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A REVIEW OF TREATMENT CONGENITAL DEVELOPMENTAL ANOMALY OF VERTEBRA - HEMIVERTEBRA IN CHILDREN

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ABSTRACT

Congenital scoliosis causing malformations of the vertebrae, often leading to severe and rigid spinal deformity in young patients, causing a gross cosmetic defect and impaired biomechanics. In rare cases of spinal diseases in children, congenital malformations occupy a very small place - from 2 to 11%. Congenital scoliosis occurs with a frequency of 0.5-1 per 1000 live births. The role of natural and environmental factors in its etiology has not been fully elucidated. Study of the premise that the spine develops due to a developmental disorder in the fifth and eighth weeks of pregnancy. The pathogenesis of hemivertebrae can be understood from the embryonic stage. Development of the spine occurs in the sixth week of pregnancy, when two late central chondrifications occur in the deviations of the vertebral bodies. By 7-8 weeks of gestation, these centers of chondrification merge, forming the primary center of ossification of the body, which temporarily opens into the anterior and posterior parts with the remainder of the notochord. Indications for surgical intervention in congenital scoliosis should be considered in terms of the severity of the existing deformity and the prospects for its further progression. With surgical intervention in childhood, it is possible to prevent the progression and further development of gross deformities of the spine.

KEYWORDS: Congenital scoliosis, hemivertebra, anterior - posterior approach, posterior approach.

INTRODUCTION

Congenital scoliosis causing malformations of the vertebrae, often leading to severe and rigid spinal deformity in young patients, causing a gross cosmetic defect and impaired biomechanics. In rare cases of spinal diseases in children, congenital malformations occupy a very small place - from 2 to 11%. Congenital scoliosis occurs with a frequency of 0.5-1 per 1000 live births.

A hemivertebra is a congenital anomaly of the spine. This can lead to three main forms of deformity: scoliosis (most common), kyphosis, or lordosis (least common). Most hemivertebrae, with the exception of some unsegmented or pinched forms, have normal growth plates, so progressive deformity occurs as they grow. As a result of local deformation and asymmetric loads, neighboring vertebrae can also have asymmetric growth. Secondary curves may further develop to help balance the torso.

Etiology and pathogenesis

The role of genetic and environmental factors in its etiology has not been fully elucidated. Research has suggested that the spine suffers due to a developmental disorder in the fifth and eighth weeks of pregnancy. These studies have emphasized that spinal malformations may occur as a result of segmentation and anomaly formation, or a combination of the two. Also, vertebral deformities in congenital scoliosis have been associated with genetic syndromes such as Alagille syndrome, spondylocostal dysostosis, and Jarcho-Levin syndrome. Идиопатический сколиоз наблюдается в обществе от 0,2% до 3%. Various statements have argued that idiopathic scoliosis has a genetic background. It has been reported that an anomaly in the 17p11 gene may increase genetic research in large families with autosomal inward transmission. The etiological factors that determine the development of congenital scoliosis are not fully explained yet. Some environmental factors can lead to the development of congenital scoliosis. Exposure to carbon monoxide during pregnancy, use of alcohol or antiepileptic drugs such as valproic acid and dilantin. Other factors are hyperthermia and gestational diabetes diabetes mellitus. Research on congenital scoliosis has been prevalent in many institutions. Some of these studies are in the field of genetics. In case of family presence, the history was demonstrated in patients with congenital scoliosis. This genetic transition is combined with autosomal recessive and autosomal recessive dominance. Chromosomal analysis showed that deletions of 2p13-13, 6q13 and 15q12 were found, causing congenital scoliosis.[1,3]

In our case, the patient had no problems associated with the growth and development of diseases, as well as problems with urination or the gastrointestinal tract. He also had a normal appearance compared to other children. Genetic research has been shown to be responsible for some congenital vertebral anomalies; however, to date, there is no clear genetic etiology of congenital scoliosis.

In more discovered cases, it was reported that the spine bone and cartilage as tissues were histologically unremarkable, namely, their location was incorrect. In addition, no molecular abnormalities of collagen have been identified. Reported in bone, cartilage, or skin. A follow-up study reported a "normal" percentage of type I admission to the thoracic region. muscles that straighten the spine on the concave side, but a significantly higher percentage of such tumors on the convex side in patients with congenital scoliosis or scoliosis of early onset. A relatively recent study reported a decrease in proteoglycan levels and water content in the cartilage endplate and intervertebral disc, especially in the change in curve concavity in congenital scoliosis. It has been suggested that, albeit secondary, due to the change in load in scoliosis, cartilage calcification and lamina closure may be of great importance in the progression of the scoliotic curve.[2]

The pathogenesis of hemivertebrae can be understood from the embryonic stage. Spinal development occurs in the sixth week of pregnancy, when two lateral centers of chondrification appear in the developing vertebral bodies. By 7-8 weeks of gestation, these centers of chondrification merge, forming the primary center of ossification of the vertebral body, which is temporarily divided into anterior and posterior parts by the remainder of the notochord. Lack of development of one of the paired chondral centers results in a lateral hemivertebra, while, more rarely, insufficiency of the anterior ossification center results in a posterior hemivertebra. The defective pathogen causes a contralateral deviation of the spine at the level of the abnormal spine, acting as a triangular wedge-shaped ossified structure within the spinal column. Based on the presence or absence of normal disc space and below the affected segment, four types of hemivertebrae can be distinguished: fully segmented (the most common type), semi-segmental, non-segmented, and strangulated. In this case, the malformation showed a fully segmented hemivertebra on CT because, according to a study by Bao B et al., a fully segmented hemivertebra is the most attractive type among others. However, fully segmented coronal Y-disc hemivertebrae have the potential to worsen due to asymmetric growth when other types are identified.[2]

Classification

Congenital anomalies of the vertebrae are classified as defects in the shape of the vertebrae, a consequence of segmentation disorders or combined defects. The most common variant of the shape defect is the wedge-shaped vertebrae and hemivertebrae (hemivertebra), which, depending on the relationship with neighboring segments of the spinal column, are classified into four types: segmented, semi-segmented, wedged and non-segmented. The two lateral hemivertebrae are called the butterfly vertebrae.[20]

The hemivertebrae are the most common case of congenital scoliosis. A segmented hemivertebra has a similar growth potential to a normal vertebra, which creates a wedge-shaped deformity that progresses as the spine grows. McMaster and David found that the degree of scoliosis produced depends on four factors: first, the type of hemivertebra; secondly, its location; thirdly, the number of hemivertebrae and their relationship with each other; and finally, the age of the patient. Semi-segmented and pinched hemivertebrae usually cause lesser scoliosis. curve than fully segmented unimpaired hemivertebrae, but may still cause significant deformity. Complete segments of unguided hemivertebrae have normal growth plates and more often require prophylactic surgery to prevent defects from being detected. Hemivertebrae facing the sidebar of the cover with the worst prognosis, two unilateral hemivertebrae and a single hemivertebra are present behind the objects.

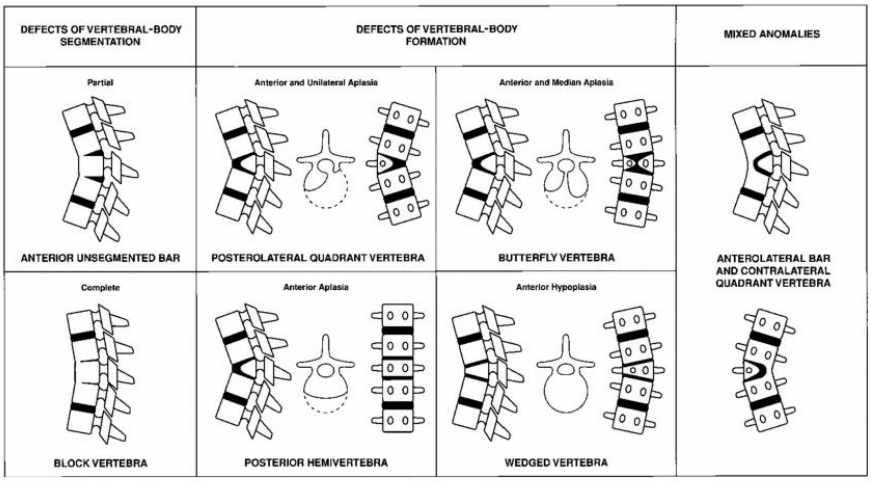

Figure 1. Drawings showing the different types of vertebral anomalies that produce a congenital kyphosis or kyphoscoliosis.

Diagnostics. The physical examination should begin with determining the height and weight of the patient, given that height plays a significant role in the progression of the curve. The skin should be checked for abnormalities such as café-au-lait spots or axillary freckles, neurofibromatosis inflammation, midline hairline for any signs of spinal dysraphia. Spinal dysraphia can also rotate in the extremities, including asymmetrical calves, hollow feet, clubfoot, vertical talus, and abnormal neurological symptoms. The spine examination itself focuses on any signs of trunk or pelvic imbalance. Chest deformity, chest or lateral wall mass, chest excursion and abnormalities, and inspiratory and expiratory chest strength should be assessed. Restriction of examination of the chest can be manifested by scoliosis syndrome and thoracic insufficiency syndrome. In young children, Adam tests for forward flexion (identification of protruding ribs in the thoracic spine or transverse processes in the lumbar spine) is not possible, but the test can be simulated with the child lying on the examiner's knee. Flexibility of the wire can be assessed by placing the child in a lateral position above the examiner's knee or by hanging the child on the examiner's arm. It is necessary to assess the balance of the spine in both the coronal and sagittal planes. Attention should be paid to trunk imbalance, head tilt, uneven shoulders and pelvic balance. Photos are an extremely important aid in the consistent evaluation of progress. includes a combination of anomalies of the nervous axis and the possibility of neurological involvement in congenital deformities of the spine, a mandatory study of motor, sensory and reflexes (including abdominal reflexes). Vital capacity screening is recommended for patients with flow and deformities. A complete spirometry examination is recommended if surgery is planned in patients with a vital capacity < 60% of normal.

Sustainable changes include relatively possible:
1) local congenital deformity of the spine, which is manifested by a combination of shoulder girdle, waist triangles, pelvic tilt;
2) fatigue, fatigue;
3) neurological deficit.

Beam indicators of instability:
1) scoliotic and/or kyphotic deformity of the spine at the level of 2–3 spinal motion segments;
2) violation of the frontal and sagittal profile of the deformed spine;
3) stenosis of the spinal canal;
4) vertebromedullary conflict.[8]

To determine the nature of the course of congenital spinal deformity in children, X-ray control is carried out at reasonable time intervals. So Shen and Arlet believe that up to 5 years it is necessary to observe the age of the child 6–9 months, from 5 to 10 years - 1 time per year, after 10 years until the completion of the development of the skeleton - about 6 months. Winter beliefs about the need to perform a spondylogram in any child with a congenital malformation of the vertebrae are usually 6 months old. in the process of growth, fearing a sudden progression of the deformity, it even manifests itself in an unfavorable course. Qualitative radiological indicators for assessing the risk of progression of congenital spinal deformity take into account, to varying degrees of significance, the morphological variant of the defect, the increase in the magnitude of the deformity in degrees per year (according to Cobb), penetration and the number of abnormal vertebrae.

A perinatal history is necessary to differentiate the hemivertebra from other causes of early scoliosis. Most often, vertebral defects or scoliosis are detected on a chest x-ray. Whether the patient has a hump or a defect found incidentally on a chest x-ray, a full examination is required, unlike scoliosis, which may be seen initially. An MRI of the spine should be done to look for possible associated spinal cord anomalies such as tethering, split spinal cord malformations, and occluded dysraphism. X-rays are used to assess the flexibility of the compensatory curves and measure Cobb angles for comparison during follow-up. Projection in the push position (by pressing the kyphosis during CT provides the best visualization of the bones, and bony septa within the canal can be identified. 3D reconstructions can highlight the posterolateral hemivertebrae very well.[3,5]

Conservative treatment
Conservative therapy included physical therapy aimed at strengthening the muscles of the back, shoulder girdle and abdominal muscles, stimulating back massage, physiotherapy, swimming and water procedures. Therapeutic physical training (static and dynamic exercises) was prescribed for children older than three years, it was recommended to perform it in a playful way every day, at least 30-40 minutes a day. Back massage was carried out in courses 2-3 times a year for 15 sessions. Symmetrical swimming styles (breaststroke, butterfly) were recommended for children over five years of age as water procedures. With an initial value of the main local scoliotic curve of deformity in the lumbar spine of less than 30°, a stable course of curvature was observed in 65% of children on the background of conservative therapy. The stability of the course of congenital deformity during dynamic observation was manifested by the preservation of the initial value of the scoliotic angle in 5% of patients, a slight progression of the local main curve, not exceeding 4° in 4 years - in 37.5% of children, a tendency to self-correction of congenital curvature within 1 -3° - in 22.5%. All children with an initial value of the manifestation of the scoliotic curve of deformity of more than 30° have a steady progression of congenital curvature within 6–13° over 4 years of dynamic follow-up.[18]

Surgical treatment Nowadays surgical treatment is used by anterior – posterior or only posterior approaches. It

depend on paints age and character of spine derformation.

The first case of hemivertebra resection was described by Royle in 1928. Since then, many studies have demonstrated positive results after hemivertebra resection with a combined anterior and posterior approach in a one- or two-stage pattern. In 1976 Leatherman, Dixon and Slabo et al. used Harrington compression rods in 25 patients and anterior and posterior distraction rods in 4 patients. In 1979, large versions of hemivertebra resections were described using a two-stage anterior-posterior procedure. They proposed resection of 50 patients with congenital scoliosis.

In patients with an increased risk of posterior element resection with spinal fusion. The angle of scoliosis averages 77° before surgery and can be corrected to 41°, and the degree of severity of the angle reaches 44.2%. The angle of kyphosis before the operation is 72°, and after the operation it increases to 40°. In 1980, Slabo et al. reported 8 lumbosacral hemivertebra resections and again performed a two-stage operation with correction from 39° to 25°; the degree of increase in the degree of danger is 35.9%. In 1986 Bergoin et al. 19 hemivertebra resections were identified in 9 children who underwent one-stage posterior and anterior surgery and achieved an average correction of scoliosis from 30° to 16° and an average correction of kyphosis from 33° to 12°. In 1990, Bradford and Boachie-Adjei reported on 7 lumbar resections of RA using a combined approach and without instruments. This showed a mean improvement in circulation of 68.1% along a curve from 47° to 15° with a mean follow-up of 4.6 years. The period of wearing a plaster cast or corset is from 7 months to 1 year. King and Lowery used Harrington rods (6 cases) or uninstrumented (1 case) after RA resection by combined approach. The mean improvement was 24.3% from 37° to 28° and patients were required to lie supine for 6 weeks to 3 months. The mean curvature reaches 54° (range 18–132°) before surgery, 33° (range 0–105°) after surgery, and 35° (0–110°) at the last velocity assessment. there was an improvement of 60% from 40° to 16°. Lazar and Hall also used one compression rod in 11 patients with nos. resection through combined approach, mean tumor enlargement 70.2% from 47° to 14°, but the mean result was only 2.3 years. Klemmit et al. used a large sublaminar suture tape in 6 patients after RA resection. through dual access. The level of evaluation of the result is 71.1% from 38° to 11°, but the duration of the observation period is only 3.5 years. Bollini et al. for thoracolumbar, lumbosacral PP. were treated with RA resection using a combined posterior and anterior view and short anterior and posterior convex fusion, respectively.[6,9,10]

Vissarionov S.V. et all. was performed from a combined (anterolateral and dorsal) approach. The technique of surgical intervention in patients of preschool age consisted of a single-stage three-stage operationAs a result of instrumental correction of spinal deformity on the lateral hemivertebrae after surgical treatment of the angle of scoliotic hearing deformity from 0 to 6°. Increase risk correction from 94% to 100%. After extirpation of the posterolateral hemivertebrae, the angle of the residual scoliotic component of bone deformity was from 0 to 4° (average 2.5°), the degree of severity correction was from 95 to 100%, the angle of the kyphotic component was from 9 to -6° (average 2.2°). 1.5–2 years after the operation, when a pronounced bone block was formed in the area of surgical intervention, the removal of metal structures was performed. After the removal of the spinal implant, the loss of correction does not occur in any case.[19]

Posterior approach - Recently, several studies have focused only on the posterior approach. Shono et al. reported posterior RA resections in 12 patients with a 63.3% improvement from 49° to a single 18° in 2001. The later procedures described are also of great importance, as operative time and the rate of significant contraction are reduced in this collection. Nakamura et al. identified 5 cases of RA resection with a 55.1% improvement for 3 thoracolumbar RAs and 31.4% for 3 lumbosacral RAs from 35° to 24°. In 2002, Ruf and Harms reported 21 consecutive cases of congenital scoliosis that were treated with RA resection using a posterior approach with a pedicle instrument. The results revealed 1 infection, 1 pedicle fracture, and 2 failure to use wire instruments.[9]

Aidogan et al. assessed the surgical and radiological results of resection of the posterior sinuses and instrumentation only from the posterior approach. They have a correction factor for gastric scoliosis of 75%, kyphosis - 84%, and kyphoscoliosis - 67%. Merger was achieved after a median of 9 months (range: 5–15 months). In 2009, Ruf et al. Completed a retrospective study of 41 aged 1–6 years with congenital scoliosis operated on via posterior resection of the right side only with a transpedicular instrument. In patients without bridge formation, the frequency of correction of the main curve was 80.5%. The craniocaudal compensatory curve improved by 80% and 76.5%, and segmental kyphosis improved from 22° to 8° points. In patients, the improvement in the main curve was 66.7%, and the frequency of correction of the cranial and caudal compensatory curve was 74.8 and 75.1%. The improvement in segmental kyphosis was 62.5% (from 24° to 9°). Posterior hemivertebra resection with transpeduncular instrumentation is an ideal procedure for early correction in young children. Correction should be done early, even in very young children.[10]

In 2011, Zhang et al. retrospectively studied 56 consecutive cases of patients with congenital scoliosis who underwent posterior resection of the right leg with transpedicular instrumentation. The mean improvement in segmental scoliosis was 72.9% from 42.4° preoperatively to 12.3° at the last follow-up, and a mean

improvement of 70% in segmental kyphosis from 42.0° to 14.5° over the same period.[11]

In 2013 Wang et al. analyzed 36 consecutive cases of patients with congenital scoliosis identified as a result of a fully segmented RA and treated with posterior resection of the RA with bisegmental fusion. Segmental scoliosis was corrected from 36.6° to 5.1° with a correction rate of 86.1%, and segmental kyphosis from 21.2° to 5.8° at the last examination. The frequency of correction of the compensatory cranio-caudal arch is estimated at 76.4 and 75.1%.

In 2014, Crostelli et al. examined 15 cases of congenital scoliosis with lumbar or thoracolumbar PP in children under 10 years of age. Patients underwent posterior resection of the RA. and pedicle screws with two levels of stabilization or with three or more levels of stabilization. The mean value of the scoliosis curve was 44° on the Cobb scale and decreased to 11° after surgery. The mean value of segmental kyphosis was 19.7° and decreased to 1.8° after surgery. Zhu et al. Examined 60 patients with congenital scoliosis (age 2–18 years) who underwent posterior resection of the right vein and monosegmental fusion. The mean preoperative angle was 41.6° which was corrected to 5.1° postoperatively and 5.3° at the last follow-up visit (87.3% correction). The compensatory curvature of the skull improved from 18.1° before surgery to 7.1° after surgery and 6.5° at the last follow-up examination. The mean Cobb angle in the sagittal plane was 23.3° preoperatively, 7.3° postoperatively, and 6.8° at the last follow-up visit. The correction factor was 70.1%.[12]

The first long-term observation of surgical outcomes of posterior RA resection and short segment fusion using segmental fixation with pedicle screws was reported by Changet et al. 52 in 2015. The mean Cobb angle of the main arch was 34.4° before surgery, 8.6° after surgery, and 12.9° at last follow-up visit. There were no crankshaft phenomena and clinical and radiological signs that could be indicative of spinal stenosis at the time of follow-up. There were also no major vascular or neurological complications associated with pedicle screws. Meanwhile, Piantoni et al conducted a retrospective study of patients with congenital scoliosis. about the hemivertebra (PP) and instrumental resection was performed through the posterior approach with remote observation. A total of 67 patients with 78 PP and 70 surgical interventions were evaluated. The data obtained showed that the mean follow-up period was 6.55 years, the scoliosis-mean preoperative angle was 38.55°, and the mean postoperative angle was 19.89°. The mean kyphosis angle before surgery was 29.98, and the mean angle after surgery was 15.41°.[13]

In recent years, researchers have attempted to evaluate the effectiveness of RA resection. Guo et al. assessed the complications and efficacy of posterior resection of the RA in young patients. The mean improvement was 83.6% in segmental scoliosis from 38.4° preoperative to 6.3° postoperative and the mean improvement was 81.9% in segmental kyphosis from 17.1° to 3.2° over the same period. The frequency of spontaneous correction of the compensatory cranial and compensatory caudal arches was 65.7% and 66.9%, respectively.[14]

In 2017, Erturer et al. evaluated the radiographic and clinical outcomes of patients who underwent deformity correction and stabilization of congenital spinal deformities using pedicle screws after completion of RA resections. The amount of correction on the coronal planes was 31%. the mean segmental angle of kyphosis was 45.7° before surgery and 2.7° in the postoperative period.[10]

Juan et al. assessed the efficacy and safety of one-stage posterior resection of the RA for unbalanced multiple hemivertebrae. The mean Cobb angle of the main curve was corrected by 73.3%. Compensatory cranial and caudal curves were corrected by 70.0% and 79.1%, respectively, and segmental kyphosis/lordosis was corrected by 65.5%.[15]

Basu et al. reported on the effectiveness of posterior RA resection in single and multi-level RA and compared the results in 2016. The average achieved rate of coronal and sagittal Cobb angle correction was 50.2% and 51.8%, respectively. As a result, it was concluded that posterior RA resection for congenital scoliosis is a safe treatment option for hemivertebrae. Lumbosacral RA can also be treated with posterior RA resection. In 2015, Zhuang et al. retrospectively studied a consecutive series of 14 cases of congenital scoliosis caused by lumbosacral RA and treated by posterior resection of LA with short segmental fusion, with a follow-up period of at least 2 years.[20]

In 2019, Miguel et al. analyzed a sequential cohort of young patients treated for congenital scoliosis secondary to a single PP and compared outcomes for thoracolumbar and lumbosacral curvature. The mean preoperative Cobb angle was corrected by 65% postoperatively and by 50% at the last follow-up. The compensatory cranial curvature improved spontaneously from 25.3° to 13.5° and finally to 19.9°. Preoperative coronary balance was corrected postoperatively but worsened at the final follow-up. The curvature of the thoracolumbar spine was corrected earlier than the curvature of the thoracolumbar spine.[20]

CONCLUSION

Indications for surgical intervention in congenital scoliosis should be considered in terms of the severity of the existing deformity and the prospects for its further progression. With surgical intervention in childhood, it is possible to prevent the progression and further development of gross deformities of the spine.

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