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Surgical Treatment of Spine Deformations after Neonatal Sepsis (Analysis of Clinical Series)

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Abstract

Background. Neonatal sepsis presents one of the current issues in modern pediatrics. The orthopedic outcomes of such a state and the possibility of treatment, in particular by surgical spinal reconstruction, are rarely analyzed. ***The purpose*** — to analyze pathology features and treatment outcomes in infants with vertebral complications resulted from neonatal sepsis. ***Materials and Methods.*** The analysis of observation and treatment of 15 infants, who have undergone neonatal sepsis which led to vertebral lesion with subsequent gross kyphotic deformity formation, is presented. ***Results.*** Average age of infants was 2.5 months when spinal pathology was diagnosed. In 7 of the 15 observations, a local angular kyphosis was revealed when the acute phase of disease was already passed („cured"). The thoracic vertebrae were most often affected, mainly Th 7-8 vertebral bodies. Average kyphosis was 53°. All infants were operated on during the period from 2006 to 2017. Each had two-stage spinal reconstruction including the anterior spinal fusion using a titanium mesh cage filled with bone autografts, or an autogenous bone graft only. At the second stage, the instrumental correction and fixation of the spine with a multi-support laminar structure were performed. Average age of patients at the time of surgery was 14 months. Average value of kyphosis correction was 27°. Further correction and anterior spinal fusion were achieved when performing the incorporation of a titanium mesh cage with bone autografts. The histological and bacteriological examination of the surgical material did not reveal any signs of infection or inflammation. Correction of deformity and restoration of the supporting strength of anterior vertebral column as a result of surgery were achieved in all cases. Various complications in the early and late follow-up period were reported in a total of 7 cases. Repeated interventions were required in two patients: in one case in the early period (dislocation of the structure supporting hook) and in one case in the long-term period (graft resorption and kyphotic deformity relapse). ***Conclusion.*** One of the complications of neonatal sepsis is severe multilevel thoracic spondylitis, the outcome of which is the formation of severe kyphosis against the background of subtotal bone vertebral destruction. The principal possibility of radical spine reconstruction in infants with achievement of good anatomical and functional results is shown.

Keywords: neonatal sepsis, spondylitis in infants, kyphosis, spinal reconstruction.

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Background

Neonatal sepsis remains a live issue of today's pediatrics: among all infant mortality cases, it ranks 2nd or 3rd with a mortality rate running up to 50% [1, 2]. At this point, a number of risk factors for developing neonatal sepsis (prematurity, pregnancy with prolonged gestosis, complex obstetric and gynecological case history, intrauterine infections), as well as its clinical and laboratory diagnostic criteria, have been identified. Concurrently, attention has been focused on the fact that the bacteriologic verification of the pathogen did not exceed 45% [3, 4, 5, 6]. A significant place is given to the differentiation of early-onset and late-onset sepsis in newborns, which is based on the age at onset of the disease. Despite the fact that the 72-hour interval is considered generally accepted, in literature there are indications on a 48-hour and 7-day time-based delineation as well as the opinion that such a division doesn't influence essentially the choice of therapy [7–10].

The most frequent neonatal sepsis local manifestations include the following: pneumonia, enterocolitis, meningitis, inflammation of soft tissues and osteomyelitis [1, 3, 4, 10]. Descriptions of clinical cases reporting spinal lesions [11, 12, 13] are limited.The outcomes of such states and the possibilities of treatment, in particular surgical reconstruction of the spine, are rarely analyzed [14]. However, along with the optimization of treatment strategies and increased survival rate of the infants who had sepsis in the neonatal period, the relevance of the issue will likely increase.

Objective: Analysis of pathology features and treatment results ininfants with spinal lesions occurring of neonatal sepsis.

Materials and Methods

Study design: retrospective mono-center series of clinical cases. The data of 15 infants who had neonatal sepsis and developed gross deformity of the spine as a result of destructive lesion of the vertebral bodies were analyzed. The data for this study were selected on the basis of the following criteria:

– retrospection time — 2017 back to 2006;

– age of primary manifestationof an infectious somatic disease corresponding to neonatal sepsis i.e. up to 90 days after birth;

– presence of destruction of vertebral bodies and kyphotic deformity which was an indication for referral to the clinic for surgical treatment;

– exclusion of specific etiology (tuberculosis, including post-vaccination form) of spondylitis through morphological and bacteriological examination, including molecular genetic studies of material from the affected area.

During the study the following data were analyzed:

– data of pre-, peri- and postnatal case history and obstetric-gynecological risk factors;

– clinical symptoms of sepsis onset state, local manifestations of infectious-inflammatory process;

– clinical symptoms of vertebral lesions, including the neurological status features;

– radiological semiotics of spinal lesions in neonatal sepsis at various stages from the initial diagnosis until the formation of spinal fusion after surgical treatment;

– results of the bacteriological and histological surgical material examination.

Average term of observation of the patient was 2±0.2 years afterthe main intervention.

The small number of selected data and long duration of retrospective dataselection (12 years) did not suppose the possibility of a complete statistical analysis.

Results

The main clinical features of the studied cohort of patients are shown in the Table below.

Due to the lack of complete radiological follow-up of the patients No. 6, 11, 13, their data were not taken into account when analyzing the relevant indicators in the group.

Table

Main clinical features of the studied cohort of patients

No	Sex	Sepsis local manifestations	Age			Spinal location of lesions before intervention	Neurological status before intervention (Frankel type) after intervention	Angle of kyphosis			Anterior spinal fusion technique	Follow-up period
			when sepsis onset occured	when vertebral lesions started	when patient unde went spinal surgery			before surgery	after surgery	at the end of the follow-up period		
1	M	Pneumonia, otitis	1 m.	4 m.	14 m.	Th 7-10	D	72°	32°	32°	TMC+ AutoB	1 year
2	M	Pneumonia	10 days	1 m.	7 m.	Th 7-10	E	62°	25°	25°	AutoB	2.5 years
3	F	Pneumonia	20 days	2 m.	8 m.	Th 7-10	E	50°	39°	63°	AutoB	2 years
4	M	Chestwall abscess	14 days	1 m.	7 m.	Th 5-8	E	47°	34°	34°	AutoB	1.5 year
5	M	Suppuration of an operational wound, pneumonia	1 m.	7 m.	21 m.	C 5-7, Th 4-7, Th 12-L1	E	80°	32°	32°	TMC+ AutoB	3 years
6	M	Colitis	2 weeks	2.5 m.	11 m.	C 3-4	D	40°	–	–	AlloB	2 years
7	M	Pneumonia, coxitis	1 m.	1.5 m.	14 m.	Th 5-8	E	40°	14°	14°	TMC+ AutoB	2 years
8	M	Pneumonia, chest wall abscess, phlegmon of the hand	3 m.	4 m.	13 m.	Th 5-8	E	45°	25°	25°	TMC+ AutoB	3 years
9	F	Pneumonia	1.5 m.	5 m.	7 m.	Th 8-10	D	54°	40°	40°	TMC+ AutoB	4 years
10	M	Meningitis, enterocolitis	9 days	12 m.	35 m.	Th 3-8	E	37°	18°	34°	TMC+ AutoB	2 years
11	M	None	1 m.	1.5 m.	9 m.	Th 9	E	–	–	–	AutoB	1.5 year
12	M	Pneumonia	3 days	3 m.	13 m.	Th 5-8	E	65°	31°	31°	AutoB	3 years
13	M	Pneumonia	1 day	4 m.	8 m.	C 4-7, Th 9-11	E	57°	–	–	AutoB	1 year
14	M	Pneumonia	2 weeks	2 m.	8 m.	Th 2-5	E	64°	33°	33°	AutoB	1.5 year
15	M	None	1 m.	2 m.	12 m.	Th 5-7	E	52°	22°	22°	TMC+ AutoB	1 year

TMCs — titanium mesh cages); AutoB — bone autografts; AlloB — bone allografts; m. — months.

Anamnesis study showed the presence of obstetric-gynecological and perinatal risk factors in 14 of 15 children: prematurity, severe forms of gestosis, infection of the mother, IVF. In one child, a septic state developed as a complication of cardiac surgery that he underwent when he was one month old (see No.5 in the Table).

Two infants developed illness in the first 72 hours (early-onset neonatal sepsis); in 13 cases, the sepsis onset developed from 3 days to 3 months after birth (average 28 days), which corresponds to the late-onsetneonatal sepsis. In all cases, the onset of disease manifested itself by hyperthermic syndrome (temperature rise above 38°C) and symptoms of intoxication. In 10 cases (67%) pneumonia was diagnosed. Six children had coxitis, gonarthritis, otitis, chest wall abscess, meningitis, enterocolitis.

Spinal lesions have never manifested themselves in the onset of disease. They were often identified accidentally during radiological examinations (CT scan, MRI) which were performedfor diagnosing pneumonia or controlling its dynamics. In 7 of 15 cases, the spinal column pathology as a local deformity was detected by the parents when the acute phase of disease was already passed ("cured"). The minimum period from the sepsis clinical manifestation onset to the diagnosis of vertebral lesions was 21 days, with an average of 2.5 months. Note that in three patients the identified changes were initially interpreted as a tumor process. Thus children were examined for a long time, consultations given and in one case a biopsy was carried out.

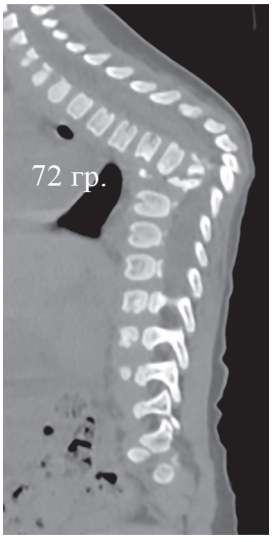

Fig. 1. Outcome of neonatal sepsis: severe kyphotic deformity — CT scan, sagittal view: subtotal destruction of Th7-Th10 vertebrae, kyphos 72°, spinal stenosis

During the active inflammatory process, all infants received near their homes an intensive medical, antibacterial therapy given to reverse the septic state. According to archival radiological data (CT scans and MRI), initially in all cases the scenario of pathology included destruction of vertebral bodies, edema of paravertebral tissues, often with an exudative component, regarded as "abscesses". Along with the soft-tissue component regression, phenomena of vertebral lesions increased up to complete destruction of the vertebrae causing a severe progressive kyphotic deformity (Fig.1).

The topography of vertebral lesions is specific: lesions of the thoracic vertebrae occurred in 14 cases, of cervical vertebrae in 3 patients, the most typical being the destruction of Th 7–8 vertebrae reported in 73% (Fig. 2).

Two-level lesions were reported in two infants, multiple (polysegmental) lesions of three or more vertebrae in 13 cases. A discrete lesion of the cervical spine was reported in one infant.

In all children, when admitted to the clinic, the symptoms appearedas a local, rigid, painless kyphotic deformity. According to imaging studies (X-ray films, CT scans, MRI), average kyphosis was 53° (min 37°; max 80°). Along with the destruction of vertebral bodies, the deformity was associated with dorsal migration of their fragments with stenosis of the spinal canal at the apex. However, only three patients had neurological disorders (Frankel type D). Soft tissue paravertebral changes were characterized by indolent edema and induration.

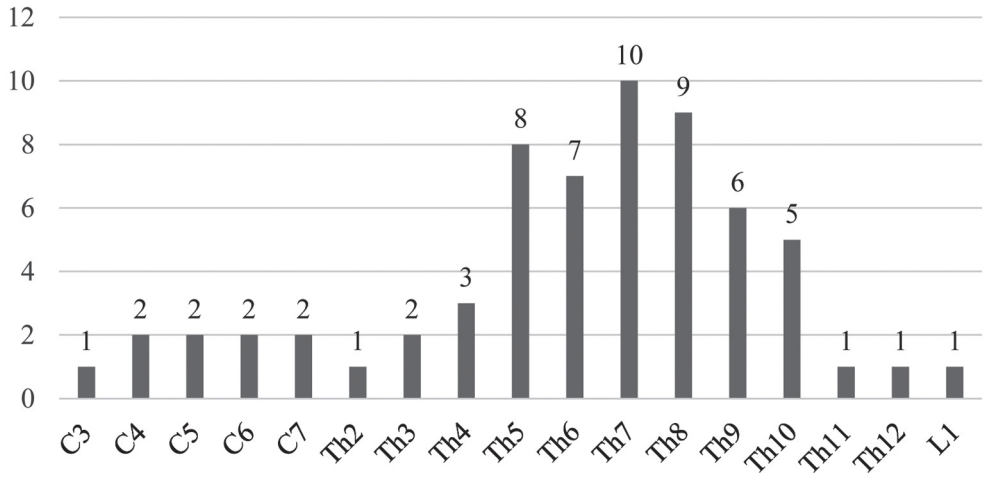

Fig. 2. Incidence of vertebral lesions in neonatal sepsis

Clinical and radiological data — progressive spinal deformity with a defect in the anterior column, in most cases involving two or more segments. This is interpreted, in terms of modern vertebrology, as a progressive kyphosis combined with destructive instabilityand failure of supporting strength of the spine. It is an absolute indication for surgical treatment, considering an unfavorable prognosis of the natural history of disease. All patients underwent reconstructive interventions aimed at restoring the spinal profile and its physiological function of support. The conditional restriction for reconstructive intervention was a baby's body weight exceeding 8 kg, which is due to the technical features of modern spinal surgery instruments used for young children. The patients' average age at the time of surgery was 14 months (min 7 months; max 3 years).

In 13 infants, the two-stage operation (anterior reconstruction and posterior instrumental correction and fixation) was performed with in one surgical session. In one infant, the treatment was separated into two operations (the anterior spinal fusion was carried out at first; the posterior instrumental fixation was performed 7 days later). This was due to severe kyphosis (80°), complexity of the first stage and hemodynamic instability. In aninfant with cervical spine destruction at the C2-5 vertebra level, the operation was limited to anterior reconstruction only (№. 6, Table).

When planning operations, we initially refused to perform a shortening vertebrotomy most commonly performed in the cases of kyphosis in adults (type VCR surgeries). It is associated with a significant shortening of the length of spine, which would negatively affect a growing child.

In 12 patients, access to the thoracic vertebral bodies was performed through right-sided thoracotomy with a rib resection. In two of them, the full extent of surgical intervention was performed from a posterior approach. In case of radical removal of pathological tissues represented by a conglomerate of bone, cartilage and scar tissue fragments, anterior decompression of the spinal canal was also performed. In no case the macroscopic signs of an active inflammatory process were detected. Post-resectional defect of the anterior column of the spine, with a length of two to five segments, was reconstructed under the circumstances of manual or temporary anterior instrumental reclination, by implanting rib fragments (7 patients) or titanium mesh cage (TMC) filled with boneautografts (8 patients). The application of TMC in infants started in the clinic in 2013.

In 14 children, the second stage was the installation of a posterior structure with laminar supports on both sides, with the formation of upper and lower "claw"-type constructs (Fig. 3). The additional correction of the deformity was carried out with the tension of the structure. The presence of residual kyphotic deformity, which was usually predicted at an initial kyphosis of more than 60°, was an indication for a monosegmental apical laminectomy. At the final stage, the posterior osteoplastic spinal fusion was performed using autograft rib fragments, which were placed onthe vertebral archesto the length corresponding to the anterior reconstruction of the spine.

In one infant with cervical spine deformity, reconstruction using an anterior-only approach ensured completing all tasks of intervention.

Average duration of the operation was 3 h 30±52 min, average volume of blood loss was 15.6±5.8% of the total circulating blood volume (TCBV). In 3 cases, despite the fact that the signs of myelopathy were present prior to the operation, the neurological symptoms fully regressed in the postoperative period. Average correction of kyphosis was 27°. In addition to that, when a titanium implant filled with bone autografts was used for anterior spinal fusion, the effectiveness was 7° higher than after interventions performed with the use of autograft ribs only.

In 7 patients (47%), various complications were reported, as follows:

– intraoperative bleeding from epidural vessels (one observation) which was treated with local hemostatic agents, hemotransfusion was performed;

– postoperative radicular syndrome (one case) was stopped by a non-operative method using a neurotropic therapy;

– complications of the early postoperative period — instability of the structure due to the dislocation of the supporting hook on the fourth day after the operation (1) and necrosis of the wound edges in the area of posterior access (1) were stopped by repositioning of the metal structure and necrectomy by stitching the wound, respectively.

In two of three cases of late-onset complications (occurred more than 1 year after surgery), a fracture (pseudarthrosis) of the anterior rib autograft was reported. In one

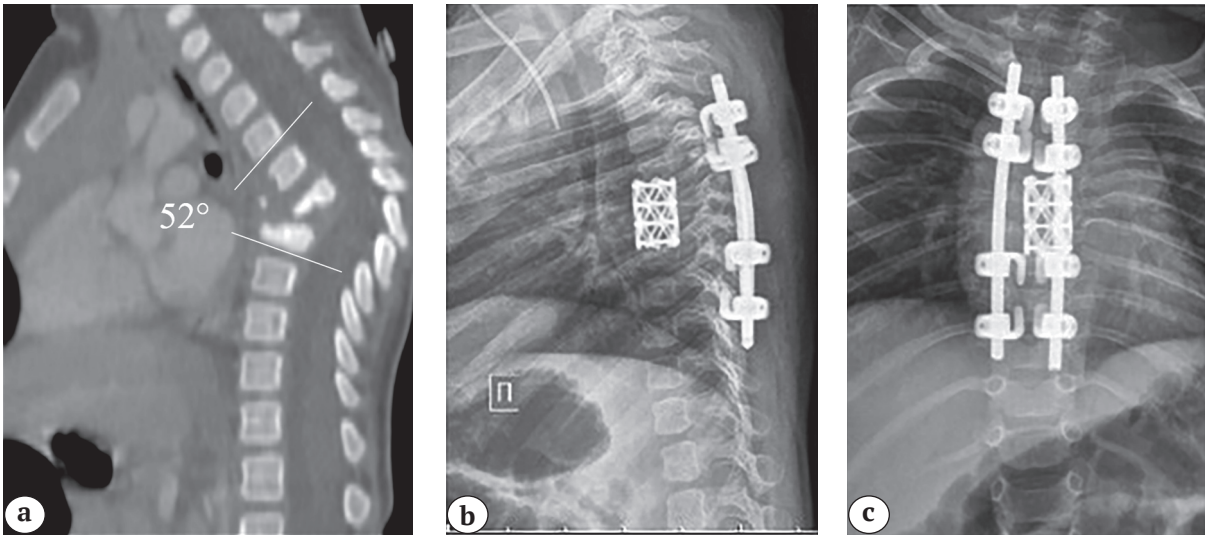

Fig. 3. Infant, 8 months old, the effects of neonatal sepsis with lesion of Th5-Th7 vertebrae:
a — CT scan, sagittal view, kyphosis is 52° combined with subtotal destruction of Th5-Th7 vertebrae;
b, c — postoperation X-rays

observation, the correction achieved during surgery was maintained by posterior stabilization and spinal fusion. In the second, there was a loss of correction by 25° (kyphosis was 63°) one year after removal of the posterior construction (2 years after the spinal reconstruction), which required repeated reconstructive intervention. In the third case, a patient underwent the anterior spinal fusion using a titanium mesh cage filled with bone autografts. After removal of the posterior construction, the loss of correction in this patient was 14°, while a total kyphosis was 34°, which does not exceed the physiological limit. This pediatric patient is followed up to the present.

In all other cases, the achieved correction is preserved. In 6 patients, the posterior metal structure has not been removed (the indication for its removal isthe anterior spinal fusion confirmed by a CT scan). These operations were performed in the last 2 years.

The histological and bacteriological study of the surgical material in all of the cases no signs of the infectious-inflammatory process activity were revealed. In addition, in all observations, there were no infectious complications recorded after surgery.

Conclusion

Severe multilevel spondylitis of the thoracic spine, the outcome of which is the formation of gross kyphosis with subtotal destruction of vertebral bodies, in our opinion, is a relatively rare but not casuistic complication of sepsis in the newborn. The analysis of case history and archival radiological data indicates quite typical characteristics of this process. The difficulties in interpreting existing semiotics, late and occasional diagnostics, incorrect treatment tactics are probably due to the rarity of pathology, as well as the scarcity of information on this problem not only in Russia but in foreign literature also.

A characteristic of the series of observations presented in the publication is a rather early age of interventions performed in the cases of a combination of gross kyphotic deformity and an extensive multilevel defect of the anterior spine. This can probably partially explain the rarity of publications on this topic. This pathology is found in pediatric vertebrology but usually in older children, particularly in specific spondylitis and tumor processes, and extremely rarely in vertebral anomalies.

The surgical tactics we used are based on the principles of modern pediatric orthopedics and traumatology — an early reconstruction of the defect with the restoration of the anatomy and function of the affected segment, taking into account the perspective of the child's growth and development.

The growth and development of the spine in children under the conditions of a multisegment anterior reconstruction necessary for treating of vertebral lesions, particularly in neonatal sepsis, is an unanswered question of modern vertebrology. A total understanding can be obtained only after 10-15 years, when the patients reach puberty. However, this aspect of the problem is still beyond the scope of this publication.

Spondylitis, as one of the neonatal sepsis, is usually diagnosed when an acute inflammatory process was already stopped. Typical radiolological manifestations of spondylitis — pronounced destruction, often polysegmental, paravertebral soft tissue component, angular kyphotic deformity — with good somatic condition of the infant allow refraining from emergency invasive diagnosis or treatment tactics. Exceptions are cases of pronounced neurological manifestations of spinal cord compression. Several of these patients were consulted by us in absentia. According to urgent indications, decompressive neurosurgical interventions were performed near their homes.

Post-destructive defect of the anterior column, causing multilevel instability of the spine, is an indication for surgery, the purpose of which is to correct the deformity and restore its supporting strength. The

extent of reconstruction as presented herein seems to us optimal because it allows avoiding the drawbacks of VCR* type shortening vertebrotomy.

The early age of pediatric patients does not preclude effective spinal reconstruction(anterior and posterior spinal fusion in combination with posterior instrumental correction and fixation). It is relatively safe due to the recession of the active process and gives the infantthe opportunity for developing fully.

To date, weight limits (body weight up to 8 kg) are not fundamental and are most likely due to the technical conformity of the spinal surgicalinstruments to the anatomical and functional state of the pediatric patient. Certainly, such operations belong to a high level of risk and should be performed in specialized clinical centers only.

The authors state that the article does not contain information that is prohibited to be published in public sources, has not been previously reported in its present form and is not being considered for publication in other periodicals.

The study partially includes data from a clinical series previously studied as part of a publication [14]. In the present study, the material is supplemented with new patients, advanced pathology analysis and modern literature.

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