THREE-COLUMN OSTEOTOMY OF THE SPINE DURING REVISION SURGERY IN A PATIENT WITH CONGENITAL ANGULAR THORACOLUMBAR KYPHOSCOLIOSIS

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A clinical case of surgical treatment of a female adolescent patient with multiple malformations, congenital thoracolumbar kyphoscoliosis, severe lower paraparesis and impaired functions of pelvic organs is presented with a review of the literature on the problem under consideration. During the course of treatment over several years, the patient underwent repeated revision surgical interventions because of implant instability and infectious complications. The last stage of treatment included a three-column osteotomy of the spine at the deformity apex with posterior instrumented fixation. A significant correction of the deformity was achieved. Based on the results of 18-month follow-up, the correction is maintained, the implant is stable. The patient reports a significant improvement in the quality of life.
Key Words: congenital kyphosis, three-column osteotomy, pediatric deformities, posterior fixation, revision intervention.

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Congenital kyphotic or kyphoscoliotic deformity is caused by vertebral developmental anomalies that prevent longitudinal growth of the anterior portion of the vertebral bodies. Isolated angular kyphosis is quite rare in children but is characterized by significant complications if untreated [23]. Most of these patients have mixed (kyphoscoliotic) deformity [5, 20, 34, 43].

Many congenital kyphotic deformities progress and subsequently become rigid [15]. Growth of the anterior portion of the vertebra stops, and kyphosis gradually increases due to growth in the posterior part of the spinal column until complete formation of the entire spine. Deformity progression may lead to respiratory failure and neurological deficits [39]. The results of bracing are almost always unsatisfactory; therefore, surgical treatment is indicated in cases of severe cosmetic defects, deformity progression, neurological deficit, or dysfunction of internal organs [31]. Historically, treatment included *in situ* stabilization, reconstruction of the anterior spinal column, pedicle subtraction osteotomy, and posterior column osteotomies [2, 3, 6, 16]. The results of numerous clinical studies have demonstrated that posterior three-column osteotomy is the most effective intervention for correcting severe angular kyphosis [18, 35, 36, 40]. In this article, we present the experience of revision surgical treatment of isolated angular thoracolumbar kyphosis using posterior three-column osteotomy in an adolescent female patient.

The patient with multiple developmental anomalies and congenital angular thoracolumbar kyphoscoliosis underwent multiple surgical interventions. The kyphoscoliotic deformity was diagnosed at birth. Since an early age, the patient had severe lower paraparesis and pelvic dysfunction. Her hereditary history was normal. At the age of 1 month, the patient underwent surgical treatment for spinal dysraphism and dural herniation; at the age of 5 months ventriculoperitoneal shunting for hydrocephalus was performed. In early childhood, the patient was diagnosed with Lennox-Gastaut syndrome. At the age of 5 years, the deformity began to rapidly progress with constant formation of bedsores in the deformity apex region. During the same period, clinical and MRI signs of Arnold-Chiari malformation were observed; the patient underwent surgical decompression of the posterior cranial fossa, resection of the C1 posterior arch, and subpial resection of the dysplastic cerebellar tonsils followed by dural reconstruction. At the age of 7 years, the patient was counseled on progression of the kyphoscoliotic deformity with the development of lower paraplegia.

Clinical examination data: lack of motor activity and sensitivity in the lower limbs, pelvic dysfunction (neurogenic bladder, fecal incontinence). Kyphoscoliotic deformity, postoperative scars after previous interventions, and a bedsore in the deformity apex region were observed (Fig. 1). The respiratory function was moderately impaired. X-rays revealed angular thoracolumbar kyphoscoliosis with a failure of segmentation of the L1–L2 vertebrae; wherein, the deformity curve between T8 and S1 was characterized by kyphosis of 93° and scoliosis of 34° (Fig. 2). Neural arch defects were present at the levels from T11 to S1. MRI revealed severe spinal cord deformity with pronounced compression and myelopathy at the deformity apex.

Case conference discussion lead to a decision to perform surgical correction of the kyphotic deformity and decompression of the spinal canal contents due to the development of neurological complications, compromising respiratory function, and chronic formation of bedsores at the deformity apex. The

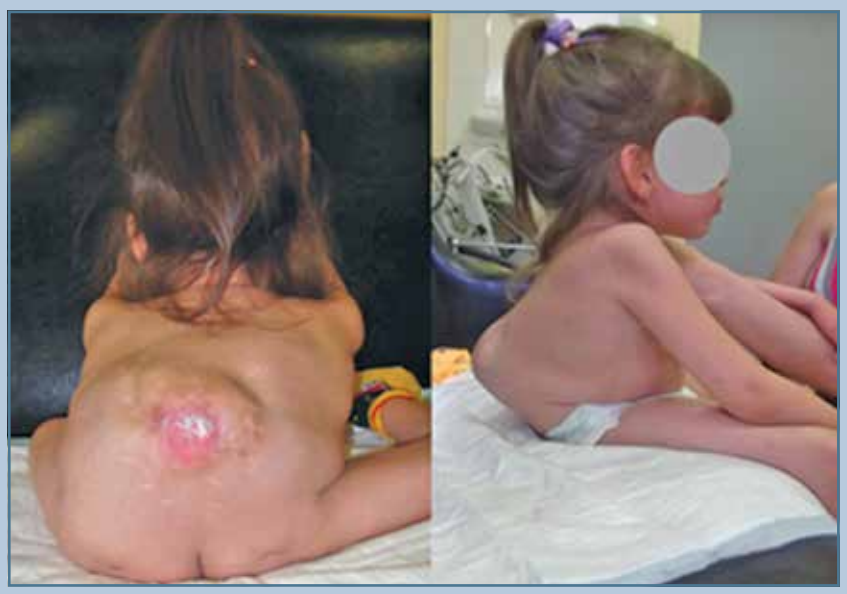

Fig. 1
Appearance of the patient at the age of 7 years: severe kyphoscoliotic deformity and a bedsore in the deformity apex projection are present

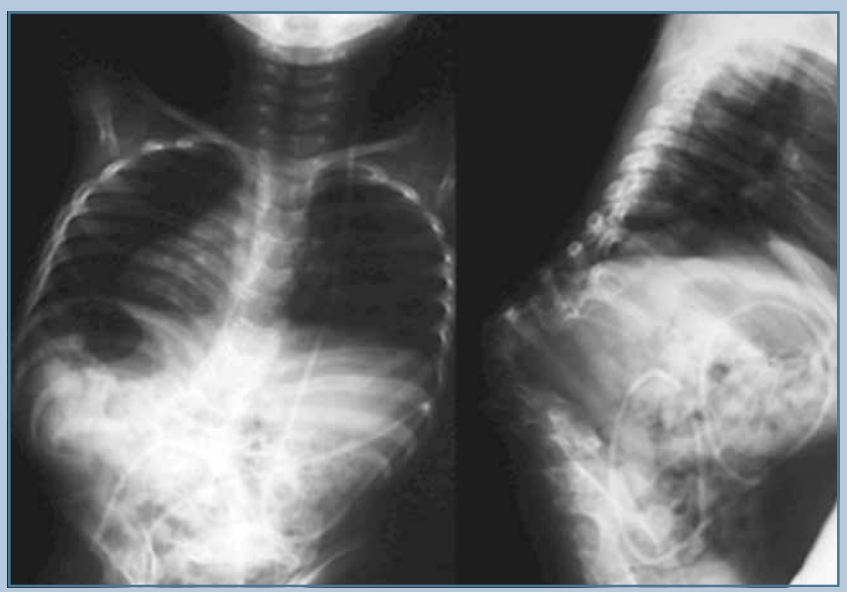

Fig. 2
Radiographs of the patient: severe kyphotic deformity, L1–L2 vertebral malformation, and pelvic hypoplasia

patient underwent posterior resection of the vertebral column at the L1 level and fixation of the spine with metal implants from the middle thoracic spine to iliac bones, which resulted in partial correction of the kyphotic deformity (about 37°; Fig. 3). However, a year later, the patient developed instability around the lower pole of instrumentation on the right: on examination, the rod protruded subcutaneously.

The right rod and two distal screws were removed during surgery; however, infection in the area of instrumentation developed within the next year, which led to fistulous tract formation with periodic episodes of fever (Fig. 4). During the next surgery, the second rod and other instrumentation elements except for screws in the thoracic spine were removed (Fig. 5), which led to a significant loss of the achieved correction. The patient received conservative treatment: bracing and a course of antibiotic therapy. Later, she was hospitalized for repeated instrumentation.

Surgical technique. The patient was admitted to the hospital for repeated revision surgery at the age of 13 years. After the course of antibiotic therapy, there were no signs of continuing infectious process. The deformity angle returned to its initial value (about 90°). Surgery was complicated by the presence of a bone block in the deformity apex area and throughout some of the previously fixed segments, which limited mobility of the deformity and the possibility of its correction. For this reason, detailed preoperative planning was performed, including generation of a CT-based three-dimensional stereolithographic model of the spine (Fig. 6). At the deformity apex, we performed a posterior three-column osteotomy of the spine as well as correction and stabilization using instrumentation from the middle thoracic spine to the pelvis with insertion of screws into the iliac wings.

Under general endotracheal anesthesia, the patient was placed on the operating table in such a way as to provide the best access to the deformity apex and to exclude pressure on the bony prominences (Fig. 7). A midline incision was made, and posterior elements of the vertebrae were accessed through the posterior approach. The existing screws in the thoracic spine were removed and replaced with screws of a larger diameter. Additional screws were inserted through the pedicles of the L4 and S1 vertebrae where the anatomical structures were preserved; also, screws were placed into

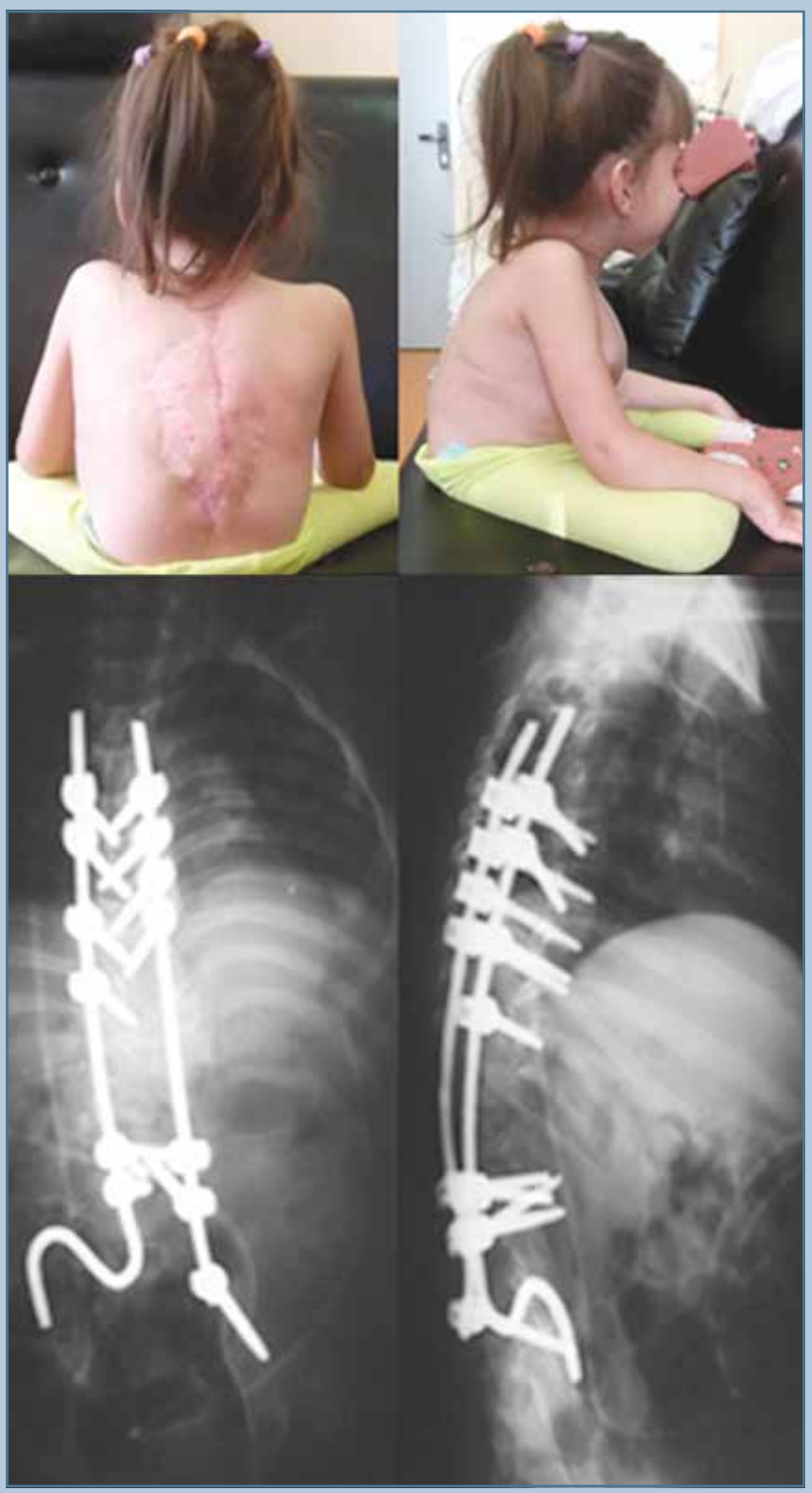

Fig. 3
Appearance and radiographs of the patient 1 month after surgery: partial surgical correction of the deformity is achieved

both iliac wings. At the next stage of surgery, a three-column osteotomy was performed at the L3 level to achieve adequate correction of the deformity. The mean arterial pressure during osteotomy was maintained at values above 80 mm Hg. The lateral surfaces of the L3 vertebra were subperiosteally isolated. During the osteotomy, temporary rods were used on the contralateral side. The posterior elements and transverse processes of L3 were resected. Then, a wedge resection of the vertebral body with preservation of the posterior cortical wall was performed. At the last stage, the posterior vertebral wall was shifted anteriorly and removed. Pre-modeled titanium rods were placed into screw heads and fixed with set-screws. During placement, step-by-step gradual compression of screw heads on the rods was performed in order to close the osteotomy region and correct the kyphosis. After radiographic control, local bone autograft obtained during the osteotomy was used for additional fusion. The wound was closed in layers, and active drains were placed. The patient was activated in a brace on the 3rd postoperative day.

The surgery time was 210 min, and intraoperative blood loss was 920 mL. A significant correction of the kyphotic deformity was achieved: 55° in the sagittal plane and 18° in the frontal plane. In the early postoperative period, there were no changes in the neurological status or infectious complications in the postoperative wound area. At the same time, significant deformity correction led to displacement of the ventriculoperitoneal shunt. The patient developed a strong cephalgic syndrome; for this reason, the patient underwent surgery for replacement of the ventriculoperitoneal shunt in a neurosurgical hospital 2 weeks after discharge. Follow-up radiography at 18 months revealed a bone block in the osteotomy area; correction of 50° in the sagittal plane was preserved. During this period, the patient had no headache episodes, bedsores in the deformity apex projection, or infectious complications. A satisfactory cosmetic outcome was achieved (Fig. 8). The patient was able to sit in a comfortable manner; dyspnea

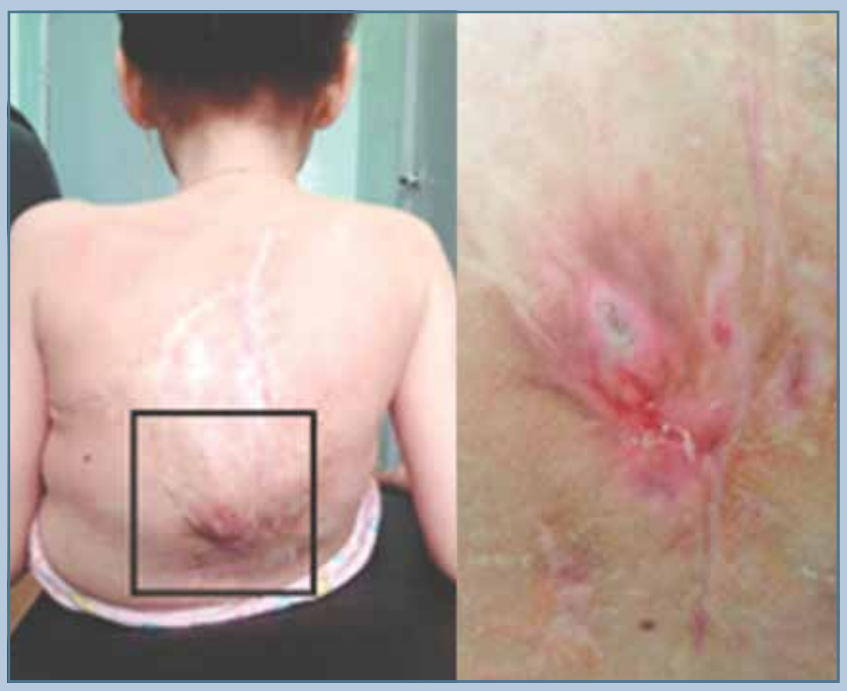

Fig. 4
Appearance of the patient and the fistulous tract formed near the lower pole of instrumentation (in the left rod projection)

episodes ceased; she noted a wider range of motion in the upper limbs and a general improvement in the quality of life.

Discussion

The purpose of surgical treatment of congenital kyphotic and kyphoscoliotic deformities is decompression of the neural structures and correction of the deformity with subsequent stabilization that is aimed to prevent progression of the underlying disease. A three-column osteotomy of the spine is recommended in cases requiring resection of the apex of kyphotic deformity, which is accompanied by compression of the spinal cord or cauda equina. In the treatment of severe kyphotic deformities caused by a vertebral fracture (traumatic or pathological) or vertebral malformations, vertebral column resection (VCR) is most often performed through the posterior approach. On the other hand, in the case of moderate deformities, in particular in the case of partial hemivertebra or impaired segmentation, a pedicle subtraction osteotomy (PSO) is usually sufficient. The

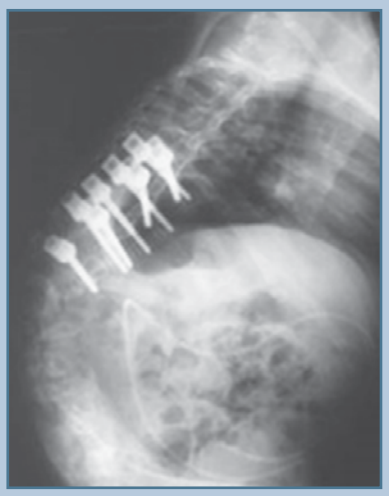

Fig. 5
Lateral radiograph of the patient after removal of the second rod and screws in the lumbar spine: screws in the thoracic spine were not removed

Рис. 6
Three-dimensional stereolithographic model of the patient's spine

three-column osteotomy techniques are indispensable in the surgical treatment of congenital kyphotic deformities as significant mobilization of the spinal segments is required for correction. In these patients, especially in the presence of other developmental anomalies, deformity progression may be associated with aggravation of phenomena such as decreased lung capacity parameters and constriction of the abdominal cavity, which is accompanied by deterioration of functional disorders. Some patients may even lose the ability to use a wheelchair [12]. Conservative treatment, bracing, and modification of wheelchairs are ineffective due to a high rate of chronic bedsores and other trophic tissue disorders similar to those observed in the presented case [14, 21, 22, 24, 25, 29]. Surgical treatment is the best choice for a number of reasons, including correction of the deformity and balance of the spine and trunk, relief or management of pain, improvement of lung capacity parameters, and prevention of bedsores in the deformity apex area [9, 10, 14, 17, 19, 30].

The choice of surgical modality should be strictly individual and be based on the opinions of several specialists, including a pediatrician, a pediatric neurologist, a pediatric neurosurgeon, and a trauma-orthopedist. These patients are at high risk of complications because the spinal cord is subjected to compression for a long time and can be additionally damaged during surgery due to excessive tension, tethering, injury to the feeding arteries, or a decrease in perfusion [34, 43]. For these reasons, these interventions should be performed with mandatory participation of a pediatric neurosurgeon [5, 20, 27, 41]. In the described case, the patient had a displacement of the ventriculoperitoneal shunt. Aggressive correction of deformity is associated with an increased risk of this complication; therefore, patients with a history of shunting surgery should consult a neurosurgeon in advance.

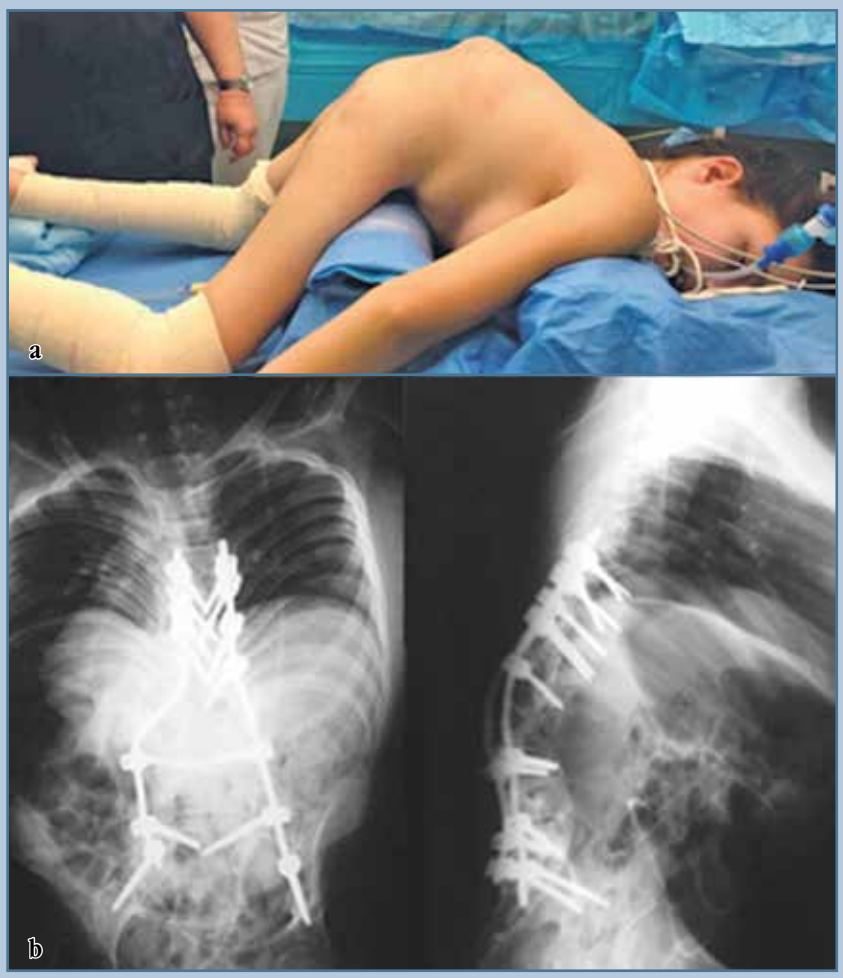

Fig. 7
Patient positioning on the operating table **(a)** and postoperative radiographs **(b)**: significant deformity correction is achieved

Traditionally, surgical correction of congenital kyphotic deformities is performed through a posterior approach if the deformity angle is less than 50°. If the angle is more than 50°, a combined (anterior + posterior) approach is more preferable [31, 39]. However, the presence of an additional approach is associated with certain drawbacks: additional surgical access, a higher risk of injury to the main vessels and internal organs, and complications associated with autobone harvesting [23, 27, 33, 37]. If surgery through the posterior approach does not provide the desired result or a pronounced bone block forms in the deformity apex region, we believe that the use of a three-column osteotomy is optimal. The three-dimensional stereolithographic model of the patient's spine enabled careful planning of both insertion of screws at strictly defined levels and the osteotomy stage to reduce risks of potential complications. Chronic bedsores at the deformity apex, history of surgical site infections , deformity severity, and bone block throughout the deformity significantly complicated the task in this patient. Single-stage surgery with a three-column osteotomy through the posterior approach was planned. Given the chronic bedsores at the deformity apex and a high risk of recurrent infectious complications, conservative treatment was not considered.

Various techniques have been proposed for correction of pronounced kyphoscoliotic deformities in pediatric and adolescent patients, including

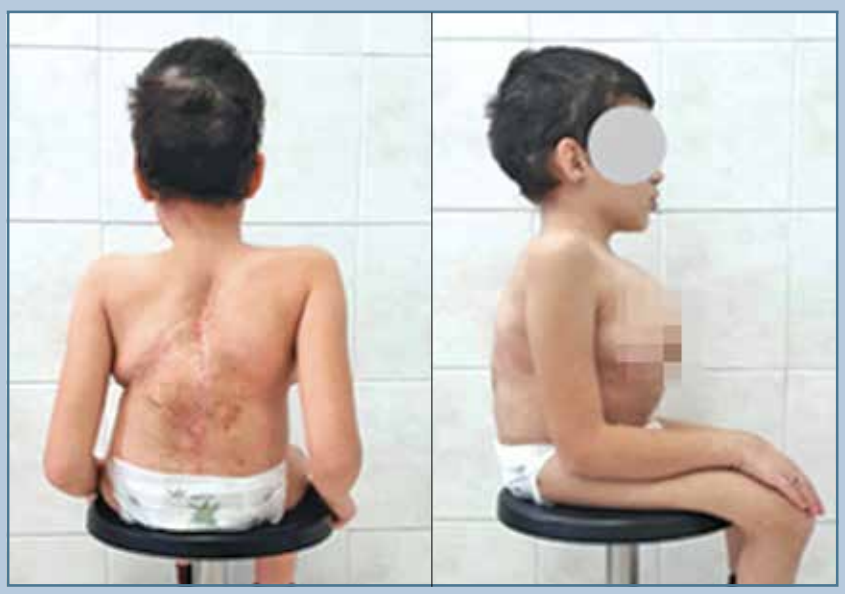

Fig. 8
Appearance of the patient on a follow-up examination 18 months after surgery

surgery through combined and isolated posterior approaches using different implants; however, no standard approach has been developed to date [7, 11, 23, 26, 28, 33, 38]. Most surgeries for congenital kyphotic deformities described in literature were primary ones and did not require revision interventions. However, the authors who have performed revision in these patients consider single-stage surgery through the posterior approach and aggressive correction to be the optimal choice [7, 42]. Surgical treatment of congenital kyphoscoliotic deformities in pediatric and adolescent patients is associated with complications, such as pseudarthrosis, CSF leakage, respiratory distress syndrome, and urogenital tract infections. According to various authors [1, 4, 8, 13, 32], the rate of these complications ranges from 9 to 49%, with the rate of infectious complications being relatively low (up to 5 %). In these cases, adequate antibiotic therapy is of primary importance. At the same time, in the case of chronic infectious complications and a low efficacy of antibiotic therapy, partial or complete removal of instrumentation system should be considered.

Conclusion

The key to successful correction of congenital kyphoscoliotic deformities is the correct evaluation of the type, level, and risk of further deformity progression with continued patient growth, as well as thorough preoperative planning. The planning stage is greatly simplified by using 3D modeling techniques. Since severe kyphotic deformities provide less favorable conditions for the spinal cord than scoliotic ones, it is extremely important to consider the limits of the possible correction amount as well as potential neurological complications. Incorrect assessment of risks may lead to serious consequences due to untimely or tactically incorrect surgical intervention or refusal of surgery. The management of these patients requires a multidisciplinary approach and treatment in dedicated centers of pediatric spinal surgery. Both primary and revision surgeries require an individual approach to each patient with consideration of an increased risk of complications associated with three-column osteotomies that provide adequate correction of deformity in many cases.

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