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Review article

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Sacralization of the L5 vertebra (Bertolotti syndrome): literature review

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Abstract

Introduction The resultant cause of pain syndrome in the lumbosacral spine in the contemporary population are dysplasia and anomalies of the development of vertebral segments, among which the sacralization of the L5 vertebra (Bertolotti syndrome) is often diagnosed. In the domestic medical literature, few publications on this pathology have been found. **Methods** Scientific studies for this literature review were obtained from PubMed, eLIBRARY, CYBERLENINKA. Eighty scientific articles were used. The search for literature sources was carried out with the following keywords: sacralization of the L5 vertebra, Bertolotti Syndrome.

Results and discussion In the course of the analysis, it was established that the rate of diagnosis of sacralization of the L5 vertebra ranges from 4 to 36 % of cases. It is equally detected among males and females. This pathology is frequently found in children and adolescents. Pain in the lumbar spine and sacrum is the leading clinical symptom of the disease. The severity of pain on the visual analogue scale mainly corresponds to 5 points. Scoliosis, spondylolisthesis, herniated disks, spina bifida posterior may exacerbate the clinical symptoms of the associated sacralization of the L5 vertebra. During the examination of patients, plain radiography, computed tomography and magnetic resonance imaging are used. Cases of sacralization of the L5 vertebra are divided into 7 types, according to the classification of A.E. Castellvi et al. (1984). Treatment of vertebrogenic pain syndrome is carried out both conservatively (drug therapy, therapeutic blockades) and surgically (resection of the L5 transverse process, decompression of the stenotic intervertebral foramina, transpedicular fusion, radiofrequency ablation).

Keywords: sacralization, L5 vertebra, Bertolotti syndrome, literature review

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INTRODUCTION

The lumbar spine and sacrum are the anatomical areas of the body that patients of various age groups complain of discomfort most frequently [1]. In the structure of their complaints, the incidence of pain in the lumbosacral area takes one of the first places [2]. It turns out that pain most often is caused by dysplasia and developmental anomalies in the spinal motion segments, among which sacralization of the L5 vertebra is frequently [3, 4]. In the domestic medical literature, there are few publications devoted to this particular pathology. If it is mentioned in the articles, then along with other types of dysplasia and anomalies

in the development of the lumbosacral area, only the incidence of its diagnosis is stated. Much more has been published in foreign studies devoted to various aspects of the sacralization of the L5 vertebra [5, 6]. Given, on the one hand, the obvious lack of domestic scientific publications on the sacralization of the L5 vertebra, and, on the other hand, the significant frequency of diagnosing this vertebral anomaly in the population, we decided to review the literature on this topic.

The **purpose** of the work is to analyze the main domestic and foreign publications on the sacralization of the L5 vertebra.

MATERIALS AND METHODS

Scientific publications for writing a literature review on the topic "Sacralization of the L5 vertebra (Bertolotti syndrome): a literature review" were obtained by searching for them in the PubMed database, electronic libraries eLIBRARY, CYBERLENINKA. In total, 80 scientific articles were studied and worked out, which

reflect the most pressing problems and aspects of the topic under this study. Domestic literary sources were seven 7 (8.25 %), and foreign ones were 73 (91.25 %). Literature sources were searched for with the following keywords: sacralization of the L5 vertebra (L5 vertebra sacralization), Bertolotti Syndrome.

RESULTS AND DISCUSSION

Ulrich et al. defined the term "sacralization" as "lumbosacral dysplasia which results in the acquisition by the lower lumbar vertebra of the signs characteristic of the sacral vertebrae: uni- and bilateral synchondrosis of the transverse process of the L5 vertebra with the iliac wing or complete fusion of L5 body with S1 body" [7].

This pathology was first described by the Italian scientist Mario Bertolotti in 1917, and since that time this variant of dysplasia bears his name, Bertolotti syndrome, in the English medical literature [8].

Sacralization of the L5 vertebra and lumbarization of the S1 vertebra are referred to dysplasia of the

lumbosacral transitional vertebrae (LSTV) that is laid in utero as a result of the exposure of a pregnant woman and fetus to a large number of damaging endogenous and exogenous factors [9, 10]. It is under the influence of these factors that disturbances occur in the formation, migration, differentiation, and segmentation of somite sclerotomes [11].

If the minimum rate of diagnosing sacralization of the L5 vertebra is 4 %, as determined by almost all authors [12, 13], the maximum rate of its occurrence reported by current researchers is varying. Thus, Ugokwe et al. reported that the highest rate of detection of this pathology is 8 % of clinical cases [14]. Approximately twice as often sacralization was diagnosed by Sharma et al. and Ucar et al., 14.1 % and 17.2 %, respectively [15, 16]. Bulut et al. analyzed 500 radiographs of the lumbosacral spine and revealed sacralization of the L5 vertebra in 21.2 % of the cases examined [17]. Ravikanth et al. diagnosed a similar pathology in 26.8 % of cases [18]. Finnish authors, Hanhivaara et al., conducted a vertebral analysis of 3,855 computed tomograms of the abdominal cavity of patients, diagnosing radiological symptoms indicating the presence of sacralization in 29.0 % of patients [19]. The maximum rate of diagnosing this pathology among patients with lumbar pain was reported by Adams et al. and Crane et al. According to the first group of authors, the incidence of this variant of lumbosacral dysplasia among the population was 35 % of cases [20], according to the second group of researchers, it was up to 36 % of clinical cases [21].

There are no gender differences in the incidence of sacralization, according to Indian authors Ravikanth et al., who studied 500 radiographs of the lumbar spine and sacrum [18]. However, Japanese researchers, Sugiura et al., provide information about the predominance of this pathology in females [10], and an international group of scientists from Mexico and the United States confirms this information reporting 60 % in a cohort of patients with sacralization of the L5 vertebra [22]. The opposite opinion is held by Jancuska et al., who reported that the incidence of sacralization among males reaches 28 % of cases, and among females 11 % of clinical cases [23].

Due to the fact that the sacralization of the L5 vertebra is a congenital pathology, it seems natural that it is often diagnosed in children and adolescents examined for vertebrogenic pain syndrome [24, 25, 26, 27]. Nevertheless, the main category of patients in whom this developmental anomaly is detected is people of mature and old age [9, 17].

It is the pain syndrome in the lumbar spine and sacrum that is the leading clinical symptom of sacralization of the L5 vertebra [28]. Pain is aggravated by prolonged standing, lifting and carrying heavy objects, turning and bending the torso [12, 29]. Pain

is frequently accompanied by numbness, paresthesias in the lumbosacral region, pain on palpation in the projection of the spinous processes of the lower lumbar vertebrae and along the crest of the sacrum [30].

The initiation and progression of pain in patients with sacralization is primarily due to the disorder of the normal anatomy and biomechanics in the lumbosacral spine [3, 31]. Further on, due to the emerging (or formed) “pseudarthrosis” between the transverse processes of the L5 vertebra and the wings of the iliac bones (or the lateral masses of the sacrum), the phenomena of osteochondrosis, spondylarthrosis and spondylosis, stenosis of the intervertebral foramen develop, and conditions are created for compression of the extraforaminal section of the nerve root L5 [23, 32]. Similar, more or less pronounced pathomorphological changes also occur in the spinal motion segments overlying the L5 vertebra, what exacerbates clinical symptoms [5, 29].

Pain may radiate to one of the sacroiliac joints, to one of the inguinal regions, to one of the lower extremities [21, 33]. The severity of the pain syndrome, including during treatment and in the study of long-term results, is determined using a visual analogue scale [34, 35, 36, 37]. Prior to the start of treatment procedures, the severity of pain syndrome assessed with VAS in patients with sacralization of the L5 vertebra corresponds, on average, to 5.2-5.5 points [18, 30, 35], but can “reach” an 8-point value [6].

It should be noted that sacralization of the L5 vertebra is not accompanied by pain in all cases. An interesting study in this regard was conducted by Turkish authors, Bulut et al., who studied 500 radiographs of patients with back pain (main group) and 500 radiographs of patients without back pain (control group). It turned out that various types of sacralization in the main group were diagnosed in 21.2 % of cases while in the control group they were encountered in 16.8 % of clinical cases [17]. Information that this pathology is often detected occasionally during a radiological examination of the spine, for example, due to injuries, was also reported by other authors [38, 39].

Other nosological forms of vertebrogenic pathology, such as scoliosis [13], spondylolytic spondylolisthesis [40], osteochondrosis in the stage of sequestration of the intervertebral disc [41], non-union of the posterior part of the vertebral arches (spina bifida posterior) [42] that accompany sacralization of the L5 vertebra aggravate clinical symptoms.

Data published in the literature on the incidence of multiplanar scoliotic spinal deformity in this category of patients are contradictory. Most of the authors inform readers that the frequency of diagnosing scoliosis in patients with sacralization is 6-18 % of clinical cases [43, 44]. However, Jain et al. report that they have experience in conservative and surgical treatment

of 20 patients with Bertolotti syndrome, and all their patients, 100 % of cases, had clinical and radiological symptoms of lumbar scoliosis [37].

The rate of diagnosing spondylolytic spondylolisthesis of the L4 vertebra in patients with sacralization can reach 60 % of clinical cases [45]. According to Kim et al., the displacement of the L4 vertebra is more pronounced than in patients who did not have dysplasia and anomalies in the development of the lumbosacral area [46]. Their conclusion is confirmed by the results of studies by a group of Chinese scientists [47]. Of interest is the publication of Russian authors who revealed sacralization of the L5 vertebra in 6.3 % of children and adolescents with a diagnosis of high grade spondylolisthesis [48]. It is paradoxical, but true. In regard to such pathology as degenerative spondylolisthesis, most authors do not consider sacralization as a probable etiological mechanism of this disease [49].

Due to the fact that sacralization is frequently characterized by concretion of the L5-S1 spinal motion segment, it seems obvious that there is a high probability of excessive mobility at the higher levels that may result, over time, in the formation of an intervertebral hernia [32, 36]. Jin et al. conducted a targeted study in 200 patients with Bertolotti syndrome (main group) and 200 patients who did not have radiographic symptoms of this developmental anomaly (control group). The aim of the study was to determine the occurrence of intervertebral hernias in the studied cohorts. It turned out that the rate of diagnosing intervertebral hernias was 71.5 % in the main group and 34.0 % in the control group. Moreover, the intervertebral disc L4-L5 was most often affected [9]. This predominant level of intervertebral hernia was confirmed by the publication of Zhang et al. According to these Chinese authors, the L4-L5 intervertebral disc is affected by a degenerative process in 81.3 % of cases, followed by the formation of a sequestered hernia at this level, while the L5-S1 intervertebral disc undergoes similar changes in 18.7 % of cases [25]. The research of Ahn et al. showed that patients with herniated intervertebral discs resulting from Bertolotti syndrome are more prone to recurrence of herniated protrusions after microdiscectomy than those patients who do not have anomalies of the lumbosacral area [50].

A group of Dutch researchers led by J.L. Bron emphasizes that degenerative changes in the intervertebral discs are recorded at a younger age in patients with sacralization of the L5 vertebra, but this difference disappears as the patients grow older, "masked" by other involutive processes in the spine [51]. These include stenosis of the spinal canal at the lumbar level [52] and abnormal tropism of the lower lumbar spinal motion segments [53], which exacerbate the clinical symptoms of sacralization [54].

Many authors that study the Bertolotti syndrome indicate that this developmental anomaly is frequently combined with defects in the posterior parts of the bodies of the lower lumbar and upper sacral vertebrae, spina bifida posterior [55] and fixed spinal cord syndrome [56]. Turkish authors, Fidan et al., studied radiographs of the lumbosacral spine in 3,132 young men aged 17 to 22 years. They revealed that 28.3 % of them had, as the authors indicate, "asymptomatic spinal anomalies". The structure of these anomalies was dominated by variants of spina bifida posterior in 16.2 % of cases and transitional lumbosacral vertebrae in 12.9 % of clinical cases [57]. In the cohort of patients with fixed spinal cord syndrome, various types of sacralization of the L5 vertebra were diagnosed in 63 % of patients [58].

The leading method of objective diagnostics of sacralization of the L5 vertebra is radiographic examination of the spine and sacrum [59]. For the examination of patients, depending on the level of the medical institution, plain radiography [9, 25], computer [5, 19] tomography and magnetic resonance imaging [60, 61] are used. These methods of radiological diagnostics are used both separately and in combination, in order to choose an approach to the planned therapy, including indications for surgical treatment [28, 37].

Plain radiography of the spine and sacrum is performed to obtain an overview of the state of this anatomical region, as standard radiographs in the anterior-posterior projection do not always provide information about the type of sacralization of the L5 vertebra [61, 62]. In these cases, it is preferable to perform radiography of the lumbosacral spine in the anterior-posterior projection at a cranial angle of 30° (Fergusson projection) [59]. The results of multislice computed tomography are more demonstrative, and on their basis, as a rule, a conclusion is made about the type of sacralization of the L5 vertebra [42, 62]. The use of magnetic resonance imaging for diagnostic purposes provides reliable information on the presence and severity of extraforaminal stenosis, impingement, and edema of the L5 spinal nerve root [60], and in some cases, the L4 and S1 spinal nerve roots [63, 64].

The use of positron emission tomography for diagnosing Bertolotti syndrome was reported in several publications by foreign authors [65].

The cases of sacralization of the L5 vertebra diagnosed with the help of radiological imaging methods are classified according to the recommendations of Castellvi et al. The classification developed by these American authors in 1984 distinguishes seven types (types I, II, III, in turn are subdivided into subtypes "a" and "b" and IV) of sacralization of the L5 vertebra and focuses on the degree and nature of the concretion of the transverse process (processes) of this vertebra with the iliac wing (unilateral or bilateral) or the lateral mass(s)

of the sacrum [66]. Modern authors present different rates of occurrence of the types of sacralization of the L5 vertebra in the cohorts studied by them. Thus, Hanhivaara et al. inform readers that type I was diagnosed in 68 % of cases, type II in 16 %, type III in 13 %, type IV in 3 % of clinical cases [29]. Chinese researchers Yao et al., who studied transitional lumbosacral vertebrae in patients with herniated intervertebral discs and spondylolisthesis, do not report on the incidence of types I or IV, while type II was diagnosed in 45.92 %, type III in 54.08 % of cases [47].

For practitioners, the published information that types Ia, IIa and IIIa, that are cases of unilateral sacralization, is valuable as they have more pronounced clinical symptoms [13], while pseudoarthrosis and concretions are more often localized on the left side [23]. The study of McGratha et al. found that the rate of diagnosing the left-sided forms of sacralization of the L5 vertebra is 48.03 % of cases, of the right-sided ones is 23.54 %, and bilateral are 28.43 % of clinical observations [5].

Despite the fact that the classification of Castellvi et al. was accepted as a guide to action by all interested specialists, research in the development of new questionnaires, scales and classifications continues. Thus, Knopf et al. presented their own four-point Onyike Grading Scale based on the assessment of location, severity and characteristics of pain in patients with sacralization of the L5 vertebra [67].

The analysis of current scientific publications devoted to various aspects of the sacralization of the L5 vertebra shows that the most important problem is errors in clinical diagnosis, and they are associated mainly with incorrect numbering of the vertebrae [68]. Many authors state with regret that patients with lumbodinia rarely have radiographs of the entire spine; most often, only the results of an X-ray examination of the lumbar spine are available for evaluation [69]. In such cases, difficulties often arise with the numbering of the vertebrae, since it is not always possible to accurately differentiate, for example, hypoplasia of the ribs from the transverse processes in the thoracolumbar junction [59]. In these cases, such anatomical formations as the iliac wing [70], iliopsoas ligament [71], right renal artery, superior mesenteric artery, aortic bifurcation, medullary cone [72] should serve as landmarks. However, the listed anatomical formations are unstable, subject to individual variability, which can lead to frequent diagnostic and then therapeutic errors [44]. In clinical practice, such errors occur with a frequency of 33 % [5] to 54 % [39]. According to the unanimous opinion of all authors involved in the study of the sacralization of the L5 vertebra, there is no reliable way to count the vertebrae without analyzing the results of high-quality imaging of the entire spinal column, starting from the level of the C2 vertebra and caudally, down to and including the sacrum [59, 69, 70]. It is this,

the most reliable method of numbering vertebrae that enables to determine the type of sacralization of the L5 vertebra and develop a treatment strategy aimed at relieving vertebrogenic pain syndrome [38, 73].

The treatment begins, as a rule, with conservative methods, which consist in prescribing physiotherapeutic procedures and drug therapy with analgesics and anti-inflammatory drugs [21, 28]. If the therapeutic effect is insufficient, local blockades are performed in the projection of the articulation between the transverse process (processes) of the L5 vertebra and the lateral mass (masses) of the sacrum [35] and/or transforaminal epidural injections of drugs from the group of glucocorticosteroids are performed [34]. The clinical results of conservative therapy are usually assessed using VAS: a reduction of 50 % from the baseline or a score of less than 3 after injection is considered adequate pain relief [37]. In some cases, the short-term positive effect obtained from local anesthesia is considered as a criterion for surgical intervention at the level of the lumbosacral junction [74].

The most common and widely used operation that satisfies patients is pseudoarthroectomy or resection of the enlarged transverse process(s) of the L5 vertebra from the posterior approach [6, 75]. This operation is used for sacralization of types I and II according to the classification of Castellvi et al., including in adolescent patients [26, 27]. A team of Australian surgeons used a retroperitoneal approach to resect the transverse process of the L5 vertebra, emphasizing that they had not previously encountered similar approaches to the pseudarthrosis of L5-S1 described in the literature [76].

Minimally invasive surgery using endoscopic instrumentation and intraoperative 3D navigation has also found application in the treatment of clinical manifestations of Bertolotti's syndrome [74, 77, 78]. The advantage of these operations is that they allow decompression of stenotic intervertebral foramina of the L5-S1 spinal motion segment with a good therapeutic effect [6, 21].

Transpedicular fusion has not been widely used, but is also used as a method of surgical intervention even in types III and IV of Bertolotti's syndrome [20]. In the few literature papers on this subject, the question of fusion extension has been primarily discussed in order to preserve the maximum possible movements in the lumbar spine [73, 79].

A number of authors consider impulsive radiofrequency ablation of L5-S1 a promising method of minimally invasive surgery in patients with pain due to Bertolotti's syndrome [32].

When analyzing the literature sources presented in this review, our attention was drawn to an interesting fact. None of the publications outlines measures to prevent the formation of lumbosacral transitional vertebrae, including sacralization of the L5. This is due, in our opinion, to the fact that the studied anomaly

in the development of the lumbosacral area is formed in utero, under the influence of many known and not yet established factors. Clinical researchers, whose study results are presented in this review, face already formed anomalies, the development of which is of a progradient nature. The proposal to conduct a preventive radiographic examination of the lumbosacral spine

in childhood and adolescence, which is proposed by a team of Russian authors [30], is also not aimed at preventing intrauterine development and the formation of sacralization of the L5 vertebra in fetuses and, if seriously discussed, would cause a large number of skeptical questions and comments from specialists in various fields.

CONCLUSION

This literature review, based on a comprehensive analysis of 80 current scientific studies, provides information about the most important aspects of such a congenital pathology of the lumbosacral spine as sacralization of the L5 vertebra (Bertolotti's syndrome). However, some issues of practical interest have not been reflected in the analyzed literature sources. Thus, the dynamic course of sacralization of the L5 related to age has not been studied. It is unclear whether it is possible in a patient with type IIa sacralization to state the presence of the same pathology but already of type IIIa or type

IV after a few years of radiographic assessment. If yes, what factors influence the progradient course of this pathology? Of undoubted interest is information about whether there are clinicoradiological parallels between the severity of pain and the type of sacralization. The literature does not provide information on the presence or absence of family forms of this disease. The unexplored questions lie "on the surface". It is expected that further study of various aspects of sacralization of the L5 vertebra (Bertolotti's syndrome), including at the genetic level, will be continued.

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