULTS OF TWO CLINICAL CASES AND LITERATURE REVIEW

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Objective. To analyze immediate results of meningomyelocele closure with simultaneous kyphectomy in newborns.
Material and Methods. In two newborns, correction of kyphosis by vertebrectomy and decancellation of the apical vertebral body was performed simultaneously during surgery for meningomyelocele
Results. In both cases, extensive mobilization of soft tissues to close the skin defect was not required, while a significant correction of kyphotic deformity was noted. The postoperative wound healed by primary intention on days 9—11. The follow-up period was 11 and 8 months. Only 34 such operations in newborns were reported in the literature. All the authors noted the absence of postoperative wound complications typical for meningomyelocele repair without an orthopedic stage. In small series with a long follow-up period there was a gradual loss of correction after such operations, but without the formation of angular kyphosis requiring repeated kyphectomy.
Conclusion. Kyphectomy in newborns with meningomyelocele provided the possibility of effective closure of the skin defect and uncomplicated healing of the postoperative wound.
Key Words: kyphectomy, meningomyelocele, kyphosis, newborn.
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Congenital kyphosis which accompanies meningomyelocele in 10–15 % of cases [1] significantly complicates surgical closure of the skin defect and is associated with a high risk of surgical wound breakdown [2], that requires long-term in-patient treatment. Meningomyelocele-related kyphosis is characterized by an extremely high potential for subsequent progression [3].

Primary kyphectomy in a newborn performed simultaneously with meningomyelocele closure not only creates much more favorable conditions for wound healing, but can also improve the orthopedic prognosis [1, 4]. This procedure, first performed by Sharrard [5] in 1966, has not been widely used to date: when analyzing the literature, we have found only 34 surgeries reported [1, 4–8]. This fact enables us to present our own observations.

The objective is to analyze technical options and short-term results of meningomyelocele closure with simultaneous correction of kyphosis in a newborn.

Clinical case 1

A newborn girl was admitted on the 1st day of life with the diagnosis: Chiari II malformation, cerebellar hypoplasia, thoracolumbar meningomyelocele. Complications were critical biventricular hydrocephalus, liquorrhea at the level of meningomyelocele, paraplegia. She also had omphalocele.

She was born at 39 weeks by C-section. Body weight was – 2,400 g, height – 42 cm, the Apgar score – 7/7. Spine X-ray showed mid-lower thoracic lordosis and thoracolumbar kyphosis of 67° with the apex at L3 (sagittal S-shaped deformity, Fig. 1a). Critical hydrocephalus and liquorrhea indicated urgent surgical treatment.

After 4-hour preoperative assessment, which included fluid resuscitation and laboratory tests, external ventricular drainage was inserted, and meningomyelocele closure was performed with L3 vertebrectomy.

Surgical technique. With the patient in the prone position, epithelioserous plate was dissected along the skin border. After mobilizing of the spinal cord and roots from adhesions, the cord was tubularized and the dural sac was formed. At the apical vertebra level, dura mater was freed from adhesions to the posterior longitudinal ligament. There were no posterior structures of the vertebrae in the defect area, the pedicles were about 5 mm in length and had lateral deviation. Pedicles of the apical vertebra were resected; the body of the L3 vertebra, adjacent discs, and endplates of adjacent vertebrae were removed subperiosteally through the pedicle base with a small curette. The correction maneuver was performed with finger pression on the apex of kyphosis. There was no significant resistance after vertebra resection. During kyphosis correction, the bodies of

adjacent vertebrae were sutured together. Sutures tended to cut through. Paraspinal muscles were sutured in midline without tension or need for significant mobilization. After that correction of the kyphosis appeared stable. The skin was sutured to complete closure, also without tension and extended mobilization.

Intraoperatively, the girl received transfusion. On the control X-ray at the age of 2 days, kyphosis decreased to 7° (Fig 1b). By the 9th day, the surgical wound healed by primary intention without any local complications. On the day 11, ventriculoperitoneal shunt was placed and simultaneous omphalocele repair was performed.

Child's follow-up was 11 months: there have been no local complications in the area of meningomyelocele repair; the kyphosis consisted 13° (Fig 1c).

Clinical case 2

A newborn girl was admitted on the 4th day of life with diagnosis: Chiari II malformation, lumbar meningomyelocele, corpus callosum dysgenesis. Complications were liquorrhea, paraplegia, subcompensated biventricular hydrocephalus.

She was born at 39 weeks by C-section. Body weight was – 3,280 g, height – 50 cm, the Apgar score – 7/8. Spine X-ray showed C-shaped kyphosis of 70° with the apex at L2 (Fig 2a). On the day of admission, after preoperative assessment, the child underwent meningomyelocele closure with simultaneous decancellation of the L2 vertebral body.

The technique of the neurosurgical stage of the surgery did not differ from that presented in the first case. After the removal of apical vertebra pedicles, decancellation of the body was performed. During finger correction of kyphosis, tension loops were put on the pedicles of the adjacent vertebrae for stabilization. The suturing of the paraspinal muscles in the midline and skin closure were also carried out without tension and significant mobilization.

The patient received a transfusion intraoperatively. On the postoperative X-ray, kyphosis was transformed into a local lordosis of 3° (Fig 2b). By the day 11, surgical wound healed by primary intention without local complications. On the day 6, external ventricular drainage was performed, then (on the day 24) ventriculoperitoneal shunt was placed. The total follow-up period of the child was 8 months.

Local kyphosis in the surgery area was 6° (Fig 2c).

Discussion

Meningomyelocele surgery in newborns usually consists in the reconstruction of the spinal cord, the formation of closed dural sac, and the closure of the soft tissue and skin defect [9]. Despite the variety of proposed plasty techniques that use cutaneous, facial, and muscle flaps [2, 9, 10], the rate of early local complications is as high as 26 % [9], and up to 12% of patients require re-surgery due to skin breakdown [10]. Kyphosis cases require the most aggressive surgical techniques and are associated with the highest complication rate [2].

Information about primary kyphectomy in newborns is quite scarce: we found only 6 reports that include 34 clinical cases (Table).

In most cases, vertebrectomy was performed. Only four patients underwent vertebral body decancellation [4]. The orthopedic stage was usually performed before the neurosurgical one [4–6, 8], while in the largest series [1] the dural sac was formed first. One patient had the vertebrotomy zone fixed with a cervical plate [8], while all other surgeries were performed by suturing the vertebral bodies [1, 4–6]. All authors note the absence of early wound-related complications [1, 4, 6, 8], and three out of the four fatal outcomes described were reported as not associated with kyphectomy [4, 6].

All the patients with a long follow-up period (one year or more) showed a gradual loss of correction [1, 4, 6] with development of a round-back paralytic kyphosis that did not affect the body support ability [1]. In one patient only, kyphosis area ulceration developed in the long-term period [1].

Both kyphectomies in newborns with meningomyelocele reported here were performed in 2021. The neurosurgical stage of the surgery preceded the orthopedic one (Crawford technique [1]). Closure of the dural sac after vertebrectomy is carried out under more favorable con-

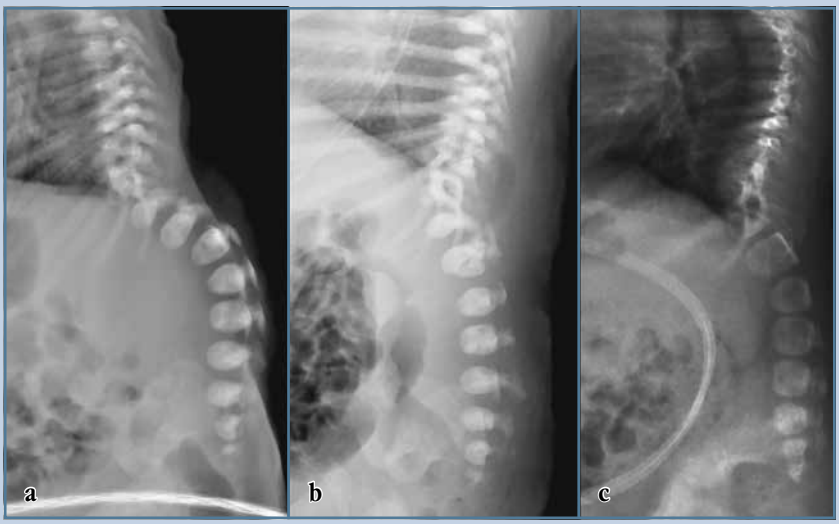

Fig 1
Lateral spine X-Ray of a newborn girl before surgery **(a)**, one day postoperatively at the age of two days **(b)**, after 11 months postoperatively **(c)**

ditions, but vertebral osteotomy with the «opened» spinal cord can be potentially more dangerous. The choice of the vertebrectomy technique in the first case was based on a more rigid S-shaped deformity. C-shaped kyphosis made it possible to limit osteotomy to decancellation of the apical vertebra. It is noteworthy that all authors who performed the vertebrectomy skeletonized the lateral and anterior surfaces of the vertebrae [1, 4, 5]. We were able to successfully perform transpedicular vertebrectomy using the egg-shell technique.

When suturing the vertebral bodies, there was tendency for the threads to cut through, while after medialization and erector muscles suturing over the posterior surface of vertebrae, the osteotomy zone was stabilized. We attribute the significant stabilizing effect of paravertebral muscles transposition and suturing to the unique biomechanical features of a newborn due to small size and body weight. A more reliable means to stabilize vertebrae was putting tension loops on the adjacent vertebrae pedicles.

Placing a plate on the posterior surface of the vertebral bodies in newborns [8] raises concerns due to its possible conflict with the spinal cord, as well as potential technical difficulties in case of the plate removal.

Unfortunately, we do not have own long-term results of primary kyphectomy in newborns. However, the obtained good short-term results of kyphosis correction without local complications are confirmed in the reports by other authors. This allows us to note the following advantages of the procedure discussed over the classic meningomyelocele closure:

1) the procedure does not require extensive mobilization of soft tissues for surgical closure of the defect;

2) in the early postoperative period, a wound heals by primary intention without trophic disorders, which is probably facilitated by the absence of local secondary changes and high restorative capacity of newborns;

3) development of less pronounced kyphosis in a long-term period compared to the natural course, is significantly more beneficial in terms of patient's life quality and possible kyphectomy and the use of endocorrectors at older age;

4) decreasing tension of the spinal cord at the kyphosis apex can theoretically have a favourable effect upon the neurological prognosis.

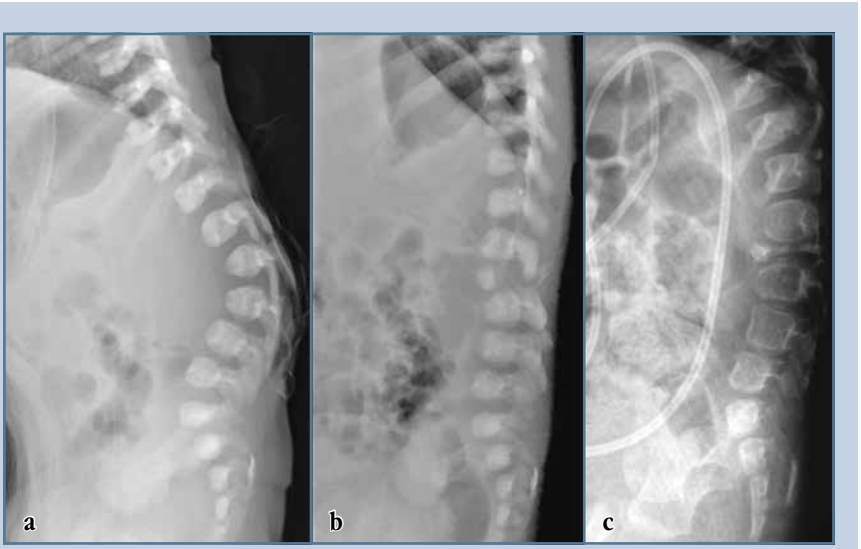

Fig 2
Lateral spine X-Ray images of a newborn girl on admission **(a)**, right after the surgery **(b)** and at the age of 8 months **(c)**

Typically, the natural history of meningomyelocele-associated kyphosis leads to inevitable progression of the deformity at the rate of up to 12° per year [3], caused by absence of stable posterior vertebral structures and the paradoxical effect of ventrally displaced erector muscles. Significant kyphosis leads to body imbalance and development of a stable pain syndrome [13], a decrease in the volume of the chest and abdominal cavities [14], iliocostal conflict [15], and inability to sit without support on arms [16]. Angular deformity complicates patient care, preventing the use of a wheelchair, while sitting on the sacrum provokes decubitus development [17]. Deficiency of soft tissues at the kyphosis apex, combined with their constant tension, results in ulceration and non-healing decubitus, which are easily infected and can lead to septic conditions [18].

Kyphosis significantly complicates the closure of skin defect during meningomyelocele repair in newborns [2, 11]. It is generally accepted to use local tissue plasty, while kyphosis correction is carried out at older age. This may be due not only to the fact that newborns do not come to the attention of spinal surgeons [1, 25], but also to the idea that such procedures are preferable in children over the age of three [1, 11]. In this age group, however, surgical treatment of meningomyelocele-associated kyphosis is carried out under extremely unfavorable local and systemic conditions. Secondary vertebrae changes complicate the interpretation of radiological data (CT), and therefore it is recommended to manufacture 3D spine models for surgery planning [12, 19, 20]. At the same time, kyphectomy with the removal of one to five vertebrae [21] and multilevel spine stabilization [22] is predictably accompanied by various and frequent complications: skin breakdown [14, 23] and fractures of hardware elements [23], development of pseudarthrosis [24], and dysfunction of the existing ventriculoperitoneal shunt [22]. A significant part of such patients are subsequently operated on repeatedly [14].

Table
Publications on kyphectomy in newborns

Source	Number of cases	Follow-up period	Repeated surgeries
Sharrard [5]	6	Up to 6 years	2
Sharrard, Drennan [6]	7	No data	No data
Eckstein, Vora [7]	1	No data	No data
Crawford et al. [1]	11	4–174 months	1 (at the age of 9 years)
Duddy et al. [8]	1	1 year	0
Özdemir et al. [4]	8	36–61 months	0

Conclusion

Undoubtedly, the system of prenatal screening will lead to a reduction in meningomyelocele incidence in newborns. Nevertheless, such cases will occur sporadically, and, consequently, the treatment of such children will remain quite difficult for both neurosurgeons and orthopedic vertebrologists.

Even a scarce but positive information about kyphectomy in newborns with meningomyelocele allows us to consider this option as a possible alternative to the standard treatment tactics. In our cases, kyphosis correction provided effective closure of the skin tissue defect and uncomplicated healing of the surgical wound, while literature reports a sufficiently favorable long-term prognosis.

Limitations of the study confidence: a small number of cases and a short follow-up period.

All authors made an equal contribution to the paper. The study had no sponsors. The authors declare that they have no conflict of interest.

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