DOI: 10.17816/KMJ2022-241

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_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_V–S_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_V spondyloptosis in children and adolescents require further study.

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

For citation: Skryabin EG. Spondyloptosis in children, adolescents and youth age patients. *Kazan Medical Journal.* 2022; 103(2):241–249. DOI: 10.17816/KMJ2022-241.

Background

Lumvar V vertebra spondyloptosis is a rare nosological form of vertebrogenic pathology in children, adolescents, and young people [1, 2]. According A.M. Lak et al., who published their article in 2020, to date, the prevalence of spondyloptosis is unknown in both the pediatric and adult population [3].

Data presented in modern medical literature on spondyloptosis frequency in growing patients is unsystematized and reflects the personal experience of individual authors in the treatment of this category of patients. Thus, the minimum incidence of spondyloptosis among children and adolescents with spondylolisthesis of the lower lumbar vertebrae was presented by V.V. Platunov et al. (0.9% of clinical cases, in 2 out of 218 treated patients) [4]. The maximum frequency of spondyloptosis diagnosed in a group of children with spondylolisthesis was announced by S.V. Vissarionov et al. (15.38% of cases, in 4 out of 26 pediatric patients operated by the authors) [5]. The average indicators of spondyloptosis diagnosis relative to the above are presented by V.V. Krutko et al. and M. Rivollier et al. (4.76% and 14.28% of clinical cases, respectively) [6, 7].

Materials and methods

Research articles for literature review were obtained from modern electronic medical databases, such as PubMed, CYBERLENINKA, eLIBRARY, and Google Scholar. Considering the small number of research articles on the topic under discussion, we considered literary sources from 1995 to 2021. Furthermore, the literature search provided links to four articles published in the 1950–1970s, and it is difficult to present contemporary information on Lv vertebra spondyloptosis without mentioning them. The search for literary sources was performed using keywords in Russian and English languages, namely spondyloptosis, high-grade spondylolisthesis, children, and adolescents.

Results and discussion

According to the literature, H. Junge and P. Kuhl were the first to distinguish spondyloptosis as

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Received 25.06.2021; accepted 29.09.2021; published 12.04.2022.

a separate, most severe, degree V spondyloptosis 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 spondylolisthesis 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, spondyloptosis 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 spondyloptosis [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 L_V vertebral arch [14].

In addition to disorders of the spinal–pelvic balance in children with spondyloptosis, 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 spondylolysis of the interarticular part of the arches of the vertebrae L_{II}, L_{III}, and L_{IV}, spondylolisthesis of the L_{IV} vertebra in a female adolescent with established spondyloptosis [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 spondyloptosis 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_I. Spondylolysis lines of the interarticular part of the L_V 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 spondyloptosis, 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_I 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 spondylolysis 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 spondyloptosis, 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_V 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, "apperta" and "occulta," were identified. Moreover,*spina bifida posterior* defects of individual vertebrae, cases of non-closure of the *hiatus*

sacralis totalis were diagnosed in 15 (23.8%) of the studied cohort, as well as anomalies in the number. The latter were represented by lumbarization of the S_I vertebra (11 pediatric patients, 17.46%) and sacralization of the L_V vertebra (4 pediatric patients, 6.34%) [24].

Objective confirmation of spondylolisthesis and spondyloptosis in pediatric patients is based on plain radiography of the lumbar spine and sacrum. This position is presented in the report of the Committee for Evidence-Based Medicine of the Scoliosis Research Society (SRS), in the section on methods for diagnosing lumbar spondylolisthesis in pediatric patients [25]. For the diagnosis and case follow-up of the pathological process, the radiograph in the sagittal (lateral) view is the most informative [26]. To establish the nature of disorders of the sagittal spinal–pelvic balance, radiography of the entire spine and pelvis with hip joints in the anteroposterior and lateral views in the patient's upright position is required [27, 28].

The normal sagittal balance is considered as a situation when, on a lateral radiograph, a vertical plumb line lowered from the middle of the C_{VII} vertebral body "passes" along the anterior superior angle of the S_I vertebra, which is the so-called *sagittal vertical axis* [29]. A variant of the norm can be a situation when the vertical plumb line in the sacrum is displaced anteriorly or posteriorly, but not by more than 2 cm in each direction [30]. Exceeding this value indicates a positive (with an anterior displacement) or negative (with a posterior displacement) sagittal balance [31]. There is a reasonable opinion that the state of the sagittal balance can only be reliably determined when assessing the radiograph of the head, trunk, pelvis, hips, legs, and feet in the lateral view, with the patient in the upright position [32].

Computed tomography (CT) and magnetic resonance imaging (MRI) performed in patients with spondyloptosis can significantly detail the nature of the pathology [33]. According to French researchers R. Vialle et al., CT enables to assess most accurately the main quantitative characteristics of the spinal–pelvic relationship of patients in the upright position and to develop the most rational approach for the surgical treatment of spondyloptosis [34]. MRI results are required to assess the anatomical changes in the soft tissue ligamentous apparatus of the lumbosacral spine, including intervertebral disks, the degree of compression of the ducal sac, and the elements of the cauda equina and segmental roots located in it [35].

Depending on the nature and degree of the diagnosed disorders in each individual child with spondyloptosis, an individual volume of surgical intervention 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 displaced L_V 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 accepted clinical and radiation criteria for evaluating the treatment results [6, 26, 40–44]. To date, the set of surgical technologies in the treatment of spondyloptosis 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 anterior 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 spondyloptosis, primarily the failure of spondylodesis, prompted the American surgeon R.W. Gaines to develop a surgical technique that consists of total vertebral body L_V resection from the anterior extraperitoneal approach (stage 1), reduction of the vertebra L_{IV}, and posterior L_{IV}–S_I 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 technique he developed [50].

Later, K. Kalra et al. modified the Gaines surgery, starting to resect only the lower part of the L_V vertebral body [51]. In Russia, the positive experience 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 Traumatology and Orthopedics [52].

With the introduction of transpedicular fusion (CD-instrumentation) into vertebrological practice, this technology was also applied in the sur-

gical 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 L_V vertebra displaced into the pelvic cavity. After successful restoration of anatomical relationships at the L_V–S_I 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 corporodesis at the L_V–S_I segment level with an autograft from the left iliac wing, which enabled 360° stabilization [55].

S.V. Vissarionov et al., in the treatment of pediatric patients with grade III–IV spondylolisthesis and spondyloptosis, successfully used one-stage surgical intervention from the posterior approach under neurophysiological intraoperative control. The surgery scope, according to the authors, consisted in laminectomy of the L_V vertebra, revision of the spinal canal and radiculolysis, restoration of the sagittal balance of the spine by reduction of the L_V vertebral body, removal of the degeneratively altered intervertebыral disk L_V–S_I, and stabilization with surgical hardware and autologous bone of the lumbosacral spine in a physiologically correct position [5, 56].

When reviewing recent scientific publications on surgical technologies inspondyloptosis treatment of growing patients, it is noteworthy that most authors prefer circular spondylodesis as the most effective method [3, 33, 36, 57]. For example, W. Molinari et al. analyzed the results of the surgical treatment of high-grade spondylolisthesis in 37 pediatric patients, depending on the surgical technique, namely L_{IV}–S_I spondylodesis *in situ* (18 patients) and 360° reconstruction (19 patients). When studying the long-term results, it turned out that, pseudarthrosis was registered only in patients with a history of posterior spondylodesis; only in 7 (38.88%) clinical cases. Revision surgeries performed in these patients in the scope of 360° reconstruction enabled to achieve adequate circular spondylodesis in all cases [58].

According to S.O. Ryabykh et al., the use of the 360° reconstruction technique with removal of the arch of the vicious vertebra L_V and meningoradiculolysis of the vertebra S_I, leading to a wide release in the scope of bone-disk-bone osteotomy at the level of L_V–S_I, and a change in the sacrum tilt angle was a key factor in achieving mobilization and radical correction of parameters of the lumbopelvic 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 C_{II} vertebra in pediatric patients of one year of age [65].

An analysis of the literature on spondyloptosis 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-

sis 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 Nawcastle–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 multiplanar spinal deformity formed in the prыesence of the posterior sphenoid hemivertebra L_I in a 6-year-old girl [76].

American authors J. Parthasarathy et al. in the surgical treatment of spondyloptosis 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

Spondyloptosis 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 spondyloptosis is the most important component of the ongoing treatment regime, and it can be achieved only by 360º 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 spondyloptosis, 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.

Funding. The study had no external funding.

Conflict of interest. The authors declare no conflict of interest.

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