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Case report

Surgical treatment of spondylitis and diaphragm relaxation in patient less than 1 year old

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ABSTRACT

Design: Case report.*Introduction:* The combination of severe post-infectious kyphosis and diaphragm relaxation is extremely rare in patient early than 1 year old. Its no publications concerning their simultaneous surgical treatment.*Case description:* 7-Month-old girl had simultaneous spinal reconstruction with anterior and posterior instrumentation and plastic of diaphragm because of sequelae of non-granulomatous spondylitis complicated by severe kyphosis (54°) and diaphragm relaxation. Between 1.5 and 3 months of live she had several infections incl. pneumonia, enterocolitis, ENT infection. Anterior fusion was done by titanium mesh with auto-rib, posterior – by compressive rods based on low-profile hooks. The deformity was reduced till 20°. 2.5 years after initial surgery and 1 year after removal of posterior instrumentation the adequate level of diaphragm and minimal (4°) loss of kyphosis correction were identified.*Conclusions:* The combination of spondylitis and diaphragm relaxation in early aged patient could be explained but it could not be confirmed as a sequelae of late-onset neonatal sepsis with a multi-focal lesions. The simultaneous surgery provided on the combined approaches (trans-thoracic and posterior) looks as optimal options in such combination of pathologies. In remains controversial how will the spine develop after so early reconstructive surgery, including in situ stable anterior fusion carried out by titanium mesh with auto-rib.

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1. Introduction

Infectious spondylitis in paediatric patients is extremely rare, and its study is usually limited to the clinical features

and treatment of spinal tuberculosis or non-specific spondylodiscitis.

Diaphragm relaxation is rare paediatric pathology also: a one-side high position of the top of the diaphragm with a normal low thoracic-aperture fixation line could be caused by

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either primary congenital abnormality (aplasia) or secondary developmental insufficiency of muscular elements (atrophy or dystrophy) of the diaphragm.

We observed a child with a combination of both forms of the pathological condition. Surgical treatment was carried out at an early age, and it involved simultaneous spinal and diaphragm correction.

2. Case description

The 7-month-old girl was admitted into our clinic in May 2013 for differential diagnosis of spinal pathology between tuberculosis (TB) spondylitis and congenital spinal abnormality.

It was the mother's second pregnancy and, while pregnant, she was diagnosed with anaemia and pyelonephritis. The patient's birth was by caesarean section because of the uterus' scar (the older sibling is healthy). The primary and secondary Apgar's scale were estimated 8/8, the weight being 3100 at birth. No infectious contact was found in the family and closed relatives.

The child was healthy up to 1.5 months with a physiological weight gain. The onset of disease associated with a high temperature (38 °C) and dyspnoea. She was hospitalised with a diagnosis of right-side pneumonia (the X-ray was not presented for revision), and treated by empiric regime of antibiotics (cefazolin, claforan and amoxiclav) with a positive clinical effect. On the X-ray control, the pneumonia was resolved, but suspicion over the high level of the right diaphragm was expressed.

One month later (at the age of 2.5 months) the temperature increased up to 39 °C associated with anxiety, dyspnoea, bilateral pneumonia, ENT (ear, nose and throat) infection and intestinal syndrome. *Staphylococcus aureus* was found in the stool. Antibacterial drugs including cephalosporin (3-rd generation), ampicillin and amoxiclav as well as anti-staph. bacteriophages. The treatment led to significant clinical improvement.

At the age of 4 months, during a stable period, the parents found the spinal deformity. After X-ray and computed tomography (CT) examinations the spinal pathology was assessed as a spondylitis. The Pott's disease (TB spondylitis) has also been proposed.

At the time of admission: the 7-month-old girl with normal motor activity; however, examination showed some lag in weight (7 kg). Her skin was moderately pale; peripheral lymphatic nodes were not increased; cordial tones, breath, bladder and bowel function were normal. The results of the clinical blood analyses, incl. leucocyte rate and erythrocyte sedimentation rate (ESR) are equal to the age. Immunological investigation did not reveal any compromise of the immune system.

Status localis: thoracic kyphosis with a prominent spinous process, pain-free palpation and no neurological deficit were observed.

X-ray (Fig. 1) and CT (Fig. 2) reveal the high level of the diaphragm top on the right side in comparison with the left side (the ribs 6 and 9, correspondingly) and destroyed vertebral bodies Th8–Th10, with a total absence of Th8 and Th9. The local kyphosis was 54°. The absence of a pronounced soft



Fig. 1 – X-ray of the thorax at the age of 7 months. The right top of the diaphragm is at rib 6; the left is at rib 9.

tissue component; the osseous inclusions in the epidural and paravertebral spaces (corresponding to the projection of the anterior and posterior ligament longitudinal) were detected.

The results of TST (tuberculin skin tests: RM 2TE and Diaskine test (Russian national analogue of the IGRA-test (interferon gamma release assay))) are negative. No ENT and neurologic pathology were revealed.

In the aggregate data, the diagnosis of TB spondylitis appears questionable. The pathology is regarded as transferred non-specific multi-focal infection with a vertebral lesion complicated by unstable kyphosis. Diaphragm relaxation could be caused by congenital abnormality as by infectious lesion (pneumonia). Because of the pure prognosis for spinal deformity progress and the risk of neurologic complication, the decision for surgery with a simultaneous correction of spinal and diaphragm pathology was made.

Two-stage one-narcosis surgery was performed on 20 May 2013: in the first stage, the position was on the left side. The right-side trans-thoracic access is via rib 7. The expressed adhesions between the visceral and parietal pleura were discovered and dissected. The diaphragm's tendon centre and sternal part appeared as a thinly stretched plate; the costal and lumbar portions were the preserved muscle structure. The right dome of the diaphragm was sutured in view of the blood supply mainly because of tendon centre duplication. The *lig. longitudinale ant.* had cut at the level of deformity. The expressed scar soft tissues changes were detected. The remaining fragments of the vertebral bodies and scar tissues were removed with anterior spinal decompression. The anterior fusion Th7–11 was achieved using a titanium mesh (inner diameter 10 mm) with an auto-rib inside it.

For the second stage, the patient was in the prone position. Th9 laminectomy accompanied by posterior Th5–L1 instrumentation was performed by low-profile multi-hooks construction.



Fig. 2 – Sagittal (a), axial (b) scans and 3D-CT (c) of thoracic spine at the age of 7 months. Destroyed Th8–Th9 body, subtotal Th10 (only the lower end plate is preserved). Partial ossifications in paravertebral tissues (b) and lig. *longitudinale posterior* (a). Kyphosis: 54°.

The general time of surgery was 3 h 20 min; the blood loss was 80 ml. The results of the surgery are presented in Fig. 3.

The post-operative period was without complications. The treatment included blood components' transfusion and antibiotics.

Results of cultural and molecular-genetic bacteriological studies of operative material were negative for *Mycobacterium* and non-specific bacteria. As for histology, signs of chronic inflammation were found.

The girl was examined at 6, 12 and 18 months after surgery: she had no complaints and was free in motor regime. Stable post-operative results of deformity correction with signs of bone fusion between the bone transplant inside the mesh and vertebral bodies (Fig. 4) were noted.

Due to the stable anterior fusion and potential risk of disc degeneration inside the instrumented zone in actively growing patients [1], 18 months after primary surgery the posterior constructions were removed. Clinical and radiological data 2.5 years after initial surgery (age – 3 years 2 months) resulted in a stable condition and no complaints. No mesh

instability occurred with a small scoliosis (5°) and loss of kyphosis' correction (from 20° until 24°, Fig. 5). The top of the diaphragm was at the same level on both sides. The patient continued to use hemi-soft thoracic-lumbar orthosis because of the mother's anxiety about non-limited activity.

3. Discussion

In our opinion, the presented case could be interesting for the following reasons:

The spondylitis had developed as evidence of multi-focal infection in a child in the first months of life. We reviewed the literature and found a publications dedicated to so-called “late-onset sepsis in early ages children”, concerning its aetiology, prevention, results of targeted and empiric antibiotic therapy, comparison with an early-onset sepsis [2–5] but only one article described vertebral lesion with severe post-infection kyphosis [6]. Unfortunately, during

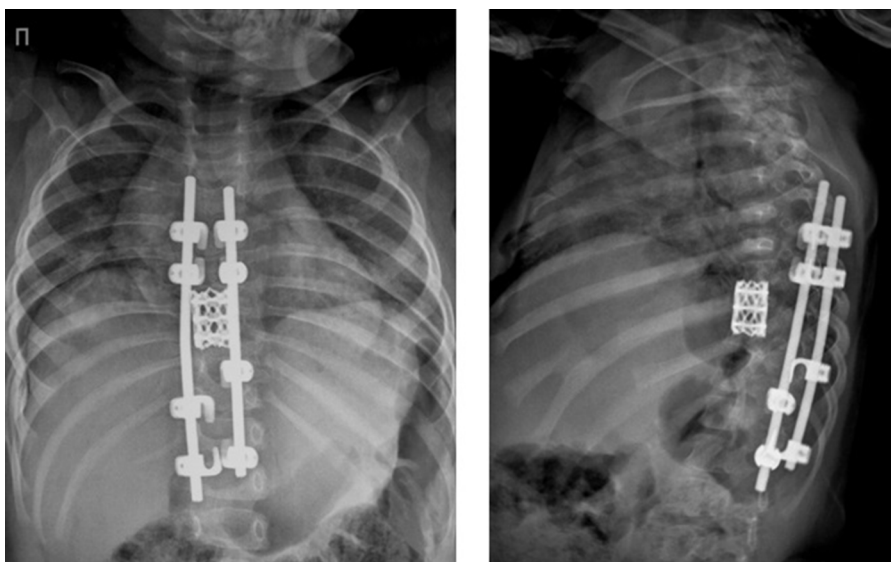


Fig. 3 – Post-operative X-rays (AP and oblique) just after surgery at the age of 8 month. The differences between the levels of the left and right tops of the diaphragm are less than one rib.

the active phase of the disease (between 1.5 and 3 months of life) before the admission to our hospital the complex bacterial and laboratory tests presently used for sepsis' diagnosis – blood culture, procalcitonin-test, C-reactive protein, neutrophil CD64 etc. [7] – were not performed. However, high temperature at the acute phase of the disease, the signs of multiple lesions (bilateral pneumonia, ENT infection and intestinal syndrome with *Staph. aureus*' isolates in the stool) and clinical effect of anti-bacterial treatment suggest that spondylitis discovered later was a consequence of suffering sepsis.

The diaphragm relaxation could be the result of congenital pathology as an effect of septic pneumonia and pleuritis, the multiple pleural adhesions confirmed the last version.

Perhaps, diaphragm biopsy could be informative to confirm the diagnosis, but it does not influence surgical decision-making.

The character of vertebral lesion was absolutely unfavourable for prognosis. Three interrelated components – spinal instability, progressive kyphosis and potential risk of neurologic complications – made surgery inevitable for patient.

Two approaches for a simultaneous plastic diaphragm and spinal reconstruction seem to be the optimal. In our opinion, the VCR (vertebral column resection) in this case could lead to the serious shortening of the spine and increase a risk of neurological complications as well as ruling out diaphragm surgery.

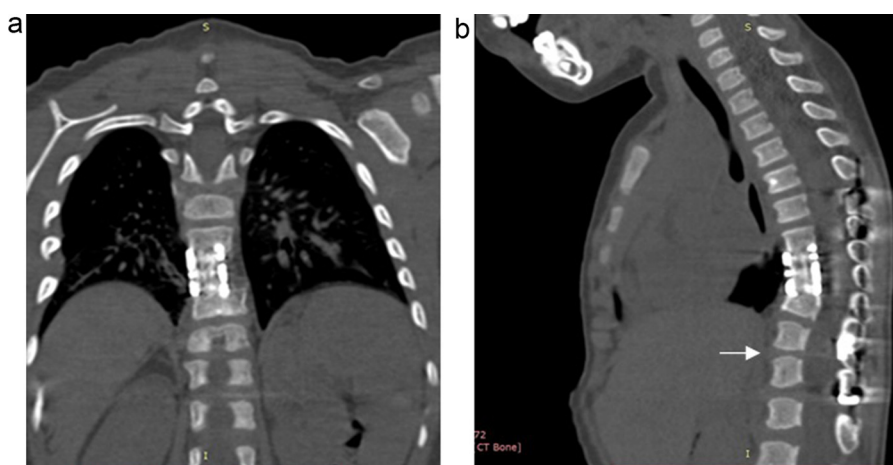


Fig. 4 – Coronal (a) and sagittal (b) CT spinal scans at the age of 2 years (1 year 4 months after surgery). The diaphragm tops are on the same level; bone transplant (auto-rib) inside the mesh completely grows with blocked vertebrae. The narrowed intervertebral disc Th12-L1 is inside the instrumentation zone (arrow, level of low hooks crew); stable anterior and posterior implant position, post-operative kyphosis is 20° (correction is 34°).

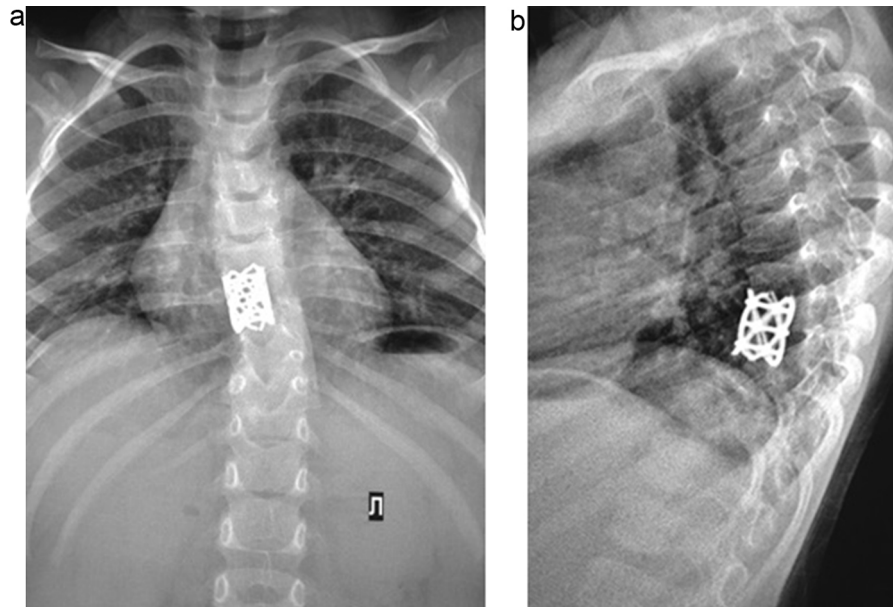


Fig. 5 – AP (a) and lateral (b) X-rays at the age of 3 years 2 months (2 years 6 months after first surgery and 1 year after removal of posterior instruments).

4. Conclusion

The case report illustrates the successful results of simultaneous surgical treatment for spondylitis complicated by severe kyphosis and diaphragm relaxation in 7-months-old patient. Despite this, it remains unclear how the spine will grow after such an early surgery. The known data concerning spinal growth after early age's operation in patients with severe tuberculosis (TB) kyphosis who underwent anterior fusion with bone graft and posterior instrumentation [1] could not be fully extrapolated to the combined (mesh + autograft) method. The appearance of minimal scoliosis deformity and kyphosis progression 2.5 years after surgery requires extending the observation.

Conflict of interest

None declared.

Acknowledgement and financial support

None declared.

Ethics

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving

humans; Uniform Requirements for manuscripts submitted to Biomedical journals.

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