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Review

Cervical hemivertebrae: A literature review on the evolution of surgical management and its results

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BACKGROUND: Congenital cervical spinal deformities due to hemivertebrae are rare and serious entities that are difficult to solve because of anatomical obstacles in the cervical region and the lack of treatment experience of such patients.

AIM: To analyze current literature dedicated to the surgical treatment of patients with congenital scoliotic deformities.

MATERIALS AND METHODS: We searched for studies evaluating the results and surgical management of patients with cervical hemivertebrae. The literature search was performed using keywords and similar articles in PubMed, Science Direct, and Google Scholar. The depth of the search was 50 years.

RESULTS: No studies have explored the issue of the surgical management of patients with congenital cervical scoliotic deformities because of the rarity and complexity of treatment. Still, data are sufficient to develop an efficient and safe algorithm.

CONCLUSIONS: Despite the complexity and lack of treatment experience in such patients, modern technologies for perioperative planning and surgical management allow for remarkable improvement of patients' quality of life without serious complications.

Keywords: hemivertebrae; cervical scoliosis; osseous torticollis; children.

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Научный обзор

Полупозвонки шейной локализации: развитие методик лечения и результаты (обзор литературы)

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Обоснование. Врожденная деформация шейного отдела позвоночника при наличии полупозвонков встречается не часто, но представляет трудную для решения проблему в связи с анатомическими особенностями данной области и отсутствием большого опыта лечения таких пациентов у большинства хирургов.

Цель — анализ доступной литературы, посвященной хирургическому лечению пациентов с врожденными сколиотическими деформациями шейного отдела позвоночника.

Материалы и методы. Проведены поиск и анализ публикаций, в которых содержатся результаты обследования и лечения пациентов с полупозвонками шейной локализации. Использовали поисковые запросы по ключевым словам в базах научных данных PubMed, ScienceDirect, Google Scholar, а также осуществляли поиск статей, близких по содержанию. Глубина поиска составила 50 лет.

Результаты. Проблема хирургического лечения пациентов с врожденными деформациями шейного отдела позвоночника освещена в литературе недостаточно ввиду малой частоты встречаемости и высокого уровня сложности лечения. Однако на основании доступных данных возможно определить эффективный и безопасный алгоритм лечения.

Заключение. Несмотря на сложности и малый опыт лечения пациентов данной группы, существующие методики периоперационного планирования и технологии хирургической коррекции позволяют значительно улучшить качество жизни и избежать развития серьезных осложнений.

Ключевые слова: полупозвонок; шейный сколиоз; костная кривошея; дети.

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BACKGROUND

Congenital scoliotic deformity of the cervical spine associated with a hemivertebra is a relatively rare nosological form in comparison with deformities of the thoracic and lumbar regions. As a rule, a hemivertebra is associated with other developmental anomalies, predominantly concrescence of lateral and dorsal structures with hypoplasia of the intervertebral discs in Klippel–Feil [1–3] or Goldenhar [4] syndromes. Patients often have hemivertebrae in the thoracic and/or lumbar regions and developmental anomalies in other organs and systems (cardiovascular and urogenital) [5]. This combination of spinal malformations greatly complicates the choice of stages of surgical treatment in these patients.

The work aimed to analyze the available literature on the surgical treatment of patients with congenital scoliotic deformities of the cervical spine.

MATERIALS AND METHODS

A search and analysis of publications over the past 50 years, reporting the results of the examination and treatment of patients with cervical hemivertebrae, were performed. Keyword search queries were used in PubMed, ScienceDirect, and Google Scholar, and articles with similar content were also included in the literature search.

RESULTS AND DISCUSSION

The major clinical manifestation is a malposition of the head and line of sight, which primarily determines the need for differential diagnostics with muscular torticollis, posterior cranial fossa tumors, and neuromuscular deformity [6]. Pain syndrome and neurological deficits are usually not registered. In some cases, they rarely develop when hypermobility and degenerative changes occur at the level of adjacent segments [7]. Although the natural course of the deformity against this type of defect is difficult to predict because of the low frequency of occurrence, in some cases, the compensatory possibilities for correcting the sight position are insufficient, and the progressive aggravation of torticollis necessitates searching for ways to address this problem. We analyzed deformities of the thoracolumbar spine that develop against a fully segmented hemivertebrae and are characterized by an average rate of progression of 3.5° annually [8]; as a result, we can state with assurance that with the same type of anomaly in the cervical spine, surgical intervention is needed. Conservative treatment using external orthoses and soft tissue interventions that are often erroneously performed are ineffective [9].

The treatment results of patients with this pathology were first reported in 1981 [10]. A. Deburge and J.L. Briard successfully extirpated the posterolateral C_{VII} hemivertebra in a 14-year-old patient with a block vertebra in the cervical region and split posterior structures using a combination of ventral and dorsal approaches to remove the bone structures of the C_{VII} vertebra. Thereafter, the deformity was corrected on the operating table in a halo apparatus against the restoration of the patient's consciousness. At stage 3, instrumental fixation was performed from the ventral approach. Postoperatively, Horner's syndrome developed, which subsequently resolved completely. Moreover, R.B. Winter and J.H. House presented their treatment approach in two cases, where they performed broad decompression, extended metal fixation, and deformity correction without removal of the hemivertebral body because of neurological deficits in the upper limb [11]. M.D. Smith followed a similar methodology, who in 1994 published a study indicating the need for early in situ fixation in patients with congenital cervical deformities and clear signs of progression [12].

For a long time, no studies have reported about this pathology, until M. Ruf and J. Harms presented the treatment results of three patients aged 4, 8, and 14 years treated between 1995 and 2002 [13]. Before surgical treatment, all patients underwent a full examination using multislice computed tomography (MSCT); however, the state and course of the vertebral arteries were assessed by MR angiography. In all patients, a combined (ventral and dorsal) approach was used; the vertebral artery was mobilized by stepwise separation of the bone canal walls of the artery using Kerrison rongeurs. This study described for the first time the coronal plane of resection of the bony parts of the hemivertebra, passing between the ventral surface of the root and the dorsal surface of the vertebral artery. The deformity was corrected by slowly tilting the head toward the apex of the deformity, after which ventral fixation was performed with a plate/wire with screws. In the case of residual kyphosis, it was supplemented with posterior instrumental fixation and spinal fusion. To ensure the safety of neural structures, somatosensory-evoked potentials must be monitored. The mean time to perform surgery was 363 (265–420) min, and the mean blood loss was 617.0 (350.0–1,000.0) mL. Significant correction of local scoliotic deformity (80% on average) was achieved, and the vicious head tilt was eliminated. One patient underwent revision surgery because a screw compressed the C_5 root, which was clinically manifested as weakness of the deltoid muscle. After replacing the screw with a shorter one, the neurological symptoms resolved completely.

M. Ruf presented the treatment results of a patient that was even more difficult to manage: the patient was a 42-year-old woman whose main complaint was severe pain in the projection of the C_2 dermatome on the right [14]. The patient was diagnosed with gross malformation of

the craniovertebral zone presenting as complete assimilation of the C_I vertebra and right-sided semi-segmented hemivertebra C_{II}. After a test foraminal block of the C_2 root, open posterior decompression and instrumental fixation were performed according to the Goel–Harms technique, and the results were satisfactory. However, after 15 months, the patient sought help for local pain in the neck and head malposition; at this time, the last complaint persisted clinically. Initially, the authors planned to resect the hemivertebra only from the dorsal approach; however, after MSCT angiography, which demonstrated a rather close location of the anastomosis of the vertebral arteries to the apex of the odontoid process, two approaches were decided. At stage 1, the apex of the odontoid process, lateral masses, and transverse process of the C_{II} hemivertebra were resected transorally, and the vertebral artery was identified. Since the patient already had a posterior metal fixation, the ventral plate was not required. The authors did not report the cause; however, resection of the arch and C_{II} hemivertebra pedicle with correction and stabilization of the deformity using surgical hardware along C_0–C_{III} was performed only 7 days after stage 2. Excellent clinical and radiological results were achieved with complete correction of the deformity and elimination of the head malposition. The postoperative course was uneventful. The authors of this report emphasized the importance of performing preoperative CT angiography to visualize the vertebral arteries. A combined approach for the resection of a hemivertebra at the level of the craniovertebral zone is a safer option that allows for achieving good clinical results. We found two more papers [15, 16], which described patients aged 15 and 21 years with developmental C_{II} hemivertebra. Both patients complained of neck pain and head malposition. Initial instrumental examination (MSCT and MRI) revealed a malformation of the craniovertebral zone. Conservative treatment with the use of analgesics, muscle relaxants, and exercise therapy was prescribed to both patients, which exerted good clinical effects. Based on the totality of these cases, this developmental anomaly is often left unnoticed for a long time, and head malposition is the major complaint that should be corrected by surgical treatment.

Q. Zhuang et al. [17] represented an interesting clinical case, where a 14-year-old patient complained of progressive spinal deformity, torticollis from birth, and back pain after exercise. Neurological examination revealed signs of myelopathy such as hyperreflexia in the upper and lower extremities and an inverted brachioradial reflex (17 points on the JOA scale). X-ray examination revealed a right-sided scoliotic deformity of 60° in the cervical spine in the presence of three semisegmented hemivertebrae C_{IV}, C_V, and C_{VI}, and a compensatory left-sided thoracic scoliotic curve of 90° was formed. According to the CT angiography data, a distinctive feature was the unique course of the vertebral artery outside the foramina of the transverse processes of all hemivertebrae. The latter circumstance made it possible to resect three hemivertebrae without vertebral artery mobilization. To our knowledge, this is the only case reported in the literature. As in previous cases, a combined approach was used; however, in a different sequence, dorsal elements (arches, articular processes, and vertebral pedicles) were resected from the dorsal approach with the installation of a provisional rod on the hook supporting elements; then, C_{IV}–C_{VI} vertebral bodies were removed from the ventral transverse cervical approach using a high-speed drill, and decortication of the endplates was performed. Thereafter, the provisional rod was removed, and the deformity was gradually corrected by slowly tilting the head to the right shoulder girdle using a contractor installed in front of the adjacent vertebral bodies. The final fixation was performed by placing the anterior cervical plate and the unilaterally located rod dorsally. Intraoperatively, during correction, the spinal cord was controlled by monitoring the motor and somatosensory-evoked potentials. The correction resulted in a decrease in the cervical scoliotic curve to 23°, as well as a regression of neurological symptoms such as the disappearance of hyperreflexia and a pathologically inverted brachioradial reflex. The achieved result was maintained up to 5 years after the intervention. The authors emphasized the need for intraoperative neuromonitoring, use of a provisional rod, and mobilization of the vertebral artery when it is in its typical location.

Since the mobilization of the vertebral artery, as described by M. Ruf and J. Harms, which involved separating it from the bone structures and spinal root, is quite risky and requires great skill and experience from the surgeon, S. Wang et al. proposed a variant of hemivertebra resection, which is mainly characterized by artery mobilization without resection of the bone canal walls, by dissecting the transverse process [18]. The results of this technique are presented in two 12-year-old patients whose hemivertebrae were removed at the C_{III} level. The authors planned the surgery using 3D-printed models of the cervical region, which displayed not only the bone but also vascular (vertebral and internal carotid) formations. The intervention was performed through a combined (ventral–dorsal–ventral) approach, and an ultrasonic bone cutter was used for safer and more precise removal of the transverse process. Other researchers performed intraoperative neuromonitoring (registration of motor- and sensory-evoked potentials) to assess the state of neural structures. The achieved correction and intervention duration (average 300 min) were comparable with the results reported by M. Ruf et al., and the volume of blood loss was significantly lower (average 200.0 mL). No complications (root or artery compression)

associated with a freely located transverse process were reported. In addition to the described technical aspect of hemivertebra resection, the authors emphasized the need to use approaches in the ventral–dorsal–ventral sequence. In their opinion, deformity correction is possible adequately and safely only with the use of a dorsally installed surgical hardware, ideally a screw structure, which is achievable after resection of the hemivertebra body first and then preliminary installation of supporting elements and dorsal structures, because the supportability of the ventral structures of the cervical vertebrae is not sufficient for direct correction. Ventral stabilization is the final and necessary stage.

A group of authors who have, perhaps, the greatest experience in the surgical treatment of patients with cervical and cervicothoracic hemivertebrae used a similar approach. In their earlier work [19], they analyzed the treatment results of 16 patients, where a combination of only ventral and dorsal approaches was used in five cases and a combination of ventral–dorsal–ventral approaches for additional anterior stabilization was applied in 11 cases. The authors deemed that the technique was indicated for a defect in bone structures at the site of vertebrotomy that was performed after deformity correction. They proposed performing the latter by contraction on the convex side of the deformity, assisted by head tilt, fixed in a Mayfield cranial brace. To fill the defect in the osteotomy zone and provide ventral support, a polyetheretherketone (PEEK) cage filled with autologous bone was used in combination with an anterior cervical plate. Perioperative planning was also performed using a three-dimensional plastic model, which, after sterilization, was used in the operating room to clarify the anatomical landmarks for the insertion of transpedicular support elements. Intraoperative neuromonitoring of motor- and somatosensory-evoked potentials was performed to assess the state of neural structures. Despite this, in four patients in the early postoperative period, a transient neurological deficit of a radicular nature was noted, which resolved within 6 months after the intervention. The vertebral artery was mobilized in the manner described by M. Ruf and J. Harms. The correction value of the main arch was 68.5% on average, the duration of intervention was 400 min, and the volume of blood loss was 675.0 mL. The treatment outcome of 12 of 16 patients was rated as “excellent” and “good.” Based on the data obtained, the authors concluded that their algorithm for selecting accesses and instrumental fixation is effective and safe.

The above work was continued in the largest study with a sample of 29 patients. It was performed in 2022 by the same team of authors in the same clinic in Beijing [20]. Moreover, it presents for the first time the results of a completely different surgical technique. Since 2016, to reduce the surgical time and blood loss volume and achieve better cosmetic results, a technique for correcting the deformity by distraction on the concave side was developed and implemented. This was probably partly facilitated by the increasing spread of 3D-printing technology, as individually manufactured implants using additive technologies were installed in some patients, and using the technology of implanting interfacet spacers to eliminate foraminal stenosis of the cervical spine in adult patients. The intervention technique involves the use of a combined (ventral–dorsal–ventral) approach; however, unlike the resection technique, deformity mobility is achieved by anterior release on the concave side, which consists in performing a discapophysisectomy and dissecting the posterior longitudinal ligament from the ventral approach. Then, support elements are installed from the dorsal approach, and segmental distraction is performed under conditions of intraoperative traction in the Mayfield clamp, after which the facet joints on the concave side are disengaged, and decortication and implantation of interfacet spacers (PEEK or titanium) are performed. After confirming that the implant is in the correct and stable position, the rods are stabilized in the supporting elements, and local fusion is performed with an autobone. In stage 3, an interbody implant (also PEEK or titanium) is placed in the ventral defect formed during distraction and fixed with an anterior cervical plate. In this case, the isolation and mobilization of the vertebral artery and root were not performed. Neural structures were monitored by intraoperative neuromonitoring. The authors distributed 29 patients by the type of intervention, where 15 patients underwent resection of the hemivertebra, and distraction of the deformity concave side was used. Comparing the results of these two methods, no statistically significant differences were found in the degree of correction of the main and compensatory arches, magnitude of the change in the angulation of the mandible, and clavicles between the two groups. In addition, the surgical duration (243 and 181 min) and volume of blood loss (342.0 and 123.0 mL) were statistically significantly less in the distraction group. The incidence of neurological complications did not differ significantly; thus, transient C_5 root paresis was noted in three and four patients from the resection and distraction groups, respectively. On average, group 2 needed implantation of more dorsal support elements and prolonged fixation by one more spinal motion segment; thus, differences in the duration of the intervention and volume of blood loss were calculated not in absolute but in relative terms (per one spinal motion segment). Nevertheless, the high efficiency of the proposed method and comparable safety were confirmed. The main advantages of the technique are as follows: resection of bone structures is not needed and isolation of the spinal roots and vertebral artery.

Congenital scoliotic deformity of the cervical spine associated with hemivertebrae is a rare and severe pathology. Compensation mechanisms for the local scoliotic curve are usually limited because of the small length of the uninvolved segment located above the defect zone, which leads to head malposition and a vicious line of sight [13]. As a result, a compensatory arch is formed at the cervicothoracic and upper thoracic spine levels. In the absence of mobility in these segments, in the presence of multiple anomalies, the frontal balance is obviously impaired with the translation of the body to the side. Conservative treatment of such patients is absolutely ineffective, and surgical correction of disfiguring deformities is the method of choice. Previously proposed methods of in situ fixation in combination with wide decompression did not eliminate the cosmetic defect, which is the main complaint of patients [11, 21]. By analogy with the hemivertebrae of the thoracic and lumbar localizations, the hemivertebra must be resected, followed by correction and stabilization with surgical hardware. However, anatomical aspects such as vertebral arteries and functionally significant roots of cervical thickening significantly complicate the treatment. Owing to the same features, resection of a hemivertebra in the cervical region always involves the use of a combined approach, in contrast to surgical interventions in the thoracic and lumbar regions, when it is possible to use only a dorsal approach [22].

In this group, the treatment issue remained undisclosed for a long time, and nowadays, it is not sufficiently covered in the literature. Almost a quarter of a century had passed between the first report by A. Deburge and J.L. Briad [10] about the successful treatment of a patient with a cervical hemivertebra and the work by M. Ruf and J. Harms, which described the resection technique that has become the standard [13]. The total number of patients presented in all articles published on this subject does not exceed 50, which also indicates the rarity of this pathology. Nevertheless, in the last decade, the interest in it has increased, which may be associated with the development of surgical technologies, anesthetic management, intraoperative neuromonitoring, blood-saving technologies, and 3D printing, which greatly facilitates perioperative planning and solves the issue of manufacturing personalized implants.

Based on the results of the literature analysis, several provisions can be distinguished. The volume of the preoperative examination must necessarily include MSCT angiography to visualize the vertebral arteries, namely, their location, direction (tortuous course), and presence or absence of developmental anomalies (hypo- and aplasia), which may affect the choice of surgical treatment. The 3D model enables planning the course of the proposed intervention in detail, and in the operating room, it helps in determining the insertion points and direction of the transpedicular support elements. The choice of an approach depends on the preferences of the operating surgeon; however, according to most authors, the combination of ventral and dorsal approaches with subsequent decisions on ventral support is trustworthy, effective, and safe. The issue of vertebral artery mobilization is the most complex and debatable because this stage involves accurate work with very important and easily vulnerable anatomical formations; therefore, the technique of transverse process mobilization, as proposed by S. Wang et al., appears more attractive [18]. Unfortunately, given the small experience of its use, it is currently impossible to decide unambiguously about its efficiency and safety. The need for intraoperative neuromonitoring during deformity correction is undeniable, which does not completely exclude the development of neurological deficits postoperatively. Fortunately, in all the cases presented, the neurological deficit was transient and was apparently due to nerve root tension and/or swelling. Finally, the extrapolation of surgical technologies used in adult patients has expanded the range of interventions for the correction of congenital scoliotic deformities of the cervical spine in children. The distraction of the concave side of the deformity with the use of interfacet spacers and interbody cages, described by the working group from Beijing, can reduce the intervention time and volume of blood loss and preserve the length of the cervical spine, although results of the implementation of a longer instrumental fixation are needed [20]. In the vast majority of cases, surgical treatment leads to an improvement in the quality of life of patients without the development of serious complications.

CONCLUSION

Thus, congenital scoliosis has an extremely rare pathology, and its correction requires careful planning. In these patients, treatment success can be achieved following the algorithms for choosing access and performing the main stages of surgical intervention. Some proposed methods are promising; however, more experience in application and long-term evaluation of results are needed.

ADDITIONAL INFORMATION

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Author contributions. *N.O. Khusainov* collected the literature data and wrote the text of the article. *D.N. Kokushin* created the concept of scientific work and edited the text of the article. *A.N. Filippova* collected literature data and wrote the text of the article. *S.V. Vissarionov* created the study design and edited the text of the article.

All authors made a significant contribution to the study and preparation of the article and read and approved the final version before its publication.

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