Spondyloptosis in children, adolescents and youth age patients

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Abstract
The study aimed to analyze scientific publications on “Spondyloptosis in children, adolescents and youth age patients”. The article analyzes 77 literary sources for the period from 1995 to 2021, presented in modern electronic databases of medical information: PubMed, CyberLeninka, eLibrary, Google Scholar. The analysis of scientific articles showed that many important issues related to spondyloptosis in children have not yet been resolved. For example, the disease incidence rate in children and adolescents is unknown. To date, an algorithm for choosing a method for surgical treatment in this category of patients has not been defined, the need and methods for reduction of a displaced L\textsubscript{v} vertebra remain debatable, the spinal fusion length is not scientifically justified, measures to prevent the appearance or exacerbation of neurological disorders have not been developed, there are no generally accepted clinical and radiation criteria for evaluating treatment outcomes. It is recognized that the severity of clinical manifestations of spondyloptosis is associated with the degree of spinal-pelvic imbalance. The range of surgical interventions is wide: from “in situ” fusion at the L\textsubscript{v}–S\textsubscript{i} motion segment to 360° reconstruction with a change in the parameters of the lumbo-pelvic balance. Successful attempts are being made to introduce into clinical practice composite models of metal structures individually made on a 3D printer, specific to the spinal-pelvic balance of a particular patient. Many authors in their publications expressed that various aspects of L\textsubscript{v} spondyloptosis in children and adolescents require further study.

Keywords: “high-grade” spondylolisthesis, congenital spondyloptosis, dysplastic spondyloptosis, children, adolescents and youth age patients.

a separate, most severe, degree V spondylolisthesis of the Lv vertebra in 1956 [8]. These German authors elaborated on the well-known classification of H.W. Meyerding, according to which all cases of spondylolysis are divided into four degrees, according to the magnitude of the anterior displacement of the lumbar vertebra involved in the pathological process, which is determined on the radiograph of the lumbar spine and sacrum in the lateral projection [9].

In subsequent years and up to recently, in the clinical practice of vertebrology, along with H.W. Meyerding’s classification, the classifications of spondylolisthesis proposed by L.L. Wiltze et al. [10], I.M. Mitbreit [11], P.G. Marchetti and P. Bartolozzi [12], and other authors are widely used.

After an international research team (Spinal Deformity Study Group, SDSG) published the results of a study on the sagittal spinal–pelvic balance in patients with degenerative spinal deformities in 2005–2006, Canadian scientists J.M. Mac-Thiong and H. Labelle developed a classification of pediatric lumbosacral spondylolisthesis [13]. The proposed classification is based on a comprehensive assessment of the three most important parameters, namely the degree of displacement of the vertebral body, degree of pelvic tilt, and degree of the spinal–pelvic balance. According to these classification criteria, spondylolisthesis in children is categorized as high-grade vertebral displacement, with a major pelvic retroversion and a great sacral tilt. Along with the above parameters of lumbosacral segment disorders, anterior displacement of the hip joints in patients is significant in the pathogenesis of spondylolisthesis [5]. This further disrupts the spine orientation relative to the sacrum and lower extremities, being a high risk factor for fracture of the interarticular part of the LV vertebral arch [14].

In addition to disorders of the spinal–pelvic balance in children with spondylolisthesis, as a rule, multiple dysplasias and abnormalities in the development of the lumbosacral spine are diagnosed, which become the foundations on which the disease develops and progresses during the postnatal period [15, 16]. Thus, M.W. Al Sebay et al. demonstrated a clinical example of the diagnosis of bilateral spondylosis of the interarticular part of the arches of the vertebrae $L_{III}$, $L_{IV}$, and $L_{V}$, spondylolisthesis of the $L_{V}$ vertebra in a female adolescent with established spondylolisthesis [17].

A research team from Singapore, under the supervision of W.M. Yue who has over 23 years of experience, analyzed the results of the complex radiological diagnosis in 27 patients who underwent surgery for spondylolisthesis due to the presence of dysplastic symptoms of the vertebrae. In all cases, the patients had a domed shape of the upper parts of the vertebra $S_{II}$. Spondylolysis lines of the interarticular part of the $L_{IV}$ vertebral arches and non-closure of the posterior part of the arches of the upper sacral vertebrae were established in 88.9% of clinical cases, respectively. The trapezoidal shape of the $L_{V}$ vertebra was registered in 74.1% of cases. Moreover, the abnormality of tropism and hypoplasia of the articular processes of the lower lumbar spinal motion segments were diagnosed in in 59.2% of patients [18].

In the above mentioned dysplasia and anomalies in the lumbosacral junction development in patients with spondylolisthesis, the domed shape of the upper sacrum is of great importance in the disease pathogenesis [19, 20]. Thus, because of complex dynamic monitoring of two girls for several years, G. Gutman et al. established that as the horizontally located upper endplate of the vertebra $S_{II}$ is transformed into a domed one, the angle of inclination and the degree of anterior displacement of the $L_{V}$ vertebra increase. Therefore, the progressive course of spondylolisthesis is especially pronounced when the patient has other dysplasias and anomalies in the development of the lumbosacral spine, primarily spondylosis of the vertebral arches [21]. Japanese researchers H. Manabe et al. experimentally established the leading pathogenetic role of the domed shape of the upper sacrum in the development of severe forms of childhood spondylolisthesis [22].

L.J. Curylo et al. confirmed that dysplasia of the posterior sacral support complex reduces the mechanical strength of the lumbosacral region and contributes to the shift of the caudal lumbar vertebra. According to the authors, out of 53 patients with high-grade spondylolisthesis and spondylolisthesis, dysplasia of the posterior elements of the spine was registered in 62% cases [23].

Notably, a study conducted by Russian orthopedists on 98 pediatric patients with various degrees of severity of spondylolisthesis enabled to objectively establish dysplasia and developmental anomalies in almost the same number of patients (in 64.27%; 63 pediatric patients). Furthermore, 30 (47.61%) pediatric patients were diagnosed with one developmental anomaly each, 19 (30.15%) pediatric patients had two, and 14 (22.24%) children had three or more developmental anomalies. In the range of anomalies diagnosed, cases of non-closure of $spina bifida posterior$ of the $L_{I}$ vertebra and/or sacral vertebrae were predominant. In total, in 63 pediatric cases of non-closure of $spina bifida posterior$ were registered in 89 vertebrae, while both of its forms, “apertura” and “occulta,” were identified. Moreover, $spina bifida posterior$ defects of individual vertebrae, cases of non-closure of the hiatus
spondyloptosis, an individual volume of surgical in-
gnosed disorders in each individual child with
chers R. Vialle et al., CT enables to assess most ac-
of the pathology [33]. According to French resear-
sonance imaging (MRI) performed in patients with
als, the degree of compression of the ducal sac,
the lumbosacral spine, including intervertebral
disks, the degree of compression of the ducal sac,
and the elements of the cauda equina and segmen-
tal roots located in it [35].

Depending on the nature and degree of the dia-
gnosed disorders in each individual child with
spondyloptosis, an individual volume of surgical in-
tervention is planned as a non-alternative method of
therapy [3, 36, 37]. The surgery aims to restore the
anatomical relationships in the lumbosacral spine,
and therefore in the entire locomotor chain of the
spine–pelvis–lower limbs, to decompress the cauda
equina roots, relieve pain, and stabilize the affected
section with the formation of a bone block [2, 5].

It should be especially noted that, according to
most experts, the issues of surgical treatment of
spondyloptosis remain debatable [7, 38, 39]. Thus,
the algorithm for choosing the method of surgical
intervention has not yet been formulated; the need
for and degree and methods of reduction of the dis-
placed L₄ vertebra remain debatable; the extent of
the spondylodesis zone has not been scientifically
substantiated; measures to prevent the occurrence
or aggravation of neurological disorders, including
through neurophysiological monitoring, have not
been developed, and there are no generally accep-
ted clinical and radiation criteria for evaluating the
treatment results [6, 26, 40–44]. To date, the set of
surgical technologies in the treatment of spondy-
loptosis varies from in situ fixation [37, 43] to 360°
reconstruction with changes in the parameters of the
lumbopelvic balance [45, 46].

According to the literature, the first surgeon
who operated on a patient with spondyloptosis was
J.A. Jenkins. This English doctor in 1936 used an-
terior spinal fusion according to the Berns method
in the surgical treatment of a 16-year-old adolescent
with spondyloptosis [47]. Since the 1960s, in the
surgical treatment of high-grade spondylolisthesis,
the L.L. Wiltse spinal fusion with autobone in situ
started to be performed [48].

Dissatisfaction in the treatment results of spon-
dyloptosis, primarily the failure of spondylodesis,
prompted the American surgeon R.W. Gaines to
develop a surgical technique that consists of total
vertebral body L₄ resection from the anterior ex-
traperitoneal approach (stage 1), reduction of the
vertebra L₄⁻⁻ and posterior L₄⁻⁻S₁ spondylodesis
(stage 2), followed by immobilization of the spine
with a corset until the formation of a bone–metal
block [49]. For 25 years, by 2005, the author had
performed surgery on 30 patients using the tech-
nique he developed [50].

Later, K. Kalra et al. modified the Gaines sur-
gery, starting to resect only the lower part of the
L₄ vertebral body [51]. In Russia, the positive expe-
rience of treating spondyloptosis in an 11-year-old
patient using the Gaines surgery was presented by
staff of the N.N. Priorov Central Institute of Trau-
matology and Orthopedics [52].

With the introduction of trapedicular fusion
(CD-instrumentation) into vertebrological prac-
tice, this technology was also applied in the sur-
surgical treatment of spondyloptosis [53, 54]. Along with traditional variant of transpedicular fusion, A.A. Afaunov et al., in the treatment of spondyloptosis in a 22-year-old patient, successfully used an apparatus for external transpedicular fixation and gradual and dosed reduction, within 34 days, of the LV vertebral displacement into the pelvic cavity. After successful restoration of anatomical relationships at the L₅–S₁ spinal motion segment level, the patient underwent a submerged stage of transpedicular fusion using a 6 polyaxial screw hardware. The final stage of treatment was an anterior corporal fusion using a 6 polyaxial screw hardware. The authors of the publication expressed their opinion that pedicle screws could definitely be prescribed. The authors of the publication expressed their opinion that pedicle screws could definitely be prescribed. The authors of the publication expressed their opinion that pedicle screws could definitely be prescribed. The authors of the publication expressed their opinion that pedicle screws could definitely be prescribed.

According to S.O. Ryabykh et al., the use of the 360° reconstruction technique with removal of the arch of the vicious vertebra L₅ and meninogaradicularulosis of the vertebra S₁ leading to a wide release in the scope of bone-disk-bone osteotomy at the level of L₅–S₁ and a change in the sacrum tilt angle was a key factor in achieving mobilization and radical correction of parameters of the lumbo-pelvic balance in severe forms of spondylolisthesis in pediatric patients [46]. Moreover, when performing circular spondylodesis with reduction, the risk of neurological complications increases significantly [59, 60].

Alternatively, an extended scope of surgical intervention on the spinal column structures, especially in patients of younger age groups, is fraught with the risk of vertebrae growth retardation of pediatric patients [61]. Literature data refute these judgments. Therefore, M. Ruf et al. retrospectively evaluated 19 clinical cases of the use of 91 pedicle screws in children aged 1–2 years, who underwent surgery for various diseases of the spine. In the long-term, one patient had a breakdown of one of the screws, and two patients had screw connection violations. The authors concluded that the use of pedicle screws is often the only way to securely fix the spine structures, and their installation does not affect the growth of the vertebrae [62].

J. Li et al., who implanted 74 pedicle screws in 16 pediatric patients aged 1–4 years, agreed with these conclusions. Postoperative CT scan showed inadequate placement of 5 (6.75%) screws, while medial malposition was not recorded in any of the cases. The long-term results of surgical treatment after 3 to 7 years were studied in 7 patients. In all clinical cases, normal shape and size of the vertebrae were recorded [63].

A. Ranade et al. report the experience of installing 88 pedicle screws with a diameter of 3.5–5.5 mm for the thoracic vertebrae and those with a diameter of 4–6 mm for the lumbar vertebrae in 16 pediatric patients under 8 years of age. In 6 (6.81%) cases, postoperative CT revealed malposition of the hardware, which was medial in one of the cases described. The authors of the publication expressed the opinion that pedicle screws could definitely be installed in the smallest children [64].

In support of the above, J. Stulik et al. reported on the safe possibility of inserting a pair of screws into the odontoid process of the C₂ vertebra in pediatric patients of one year of age [65].

An analysis of the literature on spondylolisthesis in pediatric patients shows that this pathology can be diagnosed in children during the first years of life [1, 2, 66–68]. In these cases, it should be remembered that, according to J. Dubusset, surgical interventions can lead to catastrophic consequences, since they can cause an impairment of the balanced growth of the immature spine and its surrounding structures. The author is convinced that, in modern pediatric vertebrology, the main question is when and how the surgical treatment of actively growing children with progressive spinal deformities of various etiologies should be started [69].

A study of the isolated results of treatment of pediatric patients with high-grade spondylolisthe-
sion shows, and it is paradoxical, that delayed surgery, as a rule, does not lead to serious changes in their quality of life [59]. Therefore, Canadian researchers E. Bourassa-Moreau et al. analyzed the results of treatment of 34 pediatric patients, 29 of which were operated on for high-grade spondylolisthesis. During the case follow-up, the Scoliosis Research Society (SRS)-22 questionnaire was used. The analysis results showed that the quality of life of patients operated on the spine and those treated conservatively did not differ significantly [70].

Chinese authors X. Xue et al. agreed with these conclusions and used the Newcastle–Ottawa Scale (NOS) in a similar study in the same category of patients. Furthermore, in the groups of operated and non-operated children, there was no statistically significant difference in the evaluation of the NOS scale criteria [71]. Children and adolescents, who benefit most from surgical treatment of severe spondylolisthesis, have lower baseline health-related quality of life [72].

Treatment methods used in pediatric operative vertebrology “follow” the development of general vertebral surgery [69]. Recently, after the widespread introduction of computer and robotic technologies into clinical medicine, successful attempts have been made to develop individual 3D structures used in pediatric practice [73–75]. Therefore, M.A. Gerasimenko et al. demonstrated in their article the first positive experience of 3D design and prototyping in the surgical treatment of multifocal spinal deformity formed in the presence of the posterior sphenoid hemivertebra L3 in a 6-year-old girl [76].

American authors J. Parthasarathy et al. in the surgical treatment of spondylolisthesis in an adolescent, used composite models of fixators manufactured individually on a 3D printer, designed taking into account the peculiarities of the spinal–pelvic balance of a particular patient. In their article, the authors describe the technology of the workflow for the manufacture of such products and illustrate the aspects of their use in clinical practice [77].

Conclusion
Spondylolisthesis in children, adolescents, and young people is a disorder whose relevance is determined primarily by the unresolved issues of the treatment approach. The treatment should clearly be surgical. Additionally, there is poor consensus regarding the timing and volume of surgical interventions recently.

Normalization of the disturbed vertebral–pelvic balance in children due to spondylolisthesis is the most important component of the ongoing treatment regime, and it can be achieved only by 360° reconstruction of the lumbar spine and sacrum. According to most authors, advancement in medical technologies will enable us to achieve superior results using surgical treatment in children with spondylolisthesis, with minimum risk of neurological complications in the restored anatomy of the spine, pelvis, and lower extremities, and their prospective analysis will qualitatively change the evidence and strength of recommendations for the treatment approach.

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REFERENCES
10.1097/00007632-200209150-00010.


52. A clinical case for discussion presented by the staff of the Department of Spinal Pathology of the Federal State Institution “CITO named after N.N. Priorov of Rosmedtekhnologi”. http://spineinfo.ru/infosources/case/cases_19.html (access date: 10.06.2021). (In Russ.)
75. Burnard JL, Parr WCH, Choy WJ, Walsh WR, Mobbs RJ. 3D-printed spine surgery implants: a systematic review of the efficacy and clinical safety profile of


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