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Lecture

Orthopedic management of cerebral palsy: past, present, and future

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Cerebral palsy is a neurological disease accompanied by orthopedic complications. To date, orthopedic treatment of cerebral palsy remains one of the basic approaches. Knowledge of history is the key to understanding the present and a bridge to the future. It gives us insight into the evolution of understanding of the problem, views on the possibilities of treatment, and personalized approach to the patient. The article describes the evolution of knowledge and approaches of treatment to children with cerebral palsy from ancient times to the present day. The main milestones of the neuro-orthopedic approach to diagnosis and treatment are reflected. In improving the surgical treatment of children with cerebral palsy, it is necessary to rely on a multidisciplinary approach, early detection of orthopedic pathology, personalization, taking into account the psychological and social effect of surgical treatment, and development and improvement of surgical techniques.

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Keywords: cerebral palsy; orthopedic management; history.

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Лекция

Ортопедическое лечение детей с церебральным параличом: прошлое, настоящее и будущее

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Детский церебральный паралич — неврологическое заболевание, сопровождающееся ортопедическими нарушениями и осложнениями. На сегодняшний день ортопедическое лечение детей с церебральным параличом по-прежнему актуально. Знание истории — ключ к пониманию современности и мост в будущее. Оно дает нам представление об изменении понимания проблемы, взглядов на возможности лечения, методов лечения и отношения к пациенту.

В статье описана эволюция знаний и подходов к лечению детей с церебральным параличом от древних времен до сегодняшнего дня. Отражены основные этапы становления нейроортопедического подхода к диагностике и лечению.

В совершенствовании хирургического лечения детей с церебральным параличом необходимо основываться на мультидисциплинарном подходе, раннем выявлении ортопедической патологии, персонализации, учете психологического и социального влияния хирургического лечения и на разработке и совершенствовании хирургических методик.

Лекция была представлена на научно-практической конференции с международным участием «Коррекция двигательных нарушений в комплексе медицинской абилитации детей, больных детским церебральным параличом». Санкт-Петербург, 20–21 ноября 2019 года.

Ключевые слова: детский церебральный паралич; ортопедическое лечение; история вопроса.

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ЭКО•ВЕКТОР

Modern medical science is developing most intensively at the interface of various disciplines and technologies, namely, genetics and mathematics, biology and physics, and neurology and orthopedics, whereas the merger of the latter has given rise to a scientific direction such as neuro-orthopedics.

Cerebral palsy (CP) is a neurological disease accompanied by orthopedic disorders and complications. Nowadays, the possibilities of intervention of the primary source are minor; therefore, orthopedic treatment is still relevant and will maintain to be so in the next decade.

Neuroorthopedics is one of the most interesting fields of contemporary clinical medicine, developing at the intersection of neurology and orthopedics, and it is not limited to CP. The concept of the interaction of the central and peripheral nervous system and the musculoskeletal system as a single system for controlling and implementing movements lays the groundwork for improving our therapeutic capabilities [1].

Knowledge of history is the key to understanding the present and a bridge to the future. It gives us insight into the evolution of understanding the problem, views on treatment possibilities, methods of treatment, and attitude toward the patient.

Diseases that have been known since antiquity include plague, phthisis, cholera, fractures, and scoliosis. CP has been known for the past century and a half. It was not described in the ancient world. However, this does not mean that this disease did not exist. Egyptian mummies are an inexhaustible source of biomedical knowledge. Pharaoh Tutankhamun had a clubfoot, and the ancient Egyptian Pharaoh Septah from the 19th dynasty, who ruled approximately in 1194–1188 BC, suffered from neurogenic deformity of the foot, and this is probably the oldest finding of its kind (Fig. 1) [2].

The word "paralysis" comes from the ancient Greek language. It was used by Galen (AD 130–199) to refer to "weakness and complete or partial necrosis of the nerves of the limbs." He also proposed the term "paresis," which means weakness. Books written by Greek authors (physicians and philosophers) include chapters detailing the neurological manifestations of paresis and paralysis.

Hippocrates (460–390 BC), the founder of modern medicine, drew attention to the connection between prematurity and brain damage. He was the first to mention that "in women who have given birth to lame, blind children, or children with any other defect, the fetus suffered during the month 8 of pregnancy" and that "pregnant women who have a high temperature or have lost too much weight without any for any obvious reason, give birth with difficulty and danger." Thus, even Hippocrates noticed that not only profound prematurity is the cause of profound disability [3].

In the Middle Ages and later, CP did not arouse interest from both theorists and practitioners. Only a few works of art have preserved the images of sick people (Fig. 2). In the Middle Ages, infectious diseases and numerous wars came to the fore.

The scientific study of CP was started in the nineteenth century. The morphological foundations of the problem were

Fig. 1. The mummy of Pharaoh Septah (an ancient Egyptian pharaoh from the 19th dynasty, who ruled in approximately 1194–1188 BC) revealing the oldest finding of a neurogenic deformity of the foot

Fig. 2. A fragment of the painting "Cripples" by Pieter Bruegel, depicting a crippled young man, allegedly suffering from CP

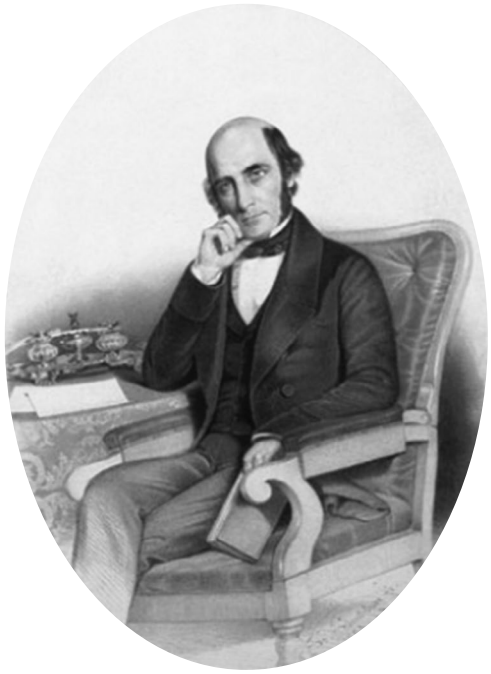

Fig. 3. Portrait of W.J. Little, who first described the orthopedic consequences of perinatal pathology

laid in the studies of great pathologists Jean Cruvelier (1829) and Karl Rokitansky (1856) and pediatrician Heinrich Henoch (1842). They reported for the first time that stillborn and liveborn babies who died early had brain changes.

In the later nineteenth century, the neurological foundations of CP were being studied, which resulted in the idea of CP as a neurological disease. Researchers known at this time include Jean-Martin Charcot (1825–1893), the founder of modern neurology, who was the teacher of a whole generation of neurologists, Jules Cotard (1840–1889)

Fig. 4. Illustration from W.J. Little's book *On the Influence of Abnormal Parturition, Difficult Labors, Premature Birth, and Asphyxia Neonatorum, on the Mental and Physical Condition of the Child, Especially in Relation to Deformities*, illustrating a typical case of a patient with a disease that was later named in his honor

who formulated the concept of the morphological substrate of lesions in CP, and Joseph Babinski (1857–1932) who described pyramidal reflexes.

The comprehensive study of CP is associated with William John Little, who was born in London in 1810 [4]. He was a peer of Nikolai Ivanovich Pirogov. At the age of 4, William John had poliomyelitis, which resulted in a paralytic clubfoot. As a child, he suffered greatly from this defect and decided to devote his life to helping the crippled, received a medical education, and became a surgeon (Fig. 3). While studying in Berlin, he met Dr. Strohmeyer who became famous for his method of treating clubfoot (closed tenotomy). Little underwent surgery, and he considered himself healthy for the rest of his life. Later, he mastered this technique and applied it widely in London.

Little was one of the founders of the Royal Orthopedic Hospital in London, which became the center of surgical orthopedics of the time for decades. He summarized his research in a book published in 1834, *On the Nature and Treatment of Deformities of the Human Skeleton* (Fig. 4). In the latter half of the nineteenth century, he published an article entitled "On the influence of abnormal parturition, difficult labors, premature birth, and asphyxia neonatorum, on the mental and physical condition of the child, especially in relation to deformities," where he summarized the idea of the etiology of CP [5].

What Little's main conclusions should be mentioned? On the nature of the disease in newborns, it was said that "a specific lesion in newborns is spasmodic stiffness of the limbs, similar to tetanus."

On the causes of CP, he said "I have seen very many cases of mental and physical deformities related to causes acting at birth, such as asphyxia, mechanical trauma during or immediately after childbirth."

On the outcome of asphyxia, he said that "obviously, most of the pediatric patients saved thanks to the skillfulness of obstetricians are not affected by this ailment." Thus, he was one of the first to declare that asphyxia can and should be controlled, and low birth-weight children must be saved.

Another specialist who contributed to solving the problem of CP was William Osler, a Canadian professor of clinical medicine at the Pennsylvania State University (Philadelphia since 1884). Based on the experience of the follow-up of 151 pediatric patients, he published the monograph *CP* (1889). It was Osler who offered this term to describe a non-progressive neuromuscular disease in pediatric patients. To a certain extent, Osler agreed with Little regarding the causes of CP as "usually starting at birth." However, he preferred the hypothesis of a traumatic origin of the disease, resulting from "meningeal hemorrhage and compression of the brain and spinal cord" [6].

The third specialist who made a significant contribution to the study of CP was the Austrian scientist Sigmund

Freud (1856–1939) (Fig. 5). In 1885, Freud began working at the Salpetrier Clinic under Charcot's supervision and published many articles and three monographs on spastic paralysis in pediatric patients. He proposed the first classification of CP. Freud proved in detail that CP is caused by damage in the brain, and not in the spinal cord. He suggested that "taking into account that the same disorders during childbirth often do not lead to the development of pathology, it can probably be congenital in nature, and birth disorders in this case are a symptom of a deeper pathology that affects the development of the fetus" [7].

Freud agreed with Little on the etiological effect of asphyxia and birth pathology and made two significant assumptions, the importance of which was appreciated 100 years later. Since asphyxia does not always precede the birth of a child with CP, in some cases, the pathological causes should be sought in the prenatal period; asphyxia may be the result of impaired intrauterine development of the brain, that is, a problematic birth is a consequence of a problematic pregnancy.

In the nineteenth century, the main position of doctors regarding the treatment of CP was "active non-intervention." Surgical treatment was considered an exception, and it had few supporters. Tenotomy remained the only option for surgical treatment (Delpekh, Strohmeyer, Dieffenbach, Little, and Pirogov). N.I. Pirogov described clearly spastic paresis in his work on lengthening of the Achilles tendon. He treated pediatric patients with CP through multiple closed tenotomies. When the use of multiple closed tenotomies began in the twentieth century as a new treatment method, authors are advised to return to the roots.

At the end of the nineteenth and beginning of the twentieth century, active development of surgical approaches to the treatment of spastic paralysis started. Lorenz (Fig. 6) performed a resection of the obturator nerve in 1887 due to the spasticity of the adductor muscles of the thigh.

The systematic treatment of CP is associated with the work of Adolf Lorenz (1854–1946), the founder of the modern school of European orthopedics. At the end of the nineteenth and beginning of the twentieth century, surgical approaches to the treatment of spastic paralysis developed rapidly. Lorenz performed a resection of the obturator nerve in 1887 for spasticity of the adductor muscles of the thigh and was a great supporter of these surgeries. However, later, this method was criticized, as it often led to complications.

A breakthrough in neurophysiology was the work of Charles Sherrington (1857–1952), who studied the spinal regulation of muscle tone and turned around the idea of reflexology [8]. N.I. Pirogov, when performing tenotomy, suspected that tendon tension was the result of the irritation of the cerebrospinal system, and only Sherrington showed how it all works, how the brain controls spinal cord reflexes, and how spinal cord reflexes depend on afferentation.

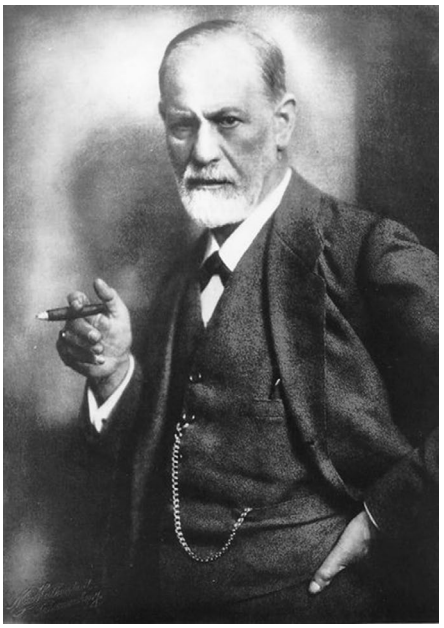

Fig. 5. Sigmund Freud studied and systematized the issues of thiopathogenesis of cerebral palsy

In 1911, Eiby published the results of the transection of the posterior roots of the spinal cord to treat pain, and in 1913, Ottfried Förster (1873–1941), who later became the personal physician of V.I. Lenin (Fig. 7), reported similar surgeries for spasticity. However, the results of this surgery were different, and R.R. Vreden wrote in 1930 that Förster's surgery was disappointing.

In 1913, the German scientist Stoffel published an article in *JBJS* on the treatment of spastic contractures by selective transection of the motor branches of the peripheral nerves.

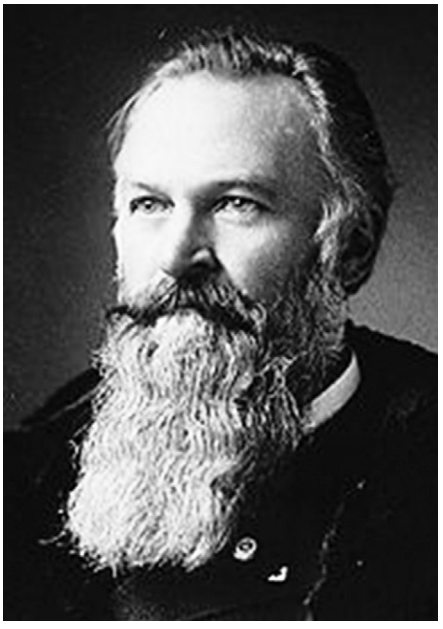

Fig. 6. Adolf Lorenz, the founder of modern European pediatric orthopedics, was the first to perform a resection of the obturator nerve branches to treat spasticity

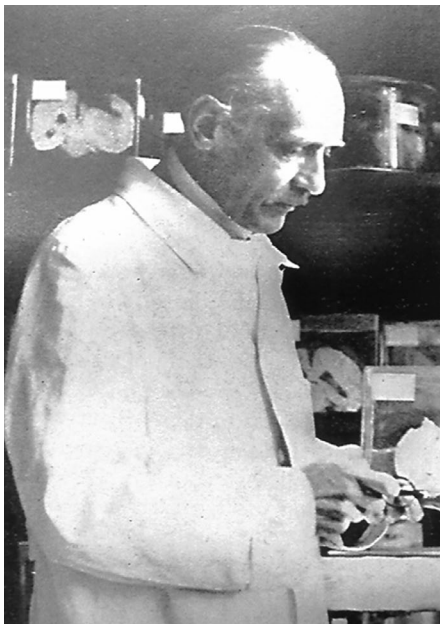

Fig. 7. Ottfried Förster pioneered dorsal rhizotomy for the treatment of spasticity

He worked out in detail the anatomy of the peripheral nerves and believed that it was possible to single out motor zones in their structure and that crossing them achieves appropriate results.

For several decades, surgical treatment has been the exception rather than the rule.

The first steps toward the idea of a multidisciplinary approach were taken in the middle of the twentieth century [9]. In 1947, the American Academy for CP and Developmental Medicine was founded, bringing together physicians from various specialties. The orthopedist Winthrop Phelps became its first president (Fig. 8). Phelps' main recommendations were relaxing procedures, achieving head and trunk control, and training reciprocal movements, which are still relevant for motor rehabilitation. He noted that long-term movement training is crucial and sometimes gives unexpected results, avoiding surgical treatment. Phelps considered surgical treatment only as an auxiliary measure in rehabilitation.

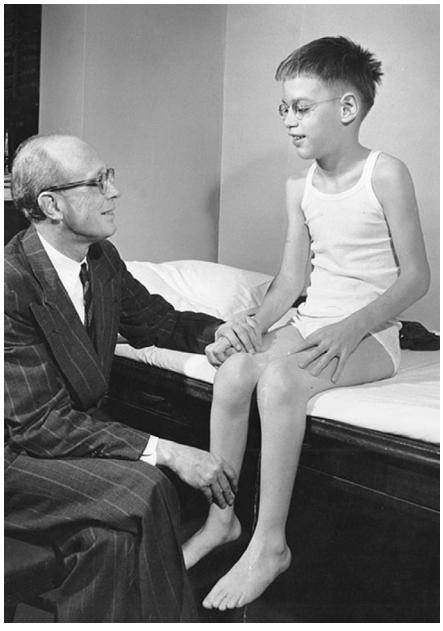

Fig. 8. Winthrop Phelps, one of the founders of a multidisciplinary approach to the treatment of cerebral palsy

In the 1950s, the limits of conservative treatment became apparent to most physicians treating CP. A brilliant constellation of surgeons in the USA and Europe (Greene, Broquay, McDermott, and Baker) started to apply surgeries that had previously been elaborated in detail in pediatric patients with poliomyelitis to correct deformities and restore muscle balance. The range of surgical interventions included neurotomy, tenotomy, muscle elongation and transplantation, arthrodesis, and osteotomy, which are still performed in an improved form nowadays [10].

In the 1970s–1980s, a new surge of interest in the ideas of rhizotomies was associated with the names of Peacock (South Africa) and Ahrens, who specified the indications for surgery, worked out the technique, and applied neurophysiological intraoperative monitoring.

In the early 1980s, Albright implemented the idea of intrathecal administration of baclofen for the long-term reduction of spasticity.

Neuromuscular blocks have entered clinical practice since 1932, when Phelps proposed the use of novocaine injections as a diagnostic and therapeutic procedure for spasticity. In the 1960s, Tardieu, Hariga, and Carpenter proposed the use of ethanol solutions with local anesthetics, and in 1993, Andrew Coman published the first results of botulinum therapy in pediatric patients with CP.

The most important achievement of modern science can be the definition of concepts and the development of CP classifications [11].

The Leningrad school for the treatment of pediatric patients with CP originates from the opening of the Turner Institute for the Rehabilitation of Physically Handicapped Children and Adolescents in 1932. At the Turner Institute, E.Yu. Osten-Saken, Z.A. Lyandres, V.M. Pigin, N.P. Kudryakova, and many others dealt with the problems of CP. The modern school was created by I.I. Mirzoeva, and under her leadership, the direction of comprehensive treatment of pediatric patients with CP was formed. Her students were Kh.A. Umkhanov, A.P. Kutuzov, and V.V. Umnov.

In parallel, the Moscow School of K.A. Semenova was developed, based on the neurophysiological principle of the treatment of pediatric patients with CP. Dynamic proprioceptive correction devices (Adelie suits) have become famous worldwide.

The third most important school for the treatment of paralytic deformities was formed in Samara under

Fig. 9. Irina Iosifovna Mirzoeva, Ksenia Aleksandrovna Semenova, and Alexander Fedorovich Krasnov are the founders of the leading national schools for the study and treatment of cerebral palsy

the headship of A.F. Krasnov. The main works of the representatives of this school focused on tendon–muscle plastic surgeries (Fig. 9).

What is the importance of surgical treatment of CP these days? Neurosurgical treatment of hydrocephalus saves lives and improves its quality. There is a wave of interest in selective dorsal rhizotomy (SDR) again. In 2018, South African researchers published an article on the history of development of this surgical intervention. In 1888, Osler presented the term CP for scientific use, and Eiby and Bennett proposed transection of the dorsal roots to treat pain. In 1898, Sherrington performed an experimental rhizotomy on cats. In 1913, Förster performed posterior rhizotomy to treat spasticity. In 1960, Gross performed partial transection of the posterior roots. In 1972, Sindu (France) developed DREZ anatomy and DREZ-tomy. In 1975, Fraioli and Guidetti used a partial intersection of afferents 1a. In 1976, Fasano proposed responses to the stimulation of abnormal beams in spasticity. In 1986, Peacock (South Africa) introduced the SDR technique as a systematic intervention for CP [12].

Interest in rhizotomies is steadily (albeit unevenly) growing, as evidenced by the number of publications on SDR (392 publications in 1978–2019, including 26 in 1980–1990; 106 in 1990–2000; 121 in 2000–2010; and 155 in 2010–2019). Thus, we are on a wave of new interest in this type of surgical treatment [13].

Dr. Park of St. Louis Children's Hospital (USA) has considerable experience in the use of SDR. From 1987 to 2018, the surgery was performed on 3897 patients aged 2–49 years.

Last year, the National Health System (NHS) in the UK published a report that SDR combined with intensive physical therapy improves overall motor function and quality of life and reduces pain to a greater extent than would be expected without SDR. Data have been used to amend the NHS policy in the UK from 2018, which resulted in public funding for SDR in pediatric patients aged 3–9 years with CP.

However, is everything so good? Studies have not provided conclusive evidence of long-term functional outcomes. The long-term effect on spasticity is unclear, and a significant need for additional treatment has been established. Immediate and long-term complications are not uncommon, but they are not properly reported. Thus, it cannot be stated that SDR solves all problems and that it is a simple surgery without risk of loss.

A relatively new technique is intrathecal baclofen therapy (IBT). The baclofen pump can be used to control hypertension in patients with severe spasticity. Both IBT and SDR may reduce spasticity and improve general motor function in chair-bound patients. The complication rate is higher after IBT, partly due to the device-associated risk. No comparative studies have been conducted on the efficiency of these two treatment methods, and important factors related to both health and patient care and comfort have not been investigated.

In the Turner Scientific and Research Institute for Children's Orthopedics, V.M. Kenis (2014) used the baclofen test to predict the outcomes of SDR. Experience in the implementation of SDR in the Turner Scientific and Research Institute for Children's Orthopedics is quite successful and ongoing.

Hip joint dislocation is another problem in the treatment of CP. At present, the direction of early detection of CP is actively developing. In this regard, the professional community should promote the widespread use of hip screening in children with CP, using the most appropriate program. If patients and their families had a choice, it would undoubtedly be the surveillance program. Specialists who wait for the implementation of the program "from above" miss the opportunity to provide quality care to patients. Every

patient with CP should be treated as having a hip disorder until proven otherwise.

Preventive surgery for progressive hip subluxation aims to prevent irreversible changes. Opinions on the efficiency of these surgeries vary; however, in general, this surgical technique is effective.

Reconstructive surgery for subluxation and dislocation of the hips is aimed at restoring the relationship in the joint and preventing pain.

What current stepwise recommendations can be given for spastic hip instability? Spastic hip instability is the second most common orthopedic problem in pediatric patients with CP, following foot deformities. Population studies have shown that it is registered in approximately one-third of pediatric patients with CP and depends on the level of the Gross Motor Function Classification System. Soft tissue surgery to treat subluxation does not always prevent progression, but it can postpone bone reconstruction to a more appropriate age. In the case of pronounced violations of the ratios, children who are unable to move need reconstructive surgery. When planning surgery, age, possibility of simultaneous bilateral intervention, and combined surgeries on the femoral and pelvic components should be taken into account. Spastic instability of the hip joint is the only one in CP that has a consensus solution, that is, the surgery should be performed on a child with CP and hip joint instability. This must be done because a morphologically well-formed hip joint is usually painless; it does not cause inconvenience in the sitting position and during transportation, facilitates personal hygiene care, and improves the quality of life.

Hip arthroplasty is currently a common surgery [14]. In 2019, American researchers analyzed the USA national database and revealed that 2,062 hip replacements and 2,193 knee replacements were performed from 2005 to 2014. For pediatric patients with CP, hip arthroplasty is indicated at a younger age; thus, there are longer hospital stays, fewer incidence of obesity, coronary heart disease, diabetes mellitus, and peripheral vascular diseases. These patients have a higher risk for general postoperative and surgical complications, specifically acute postoperative anemia. The researchers concluded that since these pediatric patients are characterized by satisfactory long-term results of arthroplasty, these surgeries can be successfully performed with careful medical supervision and surgical planning.

One of the modern achievements is single-stages multilevel surgical correction of deformities (SEMLS, single-event multilevel surgery). SEMLS is the optimal approach in modern CP orthopedics. The method is based on the postulate of James Gage, which he proclaimed in the 1970–1980s, "Pathological levers at all levels must be taken into account in order to achieve an optimal result and prevent relapses."

When comparing the efficiency of staged and single-staged surgeries, the advantages of the latter are obvious. Single-staged interventions had a significantly better effect on the gait, posture, and functional mobility of pediatric patients with CP than staged ones. Parental satisfaction was also higher in the single-staged intervention group. The only correct method of surgical treatment for CP is single-stage multilevel interventions. SEMLS is the treatment of choice, potentially leading to better outcomes.

Stepwise interventions can also be used to treat pediatric patients with CP in developing countries such as India and South Africa where the SEMLS approach is not always feasible.

Regarding the efficiency of multilevel interventions, additional interventions are required to eliminate new contractures and deformities that developed during growth in 37% of the patients.

These interventions can be considered part of multi-staged multilevel surgery; thus, when planning surgeries, parents and children should be informed about the need for additional surgical interventions. For this reason, the term single-step multi-level correction cannot be interpreted literally as surgery once in a lifetime or as multi-level single-step intervention.

The place and role of SEMLS in overall surgical strategy when choosing a treatment algorithm for pediatric patients with CP still need to be formally defined. Moreover, whether SEMLS should be the last resort measure after the failure of the primary treatment for spasticity (IBT or SDR) or whether real SEMLS should be understood as the simultaneous performance of neuromodulation and correction of orthopedic pathology (multi-level, multi-organ surgery under the same anesthesia in one surgical session) remains to be determined.

Until issues of the balance between neurosurgical and orthopedic treatment are resolved, our complex, fragile patients need to be examined and treated following a coordinated multidisciplinary program that can help families choose a particular approach for their children [15].

The assessment of the surgical prospects for the treatment of CP is difficult. We face the primary source of the problem, which is brain damage. Surgical treatment of pediatric patients with CP should be based on a multidisciplinary approach, early detection of orthopedic pathology, and personalization, taking into account the psychological and social effects of surgical treatment and the development and improvement of surgical techniques.

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Author contributions. *A.G. Baindurashvili* developed the concept and performed scientific editing of the article. *V.M. Kenis* developed the concept and wrote the article.

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

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