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Contents

„Immortalis est ingenii memoria"

Seneca

Natalia Gheorghiu *– Pioneer of Pediatric Surgery in Moldova*

Original Article

Transposition of the tibialis anterior muscle as a part of the complex treatment of congenital talipes equinovarus in children

Kykosh G.V., Korolkov A.I.

State Institution "Sytenko Institute of Spine and Joint Pathology of the National Academy of Medical Sciences of Ukraine"

Abstract

Transpoziția mușchiului tibial anterior în tratamentul complex al piciorului strâmb congenital la copil

Materialele articolului prezintă rezultatele utilizării transpoziției mușchiului tibial anterior în tratamentul complex al piciorului strâmb la copii cu vârste mai mari de 2 ani. Autorii au dezvoltat și testat clinic sistemul de tratament diferențiat complex al piciorului stârmb congenital la copii pe baza procedeului propus de I. Ponseti. Pe baza analizei tratamentului a 60 de copii cu vârsta cuprinsă între 2 și 17 ani, în 83,3% au fost obținute rezultate bune și satisfăcătoare, reducând astfel frecvența volumului clasic și a intervențiilor chirurgicale traumatice pentru această patologie cu 78%. Ortezele de corecție, bretele de producție proprie, au fost implementate în practica clinică. Acestea vor reduce costul tratamentului până la 40%.

Cuvinte cheie: picior strâmb congenital, procedeul I.Ponsei, transpoziția mușchiului tibial anterior, bretele

Abstract

The materials of the article present results of using the transposition of tibialis anterior muscle in the complex treatment of clubfoot in children older than 2 years. The authors have developed and clinically tested the system of differentiated treatment of complex congenital clubfoot in children on the basis of I. Ponseti methodics. Based on the analysis of the treatment of 60 children aged from 2 to 17 years 83.3% of good and satisfactory results received, thus reducing the frequency of the classic volume and traumatic surgery for this pathology by 78%.
Corrective orthoses – braces of own production were implemented in clinical practice. They will reduce the cost of treatment up to 40%.

Keywords: congenital talipes equinovarus (clubfoot), I. Ponseti methodics, transposition of tibialis anterior muscle, braces

Correspondence to: e-mail: korolkovmd@gmail.com; mob. +380506347105

Introduction

The clubfoot or congenital talipes equinovarus (CTEV) is one of the most frequent malformations of the musculoskeletal system in children, it occurs in 1-3 cases per 1,000 newborns (35-40% of all congenital foot deformities) [1, 9, 16]. Techniques which are currently used as a conservative treatment (durable stage plaster cast methodics) do not always lead to complete cure of the patient, and frequent relapses of deformity require subsequent extended and traumatic surgical interventions in most (up to 68%) cases [15]. To date, a large number of CTEV surgical treatment methods, which vary in volume and implementation technique, have been proposed. Analysis of results of methods proposed, of most commonly used surgical procedures also does not add optimism - primary operations relapses reach 54% [10, 13]. Currently, the technique by *I. Ponseti* became the most popular in treatment of CTEV. It is characterized by a number of differences from the "classic" methods – in principles of staged elimination of deformity components, casting technology and duration of the foot deformity correction in a plaster cast, duration of Achilles tenotomy performance and orthosis peculiarities in the rehabilitation period (the brace use). If the early treatment was provided (at the age of 3 months) – feet maintain mobility, severe muscle atrophy is not marked, the full range of motion in ankle joint and joints of the foot is maintained, and the percentage of successful results is 85 - 95% [3, 15, 17]. Works of many authors show that applying the principles of methodics by I. Ponseti in children older than 3 years, as the treatment of CTEV, can also give good results in foot deformity correction, on condition of widening indications to certain surgical interventions, particularly, a transposition of m. tibialis antetior tendon on IIIrd cuneiform bone of the foot, the so-called transposition of Anterior tibialis muscle (TTAM) which acquired significant popularity as anti-relapse surgery. It is based on the movement of insertion point outwards from the axis of subtalar joint, which turns this muscle into pronator, and aims to reduce the imbalance between the supinators and pronators. [6, 7, 11, 14].

The main indication for the transposition of tibialis anterior muscle is the dynamic supination of foot in children over the age of 2.5 years, persisting after the correction of the deformity recurrence; TTAm significantly reduces the need to perform "medial release", also prevents subsequent relapse and normalizes talocalcaneal divergence. However, it should be noted that the isolated TTAm is ineffective in rigid relapse of foot deformities [8, 13, 19].

To present the result of using the transposition of tibialis anterior muscle in the complex treatment of congenital talipes equinovarus in children older than 2 years.

Materials and Methods. 60 patients were treated by the I. Ponseti methodics in the Clinic of diatric Orthopedics of the State Institution «Sytenko Institute of Spine and Joint Pathology of the National Academy of Medical Sciences of Ukraine», including 41 boys and 19 girls, in which the transposition of bialis anterior muscle was the one of essential elements of treatment (age of patients was from 2 to 17 years (103 feet), the average age of boys - 6.2 years, of girls - 7.5 years, bilateral pathology was present in 72% (43 patients)**,** relapses of deformation were present in 67% of cases analyzed, including 34% - after surgical treatment in other hospitals, and 33% - after conservative treatment (tab. 1). The average period of observation was from 6 months to 6 years. Estimation of foot deformities' severity before and after treatment was conducted using a Dimeglio scale which allows to evaluate the effectiveness of treatment of clubfoot performed by the I. Ponseti methodic, and characterizes the condition of the foot in the range between 0 and 20 points (fig. 1): 0 corresponds to the normal foot, the deformity having 5 points or less is classified as a light degree; 6-10 as moderate; 11-15 as heavy; 16-20 as very heavy) [2, 4, 15, 18]. Before the beginning of treatment, during and after it, a comprehensive survey of patients was conducted. It included: ultrasonography of Achilles tendon (surveying its structure and length), electromyography of lower limbs, plantography, pedobarography, podometry, x-ray of foot in three projections, medical examination of neuropathologist and computed tomography or MRI examination (according to indications).

Table 1. **Division of patients according to sex and peculiarities of the previous treatment**

	Division according to the type of previous treatment			One-sided pathology	Bilateral pathology
	Primary treatment	Relapse after conservative treatment	Relapse after surgical treatment		
Boys	12	17	12	13	28
Girls	8	3	8	4	15
Total	**20**	**20**	**20**	**17**	**43**

Type of foot (by the Dimeglio scale)	Severity of foot deformities	Points
I	light degree	<5
II	moderate	6-10
III	heavy	11-15
IV	very heavy	16-20

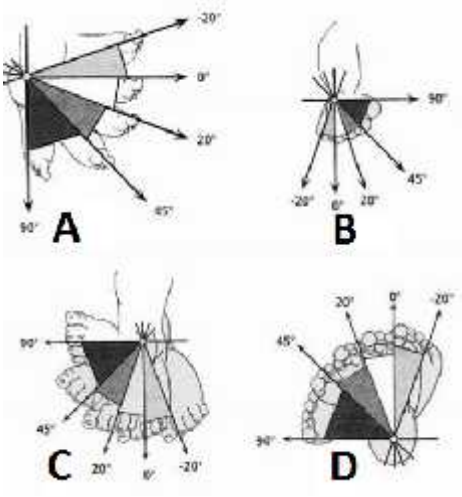

Fig. 1. Estimation of foot deformities' severity by the Dimeglio scale.

Results and Discussion

We developed and put into practice algorithmic scheme of treatment of children suffering from CTEV, based on the I. Ponseti methodics (fig. 2), in which three age groups of children were allocated: first - from 2 weeks to 2 years old, second - from 2 years old to 10 years old, and third - over 10 years old.

According to this algorithm, children aged between 2 and 10 years, after the complex examination, passed a compulsory preparatory step before surgery – stage plaster cast with performance of 4-6 corrective bandaging according to the I. Ponseti methodics (during 4-6 weeks).

The obtained data give coincides with the literature [5, 6, 12, 14] that the stage of preliminary plaster casting in children suffering from CTEV is necessary in the age of 2-10 years - it reduces the deformation of the foot and prepares tissues to a subsequent operation, and consequently significantly reduces the severity and duration of subsequent surgical intervention, but, at the same time, provides the complete correction of residual deformation of the foot, and often even both feet at the same time, in one stage. It should be emphasized that, after a preparatory plaster cast stage, in 100% of cases the stable equinus and the residual adduction of the forefoot remained.

Surgical technology consists of several elements:

- the surgical intervention is performed on the rear part of foot - either the percutaneous achillotomy (it was performed in 65% of cases) or achilloplasty (in 31,6% of cases). The posterior release of foot with incising the capsule of the ankle and subtalar joints performed in 28,3% (volume of intervention on the rear part of foot depended on the severity of the residual equinus and ultrasound results (results which indicated the presence of pathologically changed Achilles tendon and the spread of cicatricial process in the rear part of foot served as indications to undertake an extended surgical intervention));

- the tendinous portion m. abd. hallucis longus is released all along its length through a linear incision along the medial surface of the foot, followed by its Z-shaped elongation (fig. 3, 4). Then, the tendon of tibialis anterior muscle is exposed all along its length (fig. 3b) with a cut-off from the attachment site, followed by stitching by B. Cuneo (fig. 5a). Next, after the preliminary X-ray visualization of IIIrd cuneiform bone (fig. 5b), the stitched tendon of tibialis anterior muscle is carried on the dorsal surface of the foot, and, using straight bone needles, the stitching of tibialis anterior muscle across the IIIrd cuneiform bone, with moving the suture knot to the plantar surface of the foot with simultaneous bringing the foot in the position of maximum dorsiflexion, is performed (fig. 5, 6).

TTAM to IIIrd cuneiform bone in combination with the extension of abductor hallucis muscle held at 100% of children in investigated groups and, according to indications (in 62% of cases), the plantar aponeurotomy performed. "Classic", Zatsepin-like, type of surgical interference with expanded volume of surgery was performed in none of cases on that group of children.

The 2nd surgery stage supplemented to 16 children (26,6%) by the medial release with performing the dissection of capsule of cuneometatarsal, naviculocuneiform and talonavicular joints on the medial surface. The main indicator of surgery efficiency, except of all clinical deformation elements removal, was the normalization of interactions in the talonavicular joint, which was verified at the anterior-posterior X-rays of foot.

In addition to the elements listed above, cuneiform "+ -" osteotomies of cuneiform and cuboid bone using fixation by spokes (wires) or U-shaped staples were performed to 4 children.

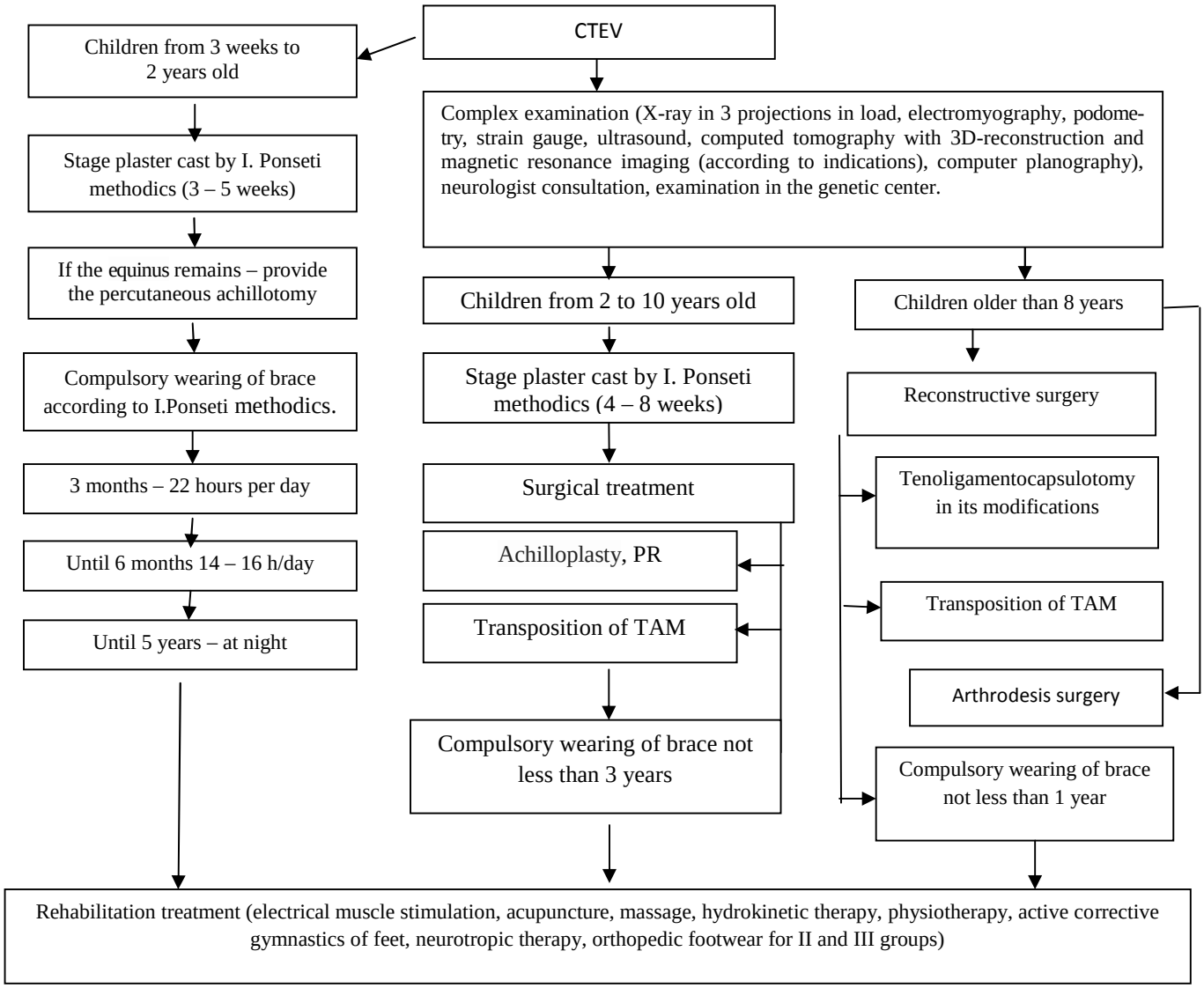

Fig. 2. Algorithm for the treatment of children with CTEV. **(**CTEV - congenital talipes equinovarus; PR - posterior release; TAM - tibialis anterior muscle**)**

When performing TTAM, in 64% of cases we detected the deviation (dystopia) at location of m. tibialis anterior - the place of its attachment was located from 0.5 to 2 cm distally than it is described for the normal foot. According to the "Atlas of Human Anatomy", this muscle "originates from the lateral condyle and the lateral surface of tibia and the interosseous membrane of leg, in the lower third of shin it goes into a long flat tendon, which lies in the tendon channel under retinaculum mm. extensorum inferius and, first, heads to the medial edge of foot and then on the plantar surface of foot where it is attached to the medial cuneiform bone and to the base of Ist metatarsal bone." However, studies conducted show a significant diversity of locations of tibialis anterior muscle insertion - see fig. 7 a, b [7, 13].

Retrospectively evaluating the performance of the muscles by the electromyography results, we found that bioelectric activity (BEA) of m. tibialis anterior was higher by 30-45% compared with adjacent muscles of the foot (m. tibialis posterior, m. flexor digitorum longus and m. flexor hallucis longus). More detailed analysis of the obtained data concerning muscles' changes in clubfoot will be presented in subsequent publications.

The complete foot correction was achieved intraoperatively in 85% of cases. That allowed us to exclude the use of spokes (in other cases, the Transossal fixation of corrected position of the talonavicular and talocalcaneal

joints using Kirschner wires was performed). After suturing the wounds, a circular plaster cast was applied for 5 to 5.5 weeks according to I. Ponseti methodics. By plaster casts' removal, braces own design (fig. 8) with a fixed orthopedic shoe were used. They have crossbeam which has same distance as shoulders width, setting the foot at 45-55° for corrected foot and at 30º for a healthy foot, with dorsiflexion of foot – at 10-20°. Against the background of fixing feet in the brace, the rehabilitation treatment – regular courses of electrical stimulation of tibia and fibula, acupuncture, ozokerite therapy, electrophoresis of foot; massage, hydro-massage; daily corrective gymnastics (held at least 3 times a day by parents) was performed.

Bracing is also an integral part of the treatment of CTEV also for children older than 2 years, wherein the compliance with their wearing is very important: the first 1.5 months after removal of the plaster cast immobilization - at least 22 hours/day, then - at bedtime, for, at least, 1 - 1,5 years.

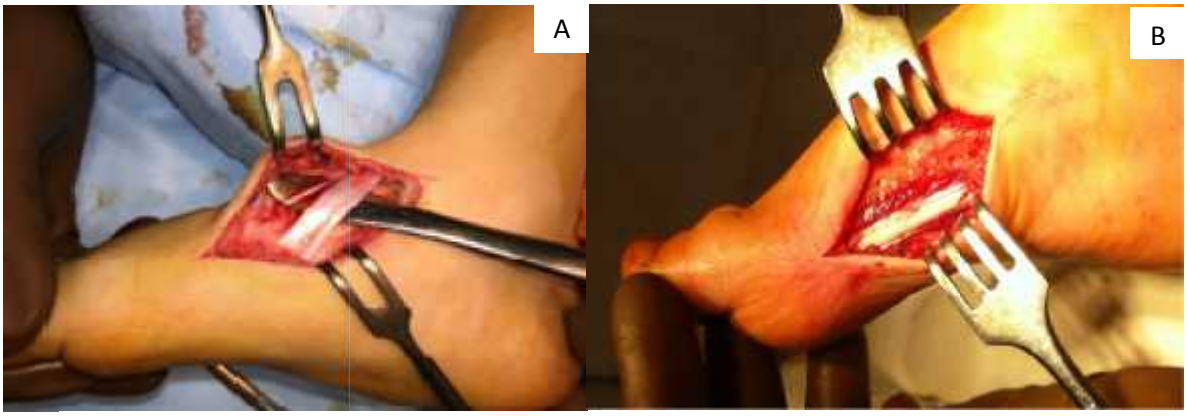

Fig. 3. Dystopic tendon of tibialis anterior muscle (A); B - contracted m. abd. hallucis longus

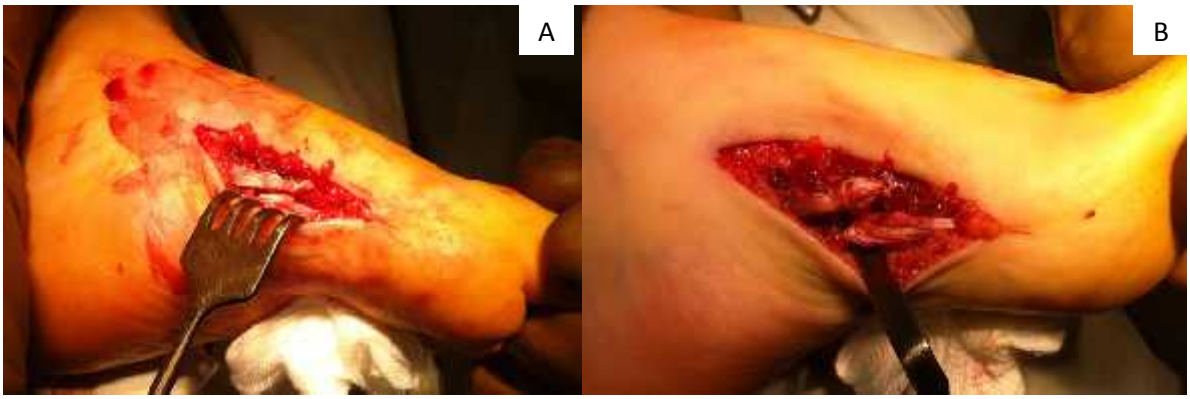

Fig. 4. Z-shaped elongation of the tendon portion of m. abd. hallucis longus

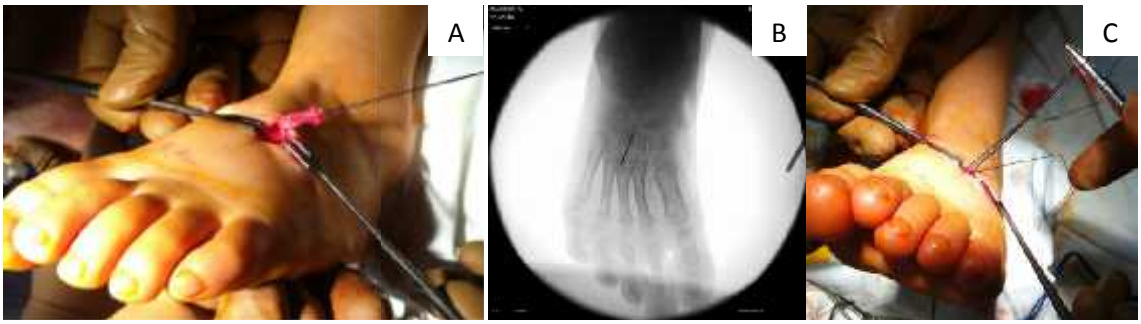

Fig. 5. Transposition of tibialis anterior muscle on III[rd] cuneiform bone: **a.** The bringing of tendon of tibialis anterior muscle stitched by B.Cuneo to the dorsal surface of the foot; **b.** X-ray visualization of III[rd] cuneiform bone; **c.** The bringing of tibialis anterior muscle across the IIIrd cuneiform bone.

Fig. 6. Feet appearance after wounds' suturing.

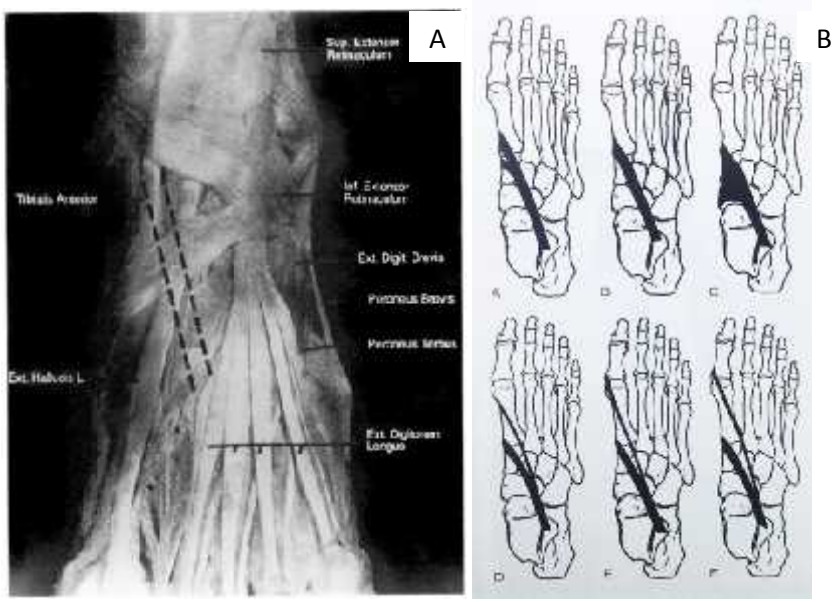

Fig. 7. A - anatomical preparation - scheme of transposition of the tibialis anterior muscle tendon to the III[rd] cuneiform bone (Congenital Clubfoot Fundamentals of treatment Ignacio V. Ponseti, 1996.); B - variants of TAM tendon attachment (Hallisy JE. The muscular variations in the human foot: A quantative study Am J.Anat. 1930).

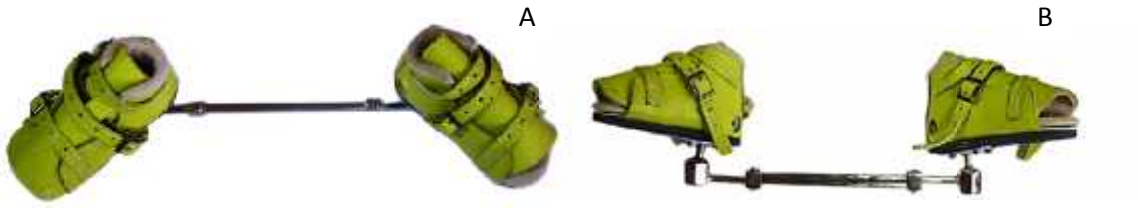

Fig. 8. Braces of own design (patent of Ukraine №95542)

We want to remind that the main purpose of bracing is to prevent relapses. According to data cited in the article by I. Ponseti "Clubfoot" in cases when parents adhered to the wearing mode of fixator, there has been a relapse in only 5 to 10% of children [13].

The results of clubfoot treatment were estimated as good (supporting foot, reached both with the help of subcutaneous tenotomy, and without it); satisfactory (foot requiring the limited rear release, transposition of the tibialis anterior muscle and the shortening of lateral support, or these operations were planned); or bad (a foot requiring a full posterior-medial release, or which was executed as planned).

To confirm the effectiveness of this technology we give an example of its use.

Clinical example. Child V., 5 years old, with a diagnosis: "Severe degree of bilateral congenital talipes equinovarus" (10 points by Dimeglio scale). Relapse of deformations of both feet long conservative treatment (plaster cast by "classical methodics" during 7 months) (fig. 9, 10).

The staged correction of feet deformities was done by I. Ponseti methodics for 6 weeks with changing 5 plaster casts. After their removal, subject to stable equinus of foot, residual adduction of anterior part, the surgical intervention was performed in the following volume: Partial percutaneous achillotomy, transposition of TAM to IIIrd cuneiform bone of both feet, the m. abductor hallucis lengthening on both feet, right-sided plantar partial fasciotomy.

In fig. 11 and 12 - in the long term, after 12 months after surgery, feet are mobile, dorsal flexion of feet is sufficient, there was no tendency to relapse observed. No complaints of the patient on pain in feet. Fixing the feet in the braces is continued at the time of sleep against the background of regular courses of conservative rehabilitation treatment. In fig. 13 (a, b, c), confirming the effectiveness of the correction provided, the static and dynamic barometric data is presented, that shows the normalization of the load on feet, for both the posterior and anterior parts.

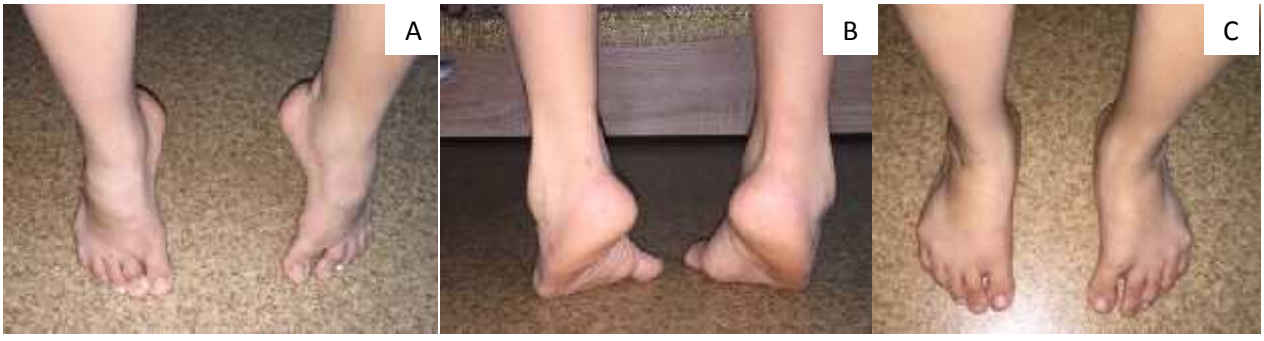

Fig. 9. The child V., 5 years old, feet appearance before treatment begins

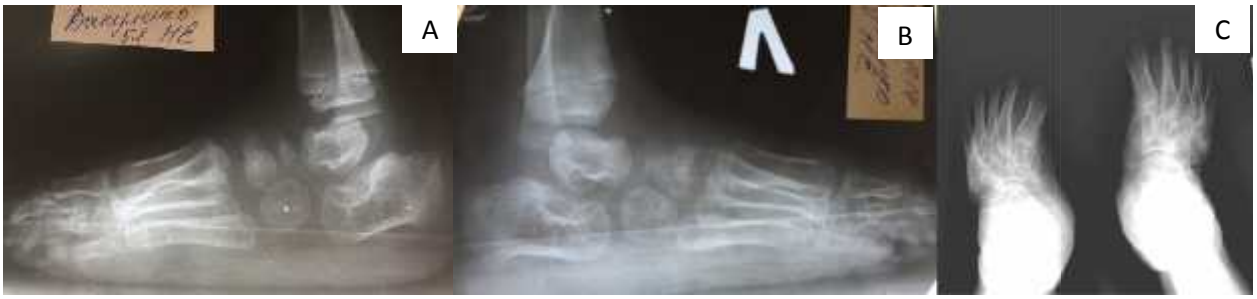

Fig. 10. X-ray images of feet of the child V., 5 years old, standing with the load before treatment

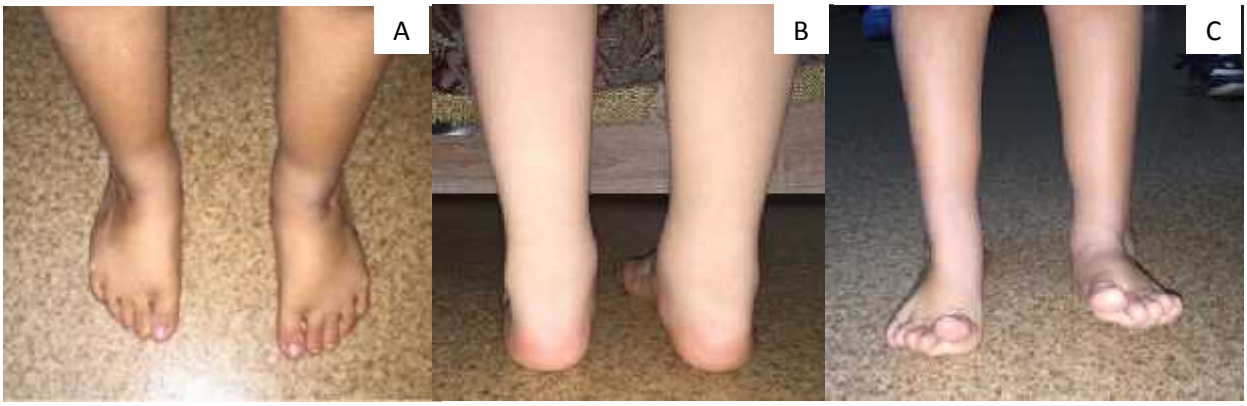

Fig. 11. Feet appearance – the child V. stands, 6 years (12 months after treatment)

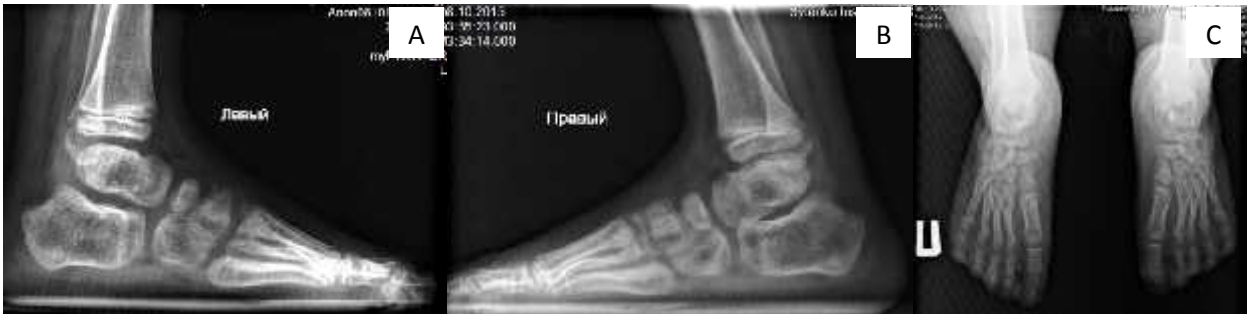

Fig. 12. X-ray images of feet of the child V., 6 years old, standing with the load (12 months after treatment)

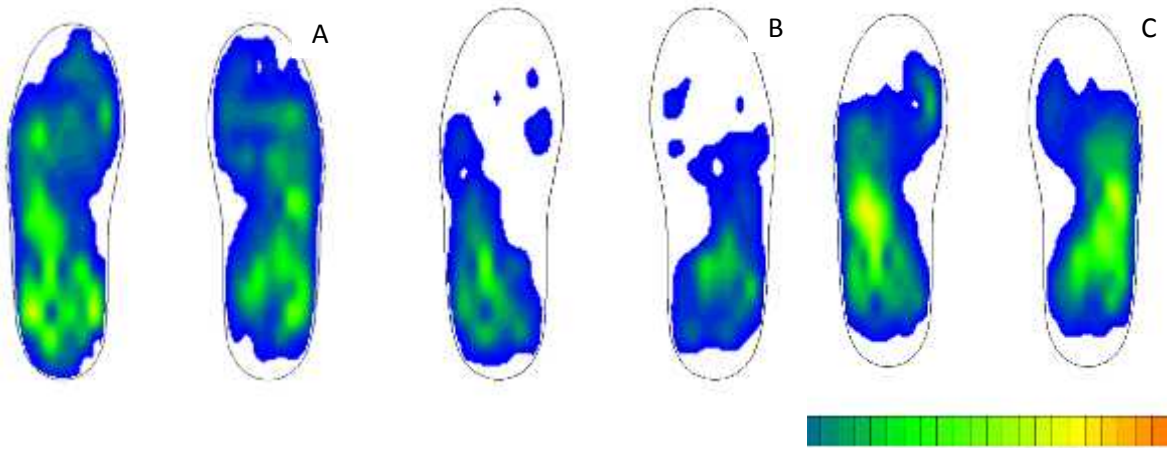

Fig. 13. Dynamic barometry of the feet of child V., 6 years, 12 months after surgical treatment: A - average value; B - standing without movement; **C** - biggest load for the longest period of time during a roll-over)

Conclusions

1. The system of differentiated treatment of complex congenital talipes equinovarus in children based on the I. Ponseti methodics, which is developed and clinically tested, allows the individual approach in solving the problem of this disease's treatment, and the frequency of the "classic" voluminous and the traumatic surgical interventions is reduced by 78% for given pathology. The use of staged plaster cast methodics by I. Ponseti on the preoperative preparation of children in the age between 2 to 10 years is an effective way of significant reducing the severity and duration of the surgery, allowing simultaneous correction of residual deformities of both feet at one surgical stage.

2. Transposition of the tibialis anterior muscle is an essential element in anti-relapse complex treatment of congenital talipes equinovarus by I. Ponseti methodics in children aged from 2 to 17 years, but its performance allowed to obtain 3,3% of good and satisfactory results while respecting the technology, peculiarities of treatment and terms using fixation brace.

3. Using domestically produced brace reduces individuals treatment cost by 40%, excludes durable plaster casting of child's foot, does not lead to severe muscle hypotrophy and neurological complications, and allows child to start stages of a comprehensive rehabilitation treatment earlier, resulting in persistent good anatomical and functional results.

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Application of the hydrosurgery system and plasmajet for thoracoscopic pleural cavity debridement in children with fibrinothorax

Bataev S.[1], Rozinov V.[1], Ignatiev R.[1], Zurbaev N.[2], Fedorov A.[1], Molotov R.[1], Afaunov M.[1], Karpovich S.[2], Smirnova S.[2], Tsilenko K.[1]

Pirogov Russian National Research Medical University,
National Research Institute of Pediatric Surgery, Moscow[1]
Speransky Children's Hospital, Moscow[2]

Abstract

Aplicarea sistemului de hidrochirurgie și a plasmajetului în debridarea toracoscopică a cavității pleurale la copii cu fibrinotorax

Toracoscopia a devenit o modalitate favorizată în tratamentul empiemului pleural la copii. Cu toate acestea, factorii care afectează rezultatul administrării toracoscopice rămân neclari.
Scopul studiului este de a demonstra eficiența sistemului de hidrochirurgie "Versajet" și a unității de plasmă "Plasmajet" în tratamentul toracoscopic al copiilor cu complicații pleurale ale pneumoniei.
MATERIALE SI METODE. În perioada 2015-2017, la Spitalul de Copii Speransky din Moscova, 377 copii cu pneumonie au fost tratați, la 62 pacienți (16,45%) din 377 am efectuat drenajul cavității pleurale, 14 pacienți cu vârste cuprinse între 1,6 și 15 ani (în medie, 3,2 ± 3,8 ani) cu empiem pleural au fost operați. S-a efectuat decorticarea pulmonară toracoscopică cu sistem de hidrochirurgie (Versajet-2). Sistemul hidrochirurgical este un instrument chirurgical bazat pe impactul jetului de apă de mare viteză asupra țesuturilor necrotice și inflamate, combinând avantajele debridării țesuturilor moi și evacuarea acestora prin pulsarea jetului de apă. Designul tubului de evacuare și apropierea acestuia de jetul lichid creează un vacuum local, care îndepărtează efectiv fibrina și conținutul lichid prin efectul Bernulli. Consimțământul informat a fost obținut de la toți părinții înainte de operare, iar procedura însăși a fost aprobată de comitetul de etică locală.
REZULTATE. Recuperarea și reabilitarea fără particularități au fost în 13 cazuri. La un pacient cu empiem al cavității pleurale pe dreapta și leziune organică severă a sistemului nervos central a perioadei postoperatorii după o toracoscopie convențională complicată de recurența empiemului pleural.Retoracoscopia cu debridarea cavității pleurale de către sistemul de hidrochirurgie efectuată la șase zile după operația inițială, a avut rezultate satisfăcătoare. În cazul hemoragiei intraoperatorii, a fost efectuată coagularea plasmei de argon, obținându-se aerostasie completă la 2 pacienți și hemostază - la 1 pacient. Durata medie de funcționare a fost de 90 de minute (± 15 minute). Drenajul cavității pleurale este eliminat în ziua a 3-a sau a 4-a după operație. Copiii externați în ziua a 10-a (± 1,2 zile). Examinarea cu ultrasunete și cu raze X la patru luni după intervenția chirurgicală a confirmat absența inflamației în parenchimul pulmonar și reexpansarea plină pulmonară la toți pacienții.
CONCLUZIE. Aplicarea sistemului hidrochirurgical în timpul toracoscopiei asigură o debridare sigură și eficientă a cavității pleurale, decorticările plămânului fiind fără deteriorarea severă a parenchimului pulmonar și crearea condițiilor pentru reabilitarea precoce a plămânilor compromiși.

Cuinte cheie: sistem hidrochirurgical, toracoscopie, decorticare, cavitatea pleurală, fibrinotorax

Abstract

Thoracoscopy became a favored modality in pediatric pleural empyema treatment. However, factors affecting outcome of thoracoscopic management remain unclear. The purpose of the study is to demonstrate efficiency of hydrosurgery system "Versajet" and plasma unit "Plasmajet" in thoracoscopic treatment of children with pleural complications of pneumonia.

MATERIALS AND METHODS. During the period of 2015-2017 at the Speransky Children's Hospital in Moscow 377 children with pneumonia were treated, in 62 patients (16.45%) of 377 we perform drainage of pleural cavity, 14 patients from 1.6 to 15 years of age (on average, 3.2 ± 3.8 years old) with pleural empyema were operated. Thoracoscopic lung decortication with hydrosurgery system (Versajet-2) was performed. Hydrosurgery system is a surgical instrument based on impact of high-speed jet of water on necrotic and inflamed tissues, combining the advantages of soft tissues debridement and evacuation them by pulsating water jet. Design of the evacuation tube and its proximity to liquid jet creates a local vacuum, which effectively removes fibrin and liquid contents by Bernulli effect. Informed consent was obtained from all parents before operation, and procedure itself received approval from the local ethics committee.

RESULTS. Recovery and rehabilitation was uneventful in 13 cases. In one patient with empyema of right pleural cavity and severe organic lesion of central nervous system postoperative period after conventional toracoscopy complicated by recurrence of pleural empyema. Rethoracoscopy with debridement of pleural cavity by hydrosurgery system performed six days after initial operation, with satisfactory results. In the event of intraoperative air leak or hemorrhage, application of argon plasma coagulation had been performed achieving complete aerostasis in 2 patients and hemostasis - in 1 patient. Average operation time was 90 minutes (± 15 minutes). Drainage of the pleural cavity removed on the 3rd or 4th day after surgery. Children discharged from the hospital on 10th PO day (± 1.2 days). Ultrasound and X-ray examination four months after surgery confirmed the absence of inflammation in the lung parenchyma and full lung reexpansion in all patients.

CONCLUSION. Application of hydrosurgycal system during thoracoscopy provides safe and effective debridement of pleural cavity, decortications of the lung without severe damage to the lung parenchyma and create conditions for early rehabilitation of compromised lung.

Keywords: hydrosurgycal system, thoracoscopy, decortications, pleural cavity, fibrinothorax

Correspondence to: E-mail: khassan-2@yandex.ru; Address: 109559. Krasnodarskaya st. 72-3-219. Moscow. Russia. +79260169119

Introduction

Balance between the resistance of the child's organism, virulence of bacteria and time of appropriate pharmaceutical treatment initiation [6] determines risk of developing pleural empyema during complicated pneumonia course. Independent risk factors for development of empyema include congenital and acquired diseases, such as immunodeficiency, viral infection and organic disease of central nervous system, cancer, Down syndrome, congenital thrombocytopenia, tuberculosis, congenital heart disease, preterm birth, oesophageal disorders and cystic fibrosis [1]. There are three stages in natural course of empyema thoracic: exudative, fibrinopurulent, and organizing stage. These stages developing in continuum if inadequate treatment persist. Two last stages are called complicated parapneumonic effusions and may be a reason to perform surgery for lung reexpantion [4]. Recently, in cases of pneumonia with pleural complications, most surgeons perform thoracoscopic decortication. Success of surgery for this disease depends on the quality of removing fibrinopurulent exudate out of pleural cavity [5]. However, the vacuum aspiration used in thoracoscopy and the mechanical elimination of the pathological contents do not always allow complete debridement of the affected pleural surfaces. In addition, during attempts of full debridment bleeding and bronchopleural fistulas often occur, because of damage to inflamed parenchyma of lung and pleurae, especially if their tight fusion exist [3]. In connection to this, it is relevant to use a hydrosurgical unit for the gentle debridement of affected surfaces and a gas-plasma argon coagulator for provision of intraoperative aerostasis and hemostasis.

Patients and Methods

From 2015 to April 2017, in various departments of the G.N. Speransky Children's Hospital was on treatment 377 children with a diagnosis of acute community-acquired pneumonia. Of these, 64 patients (16.58%) had a complicated course of empyema of the pleural cavity, 40 children from 64 with pleural complications of destructive pneumonia required drainage of the pleural cavity. In 14 (21.8%) of 64 children, intensive therapy was ineffective for 4 days after drainage of the pleural cavity, patients underwent x-ray and ultrasound examination of pleural cavities in dynamics, computed tomography was performed in 5 patients. Detection of the persistence of loculated empyema according to CT and / or ultrasound of pleural cavities was an indication for thoracoscopic sanation of

the pleural cavity. Usually used in this operations vacuum aspiration of content and mechanical extraction of fibrinous-purulent overlays cannot ensure complete debridement of the inflamed surfaces, especially if the disease is long-standing or severe. Attempts to completely debridement are complicated by aspiration of air from the pleural cavity during thoracoscopy, which leads to loss of visualization of the operating field and contamination of the chamber. Elimination of these shortcomings leads to an increase in the operation time. In addition, to ensure the complete removal of nonviable tissues is hampered by the possible sucking up of the aspirator to the affected areas, which usually causes additional bleeding and increases the threat of formation of iatrogenic bronchopleural fistulas. In turn worldwide experience of using hydrosurgical dissection and debridement unit "Versajet 2" in situations that require a delicate attitude to the tissues of the body [2] led us to understand the possibility of its use in children with fibrinothorax.

INTERVENTION - SURGICAL TECHNIQUE

At the preoperative stage, location of maximum accumulation of exudate in the pleural cavity were determined using instrumental methods and later aspirated by pleural puncture. This ensures the creation in the pleural cavity of the necessary space for the safe trocar insertion and introduction of endoscope. Location of the first port depends from degree of lung collapsing and position of diaphragm. After this, removal of exudate residues from the pleural cavity and separation of fibrinous peels between pleura sheets followed by washing out pleural cavity with saline were performed. Then we proceed with elimination of fibrinous-purulent deposits from the visceral and parietal pleura sheets and the decortication of the lung with the tip of the "Versajet" hydrosurgical unit, introduced by the port-free technique (fig. 1). Debridement was performed until the appearance of minimal diapedesis bleeding from the surface of the lung, which indicated sufficient cleaning from purulent-fibrinous peels (fig. 2). In the event of an air leakage or bleeding from the parenchyma or chest wall, we perform coagulation with stream of argon hot plasma by Plasmajet unit to achieve complete intraoperative aerostasis and hemostasis. Operation ends with a classical drainage of the pleural cavity.

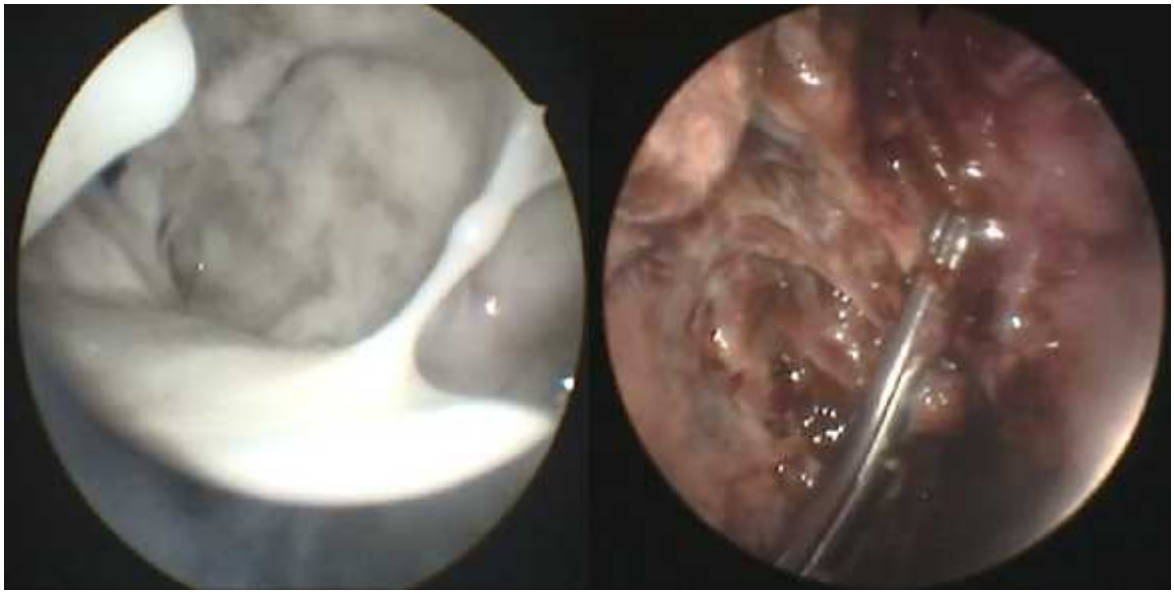

Fig. 1. Insertion of the instrument and the beginning of hydrosurgical debridement of pleural cavity

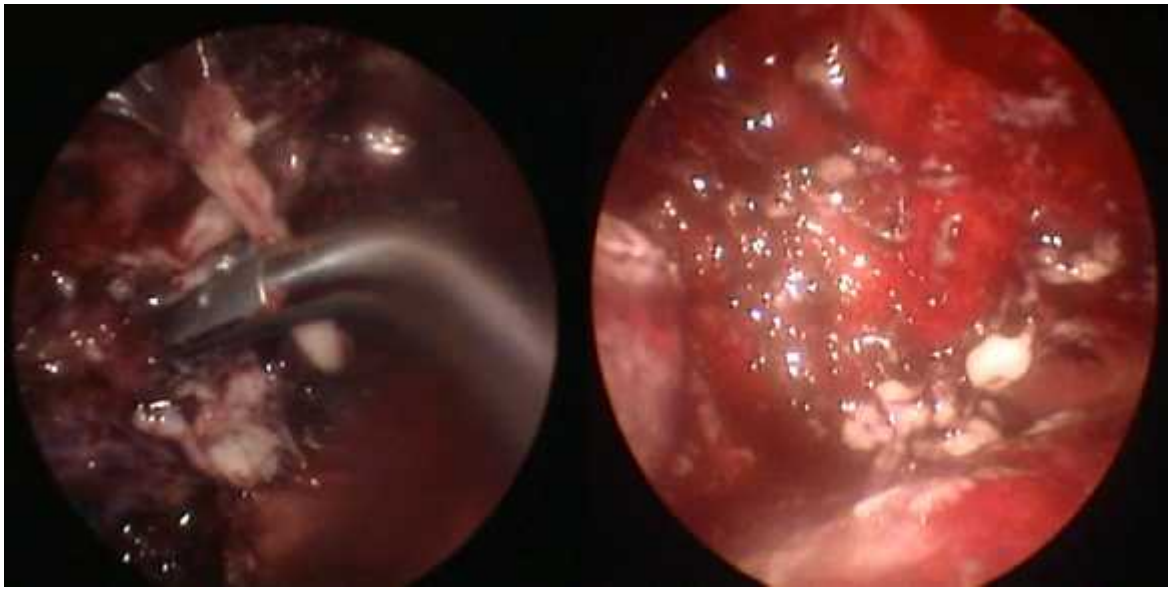

Fig. 2. Complete cleaning of pleura from fibrinopurulent peels

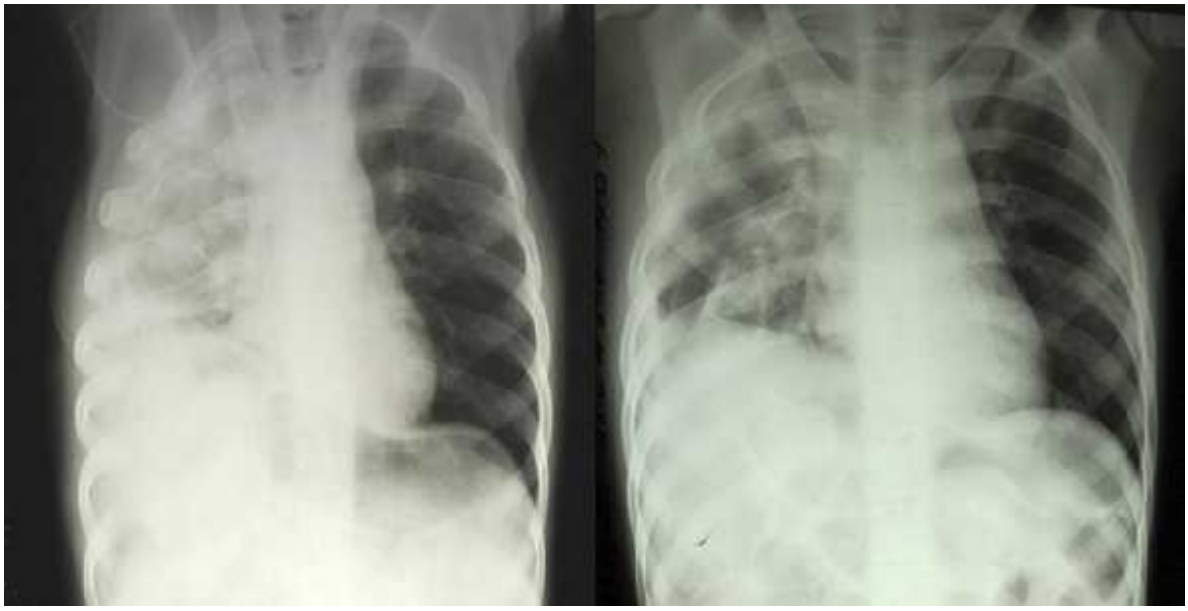

Fig. 3. Postoperative result of hydrosurgical debridement on the 4th postoperative day

Results and Discussion

In all cases, the postoperative period proceeded without complications. Average duration of operation was 90 minutes (± 15 minutes). Drainage from the pleural cavity was removed on 3th day (± 1.1 days) after the operation. PCR analysis of fluid from the pleural cavity showed that in 6 cases Str.pneumoniae was the causative agent, in one - Staph. Aureus, in 7 cases it was not possible to identify the pathogen. After operation, children discharged from the hospital on 10th POD (± 1.2 days), average duration of the hospitalisation was 19 days (± 2 days). Repeated thoracoscopies after hydrosurgical sanation were not performed (Fig. 3). According to chest radiographs and ultrasound of pleural cavities at 3 and 6 months, it was found that all patients lack fibrin overlap and inflammatory changes in the parenchyma of the lungs and pleura

One of the advantages of the hydrosurgical method that it's based on a saline solution that is safe for the patient and does not cause an allergic reaction. High-speed supply of sterile liquid allows perform excision of tissues with one-stage aspirating of detritus and liquid contents, which reduces the time of operation. The advantage of this method lies in the fact that at high values of the power of the apparatus, it is possible to debride dense fibrinous fusion in the pleural cavity. Together with the high accuracy of the liquid flow and the thin depth of cutting of the tissues (1 mm), the water jet device allows selective removal of non-viable tissues, which can reduce the risk of bleeding and damage to the pulmonary parenchyma. Application of argon-plasma coagulation to achieve aero and hemostasis avoids additional manipulations in the pleural cavity and prevents the development of severe complications.

Conclusion

Application of hydrosurgical and plasma scalpel allow us conduct desirable debridement of inflamed surfaces, with the relief of possible complications by aerostasis and hemostasis, what creates favorable conditions for reexpansion of compromised lung and relief of intoxication symptoms for a faster recovery of the patient. However, these statements should be researched in randomized controlled studies.

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The influence of genotype and chronic infection on pulmonary phenotypes in patients with cystic fibrosis

Şciuca S.[1], Barbova N.[2], Selevestru R.[1], Balaneţchi L.[1,5], Cepraga V.[1], Vlas C.[1], Ciupac I.[3], Crivceanschi E.[3], Ceban A.[4]

University of Medicine and Pharmacy „Nicolae Testemitanu", Clinic of Pneumology, Department of Pediatrics, Chisinau, Republic of Moldova[1]
PMSI Mother and Child Institute, Center of Reproductive Health and Medical Genetics, Chisinau, Republic of Moldova[2]
PMSI Mother and Child Institute, Radiology and Imaging Section, Chisinau, Republic of Moldova[3]
PMSI Mother and Child Institute, The Scientific Laboratory of Microbiology and Immunology, Chisinau, Republic of Moldova[4]
Children Emergency Hospital, Department of Pediatric Surgery and Orthopedics, Cluj Napoca, of Romania[5]

Abstract

Influența geneticii și a infecției asupra fenotipurilor pulmonare la pacienții cu fibroză chistică

Fibroza chistică (CF) este o boală ereditară autosomal-recesivă care evoluează cu un conținut ridicat de cloruri de transpirație, boală pulmonară cronică, insuficiență pancreatică cu maldigestie, malabsorbție și scădere în greutate. Sunt analizate interrelațiile dintre fenotipul modificărilor bronhopulmonare la 3 pacienți cu fibroză chistică și colonizarea infecțioasă cu germeni patogeni. Diagnosticarea CF la copiii evaluați a fost confirmată prin cercetarea ADN moleculară pentru a determina mutațiile CFTR. Sa raportat că infecția cu Pseudomonas aeruginosa accelerează procesele distructive ale parenchimului pulmonar și contribuie la răspândirea fibrozei pulmonare, bronhiectazei, emfizemului, distrugerii pulmonare, fibrozei focale sau difuze, pleurezii, toate fiind vizualizate și diagnosticate în examinările imagistice.
Relația dintre genotip și fenotip a fost investigată la pacienții cu CF și infecții pulmonare cu diferite etiologii. Asocierea a două mutații CFTR și a infecției cu P. aeruginosa au fost asociate cu fenotipurile clasice severe ale CF.

Cuvinte cheie: fibroză chistică, CFTR, infecție, fenotip, copii

Abstract

Cystic fibrosis (CF) is an autosomal-recessive hereditary disease that evolves with high content of perspiration chlorides, chronic pulmonary disease, pancreatic insufficiency with maldigestion, malabsorption and weight loss.
The interrelations between phenotype of bronchopulmonary changes in 3 patients with cystic fibrosis and infectious colonization with pathogenic germs are analyzed. Diagnosis of CF in evaluated children was confirmed by molecular DNA research to determine CFTR mutations. It has been reported that *Pseudomonas aeruginosa* infection accelerates the destructive processes of pulmonary parenchyma and contributes to the spread of pulmonary fibrosis, bronchiectasis, emphysema, pulmonary destruction, focal or diffuse fibrosis, pleurisy, all of which are visualized and diagnosed in imaging examinations.
The genotype-phenotype relationship was investigated in patients with CF and pulmonary infection of different etiologies. The association of two CFTR mutations and *Ps.aerugenosa* infection were associated with the classical severe phenotypes of CF.

Keywords: cystic fibrosis, CFTR, infection, phenotype, children

Correspondence to: E-mail: svetlaba.sciuca@usmf.md; mob. +373 79471374

Introduction

CFTR gene mutations and their role in CF pathogenesis remain challenging for both researchers and physicians, so more information on genetic variation would help to study the effect on the phenotype of the disease [12]. CF is the most common autosomal recessive disease in the Caucasian population with very high mortality and is caused by mutations in the CFTR gene [8]. This is a hereditary disease that evolves with high content of perspiration chlorides, chronic pulmonary disease, pancreatic insufficiency with maldigestion, malabsorption and weight loss. The CF phenotype is very heterogeneous even among siblings carrying identical CFTR mutations, suggesting that the severity of the disease is affected by the CFTR genotype and other causal factors [13]. More than 2000 mutations and polymorphisms have been identified on the CFTR gene [9]. An association between the various severe genotypes and CF pulmonary phenotype has been reported in various international studies [2]. The term diagnosis of the disease may be useful in the prognosis of the cystic fibrosis phenotype. Thus, a late diagnosis age may be the result of an unsatisfactory system of health system administration that could lead to a substantial progression of the disease due to inadequate treatment, creating a negative association between age to diagnosis and survival. The findings of various investigators reveal significant deviations between diagnostic term and mode. There are children diagnosed early by prenatal screening without meconial ileus, early with manifest manifestations of the disease or late progression of the disease due to absent treatment [5, 10].

Aim. Evaluation of the influence of CFTR genetic mutation and pulmonary infection on the phenotype of bronchopulmonary changes in patients with cystic fibrosis.

Materials and Methods

Three patients with CF were evaluated in the study, who performed periodic stationary treatment at the Cystic Fibrosis Center in the Pneumology Clinic, Mother and Child Institute.

The positive diagnosis of CF was confirmed based on anamnestic data, clinical examination, paraclinical investigations, automated welding test (Macroduct, USA, Exudose, France). Through informed consent, molecular DNA research was performed to determine CFTR mutations. Pathogenic germs were examined bacteriologically from bronchial secretions and identified by cultural methods using the „WalkAway-96" device, Siemens, Germany.

The imaging examination included ultrasound results, pulmonary radiography. The imaging evaluation of bronchopulmonary lesions was performed by thoracic HRCT (Aquilion 32, Toshiba, Japan). Spiral pulmonary computed tomography (CT) is a radiologically binding exploratory, highly informative exploratory examining and detects structural changes in the bronchopulmonary system. Pulmonary CT provides high information in the assessment of the bronchopulmonary substrate, thus advanced lung bronchial pulmonary emphysema, atelectasis, bronchiectasis, fibrosis, pulmonary sclerosis have been identified in CF.

Results and Discussions

It is believed that the determinant in manifestation of phenotypic expression in patients with CF are inherited mutations. In the Pneumology Clinic, several studies have been conducted to diagnose and monitor several CF patients. Three children with CF mixed with different severity of the disease were evaluated in current research. The CF diagnosis was established based on the results of two sweat chloride tests (>60 mmol/l). The history of the disease, CF diagnostic age, CFTR genotype, body mass index (BMI), pulmonary function test results in children older than 5 years, exocrine pancreatic status were evaluated in the study children. Bacteriological results were obtained from each patient during hospitalization with the identification of pathogenic airway disease.

All patients exhibited characteristic CF symptoms with manifest respiratory and digestive signs. All children with CF have been found to have elevated concentrations of chloride in sweating. The diagnosis of this disease was confirmed in all infants during the first 6 months of life with signs of recurrent bronchopulmonary infections (severe obstructive bronchitis, tremors, recurrent pneumonia, nutrition disorders). Early diagnosis induces suspicion that at the onset of the disease, the children make a severe form or a "severe" phenotype of cystic fibrosis may be assessed for a shorter survival. Alternatively, an early diagnosis age could be the result of prenatal/neonatal positive screening, which generally identifies infants with cystic fibrosis at pre-symptomatic stages, and could thus lead to better survival [5, 10].

However, an early diagnosis was made, but the presence of severe mutation conditioned an evolution with severe respiratory symptoms over time and a nutritional compromise attested at the time of assessment. From confirmation of diagnosis, children were periodically monitored with enzyme replacement therapy and antibacterial treatment for respiratory infections.

The most severe general state was found in the girl who is 8 years old. During last staying in the hospital she had purulent wet productive caugh, hardly expectorated, loss of appetite, fatigue, diffuse bilateral submatity, bilateral diminuated vesicular murmur on

the background of wet medium rales. Respiratory frequence – 45/min, pulse – 150beats/minute, SaO_2 – 43%, FVC - 42%, FEV_1 - 29%, IT - 63%, and the weight of 17,5 kg (p 0-3), height – 116 cm (p 0-3), BMI – 12,96

The boy with the same age also presents a severe form of the disease. He is hospitalised with wet caugh with purulent expectorations, dyspnoea at physical effort, perioral cyanosis, bilateral medio-basal submatity and bilateral medium bulous rales. Respiratory frequence – 28/min, pulse – 95/min, SaO_2 – 96%, FVC - 54%, FEV_1 - 51%, IT - 85%, mass during hospitalisation – 22kg (p 10-25) , height 125 cm (p 25-50), BMI – 14,1. The boy who is 14 years old has a controlled state of the disease, being hospitalised with productive wet purulent caugh, loss of appetite, fatigue, bilateral diminuated vesicular murmur in the inferior segments on the background of wet fine rales. Respiratory frequence – 24/min, pulse – 93/min, SaO2 – 95%, FVC - 78%, FEV_1 - 70%, IT - 77%, weight – 42kg (p 10-25), height – 160 cm (p 25-50), BMI – 16,4

CF is a complex disease with multisystemical involvement. Correlation between the genotype and the severity of disease varies in function of affected systems and organs. Phenotype expression in respiratory system varies a lot and there were found few correlations between the genotype fenotype and pulmonary fenotype. An exception is the fact that patients with pancreatic insufficiency have, generally, a pulmonary disease which is more severe than in patients with normal pancreatic function [3].

The main purpose of our study was to investigate if the mutations had an impact over pulmonary modifications in CF and if chronic colonisation with gram negative infection with *Pseudomonas aeruginosa* differs between patients (table 1).

***Table 1*. Infectious disease in children with CF at the last admissions for 2 years**

Girl, 8 years old - F508del/F508del	**Boy, 8 years old - F508del/W1282X**	**Boy, 14 years old - F508del/F508del**
Pseudomonas aeruginosa 10^5	*Pseudomonas aeruginosa* 10^5	*Kl. pneumoniae* 10^5
BNGN 10^6	*Staphylococcus aureus* 10^6	*Corynebacterium spp* 10^5
Flavobacillus spp 10^4	*Candida albicans* 10^3	*Staphylococcus aureus* 10^4
Staphylococcus aureus 10^6		
Candida albicans 10^3		
Candida glabrata 10^3		

In every child there was at least one mutation F508del. The same CFTR mutation in homosigous state F508del/F508del was found in the girl with the most severe form of CF and in the boy with controlled form of the disease (with rare hospitalisation). In the 8 years old boy who has a more severe variant of the disease was found the F508del/W1282X genotype.

There is presented the result of an international study Geborek A., et al. 2011 , where the authors find the role of CFTR genotype upon the ailing phenotype of the patients with CF (table 2, 3). This way heterozygous mutations F508del are more severe than homozygous ones [6]. Patients with 2 mutations of the first class had inferior pulmonary function (FEV_1 și FVC) than the ones who had a combination between mutations of the first and second class, or two mutations of the second class.

In conclusion, patients with CF who have 2 mutations of the first class risc to develop a more severe pulmonary disease than the patients who have at least one mutation of the second class [6].

The results of the patients involved in our study show us the fact that the genotype CFTR F508del/F508del leads to a severe pulmonary disease if associated with chronic colonizaton with *Pseudomonas aeruginosa*. With all these said, in a study based on Register of Cystic Fibrosis HuiChuan J. Lai et al. 2004 compared with the "homozygous Δ*F508*" group, "severe genotype other than homozygous Δ*F508*" was associated with a significantly lower risk of shortened survival, whereas the "mild genotype" group had lower risks of shortened survival and *P. aeruginosa* acquisition (table 4). No differences in the risk of having poor lung function were found among all genotype groups [7].

Table 2. **The different CFTR mutations according to functional class** [6]

Class I	Class II	Class III	Class IV	Class V
1717-1 G- > A	F508del	G551D	297 C- > A	2789 + 5 G- > A
3659delC	S945L	R560T	R117C	3849 + 10 kb C > T
394delTT			R347P	A455E
R553X			T 3381	3849 + 10 kb C-T
621 + 1 G- > T				
E60X				
G542X				
W79R				
W1282X				

Table 3. **The different CFTR mutations combinations of class I and class II mutations** [6]

Class I/class I		*Class I/class II*		*Class II/class II*	
1717-1 G- > A/1717-1 G->A	n = 1	3659delC/S945L	n = 1	F508del/F508del	n = 165
3659delC/3659delC	n = 5	3659delC/F508del	n = 23	F508del/S945L	n = 5
3659delC/394delTT	n = 7	394delTT/F508del	n = 38		
394delTT/394delTT	n = 4	621 + 1 G->T/F508del	n = 6		
R553X/E60X	n = 1	E60X/F508del	n = 4		
		G542X/F508del	n = 1		
		R553X/F508del	n = 2		
		W79R/F508del	n = 1		
		W1282X/F508del	n = 1		
		1717-1 G->A/F508del	n = 1		
Total	**18**		**78**		**170**

***Table 4.* Associations of baseline risk factors with survival and lung disease outcomes in US patients reported to the 1986–2000 Cystic Fibrosis Foundation Patient Registry** [7]

Baseline risk factor	Survival (*n* = 13,690)		FEV_1† <70% (*n* = 3,320)		*Ps. aeruginosa* acquisition (*n* = 5,290)	
	OR†	95% CI†	OR	95% CI	OR	95% CI
Diagnostic group (compared with SCREEN†)						
MI†	2.59**	1.28, 5.25	1.18	0.78, 1.80	1.18*	1.01, 1.36
FH†	2.21*	1.06, 4.61	0.99	0.57, 1.73	1.25*	1.01, 1.56
SYMPTOM†	2.63**	1.31, 5.29	1.15	0.77, 1.71	1.13	0.98, 1.30
Female sex	1.27***	1.14, 1.41	1.11	0.95, 1.28	1.10**	1.02, 1.17
Genotype (compared with Δ*F508*/Δ*F508*)						
Severe genotype other than Δ*F508*/Δ*F508*‡	0.76***	0.67, 0.86	0.88	0.74, 1.05	1.03	0.95, 1.11
Mild genotype§	0.51***	0.37, 0.70	1.16	0.55, 1.33	0.65*	0.42, 1.00

* $p \le 0.05$; ** $p \le 0.01$; *** $p \le 0.001$.
† FEV_1, forced expiratory volume in 1 second; OR, odds ratio; CI, confidence interval; SCREEN, group of patients without meconium ileus and identified via prenatal/neonatal screening; MI, group of patients with meconium ileus; FH, group of patients with a positive family history without symptoms; SYMPTOM, group of patients with symptoms other than meconium ileus.
‡ Includes Δ*F508*/non-Δ*F508*-I, II, III and non-Δ*F508*-I, II, III/non-Δ*F508*-I, II, III; refer to table 1 for specific mutations categorized to classes I–V.
§ Includes Δ*F508*/IV, V; non-Δ*F508*-I, II, III/IV, V; and IV, V/IV, V. See table 1 for specific mutations categorized to classes I–V.

Patients presented in our study who have CF have the advantage of a health care system, which works well, with the same treatment in the country. However, the difference regarding the adjustment for colonisation with *Pseudomonas aeruginosa* is significant. In addition to it, in the protocol of the study, all the patients were evaluated in the same clinical condition, during a exacerbation, which required hospitalization for medication, this way standartizing the investigation even more. According to the study chronic infection with *Pseudomonas aeruginosa* affects the evolution of the disease in presented children. It seems that socio-economical status of the pacients is less likely to affect the severity of the disease because of the medical and social assistance of the medical system, which fully supports the costs for the medication, as well as health care.

Homozygous patients for the F508del mutation have a wide variety of phenotypic variations [1]. Another posibility is the aspect related to the environment, such as chronic infection in combination with modifying genes or by itself, may contribute to progressive evolution with aggravation. According to Navarro H., 2002 study, pancreatic insufficiency and worse nutritional state were common for patients with F508del mutation. Respitarory implication was variable, both for heterozygous and homozygous patients. Another severe mutations, such as W128X and G542X, determined clinical manifestation similar to those found in F508del mutation [11].

Another study reveals that the average age of establishing the diagnosis of CF is 34 months, and the majority of the patients (73%) had pancreatic insufficiency during the evolution of the disease. The presence of exocrine pancreatic insufficiency was significant in the group of homozygous patients with F508del mutation, also the death index was high for the group of people with this CFTR mutation [4].

The character of expectorations in children with CF is related to the bacteriological spectre of the pulmonary infections : pyogenic germs determines production of abundant bronchial secretions, purulent ones, with fetid sputum in infectious exacerbation. It was determined thoracic deformation (emphysematous thorax, dorsal kyphosis) in 8 years old girl, which is a clinical expression of a severe broncho-pulmonary pathological process. In time progressive respiratory insufficiency develops, chronic severe persistent xypoxy determined the formation of digital hypocratism, wich is less observed in other two children.

Radiological pulmonary explorations (fig. 1, 2, 3) of the patients with CF show signs of bronchitis, broncho-obstructive syndrome, thoracic distension with emphysema, pulmonary hypertransparence expressed more in apical segments, segmentary opacities, regions of consolidation, alveolar opacities, bronchiectasis “in bouquet” (superior right lobe is frequently affected).

Separate description of latest radiological films of these children detects significantly different affection in them. A more severe affection is seen in girl's homozygous affection *F508del* (fig. 1) and boy's heterozygous mutation *F508del* (fig. 2), also having chronic infection with *P. aeruginosa*.

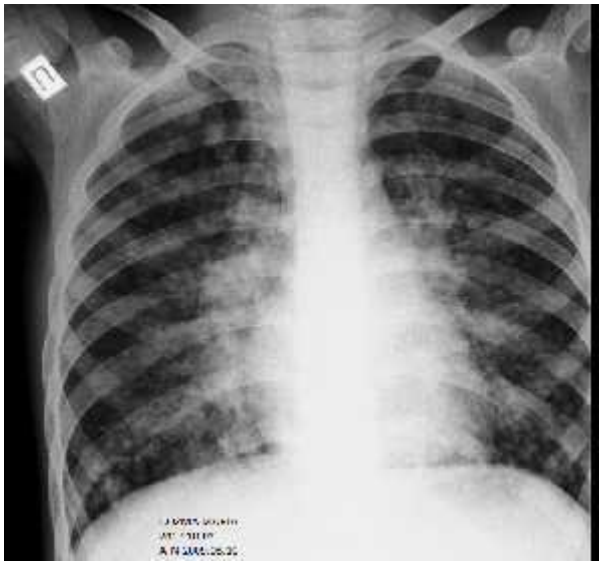

Fig.1 Thoracic Rx (2.10.2017) – aerated lungs. Homogenous hili, dilated on the background of enlarged lymph nodes. Pulmonary pattern presents cystic distensions of different dimensions. Mediobasal on the right – aeric bronchogram. CTI 0,42. Sinuses are clear.

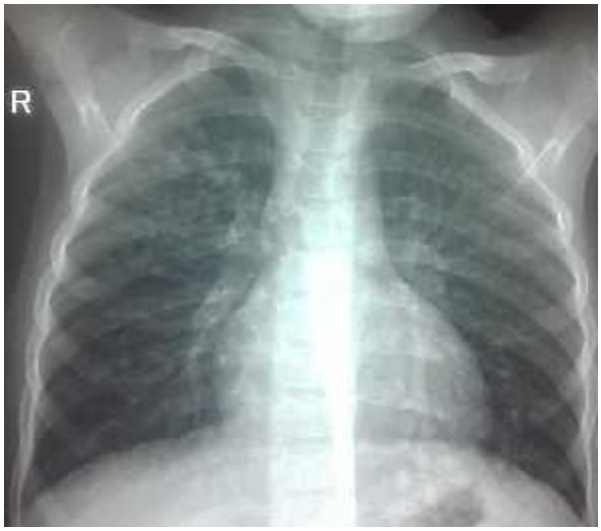

Fig. 2. Thoracic Rx (05-04-2017) – patchy pneumatised lungs. Fibrotic pulmonary pattern, distorted apically on the right and basal on the left. Enlarged homogenous hili. There are determined cystic heteromorphic deformations, heterogenous with erased contour, from 2.0-2.4 to 2.5-2.9 apically on the right S3 and basal on the left S5, S10. Mediobasal on the right and basal on the left – pneumonic infiltrations. CTI 0,44. Sinuses are clear.

There are seen irreversible bronchopulmonary modification in boy with homozygous mutation *F508del* (fig. 3), but modifications are less expressed and there is no chronic infection with *P.aeruginosa* which favors a better quality of life.

Fig. 3. Thoracic Rx (1.12.2017) – fibrotic, endured and deformed pulmonary pattern. Cylindric and varicous expansions of the bronchi, predominant in superior pulmonary areas. Thick-walled bronchi. Pulmonary hili are structured. Infiltrative and nodular opacities are not seen. "In drop" cord . CTI 0,42. Sinuses are clear

Computed tomography examination (CT) of the thoracic cavity (fig. 4, 5, 6) is a particular and highly informative method for assessment of bronchopulmonary changes in CF patients, which reveals signs of bronchopulmonary alterations in these children. Important CT findings are evident in a girl with a homozygous mutation in *F508del* (fig. 4). There are bilateral diffuse distributed bronchiectasis (cylindrical and varicose), of moderate grade (bronchi size increased 2-3 times compared to adjacent vessel), predominantly affecting upper lobes, associated with moderate peribronchial thickening. Intraluminal mucus deposits are detected bilateral in segmental and subsegmental bronchi. Centrilobular lung opacity of Y- or V-shaped structure represents mucus deposits in terminal bronchioles, which is suggestive for bronchiolitis. There are ground glass opacities with bilateral perihilar distribution. Multiple pleuropulmonary adhesions bilaterally distributed (predominantly in upper lobes). Diffuse accentuated vascular pattern. Moderate bilateral hilar

enlargement. Minimal bilateral pleural effusion. Multiple mediastinal lymph nodes (paracaval, subcarinal), ovoid and round in shape, homogeneous structure, well-defined contour, soft-tissue density (+35+40 Uh), without calcification.

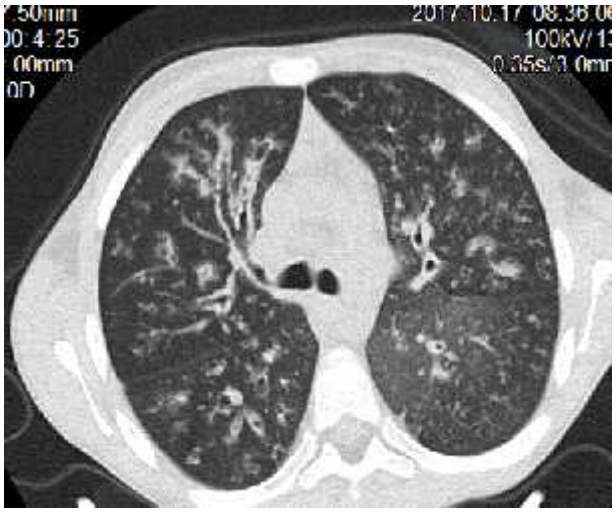

Fig. 4. *F508del/F508del*. CT of the thoracic cavity (17.10.17) bronchopulmonary changes with cylindrical and varicose bronchiectasis, bronchiolitis. Mediastinal lymphadenopathy, probably reactive. Minimal left pleural effusion.

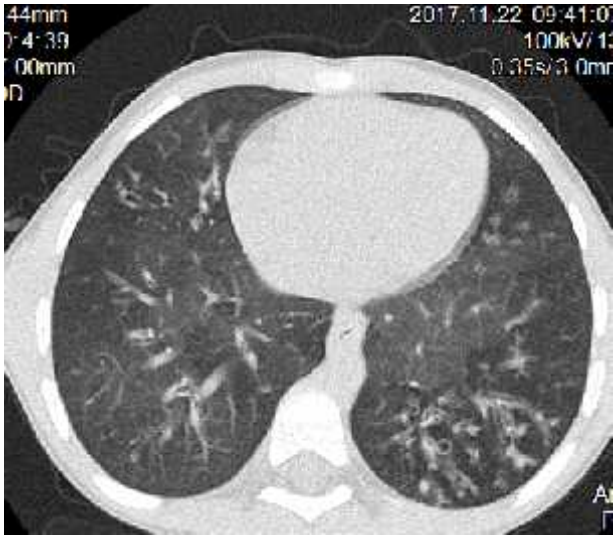

Fig. 5. *F508del/W1282X*. CT of the thoracic cavity (22.11.2017) cylindrical and varicose bronchiectasis in S_1, S_2, S_4, S_5, S_6 of the right lung and in S_5, S_9, S_{10} of the left lung. Minimal left pleural effusion..

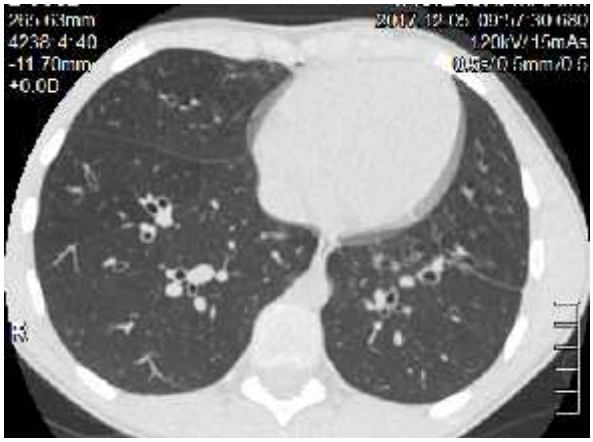

Fig. 6. *F508del/F508del.* CT of the thoracic cavity (14.01.2016) broncho-pulmonary changes in the context of moderate grade cystic fibrosis, in exacerbation. Bronchiolitis. Pulmonary infiltration with atelectatic component in the right lung S5

CT of the thoracic cavity (fig. 5) in a boy with a heterozygous mutation in F508del reveals following bronchopulmonary changes: bilateral bronchiectasis (cylindrical and varicose) distributed in S_1, S_2, S_4, S_5 and S_6 of the right lobe, and S_5, S_9, S_{10} of the left lung, of moderate grade (bronchi size increased 2-3 times compared to adjacent vessel). Moderate peribronchial thickening. Few pleuropulmonary adhesions with bilateral distributions. Minimal bilateral pleural effusion. Diffuse accentuated vascular pattern. Calcification of the arterial ligament. Moderate bilateral hilar enlargement. Irreversible bronchopulmonary changes, with less important CT findings (fig. 6) compared to the previously described patients, in a boy with a homozygous mutation in F508del are limited to bronchiectasis (predominantly cylindrical, several varicose) distributed diffuse in both lung fields, of moderate grade (bronchi size increased 2-3 times compared to adjacent vessel). Minimal peribronchial thickening (bronchial wall thickness is equivalent to the adjacent vessel diameter). Intraluminal mucus deposits are detected bilateral in segmental and subsegmental bronchi. Centrilobular lung opacity of Y- or V-shaped structure represent mucus deposits in terminal bronchioles, which is suggestive for bronchiolitis. Pulmonary infiltration with atelectatic component in the right lung S_5. Multiple pleuropulmonary adhesions distributed bilateral in apical and basal segments. Diffuse accentuated vascular pattern; moderate bilateral hilar enlargement.

A big potential in medication of patients with CF has the implementation of neonatal screening programme, and a double-check programme of diagnosis in screening children in precocious states of the disease for preventing pleuro-pulmonary complications, improvement of the prognostic and lifespan.

Conclusions

1. Pulmonary involvement in infectious episodes with aggresive germs in children of the same age with CFTR F508del/F508del and F508del/non-F508del genotypes determines bronchopulmonary chronicity. Progressive evolution of pathologic pulmonary process in these children was determined by such resistant germs as *P. aeruginosa*, *S.aureus*, which accelerates the destructive process of lung parenchyma and contributes to the extension of pulmonary fibrosis and formation of bronchiectasis

2. Patients with severe mutations of cystic fibrosis present a serious respiratory, pancreatic and nutritional involvement. The frequence and severity of clinical manifestations of bronchopulmonary system is correlated with F508del genotype.

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Research Article

Morphopathological aspects of uncomplicated omphalocele in new-borns

Babuci S.[1], **Negru I.**[1, 2], **Petrovici V.**[2]

Laboratory of surgical infections in children of NicolaeTestemitanu SUMPh[1]
Department of clinical morphopathology of PMSI Institute of Mother and Child[2]

Abstract

Aspecte morfopatologice în omfalocelul necomplicat la nou-născuți

Scopul studiului constă în elucidarea particularităților morfostructurale și morfofuncționale ale peretelui și zonei regionale a omfalocelului necomplicat în estimarea prognosticului evoluției complicațiilor locale și alegerea tacticii de tratament.
În lotul de studiu au fost incluși 18 nou-născuți din nașteri la termen de gestație de 37-40 săptămâni, cu masa ponderală la naștere 2850-3780 g, dintre care 4 (22,2 %) cu omfalocel major și 14 (77,8 %) cu omfalocel minor și mediu necomplicat, predominând formele de dimensiuni medii. Examinării morfologice au fost supuse regiunea limitrofă la nivelul peretelui abdominal, peretele și regiunea bolții omfalocelului și sectoarele din cordonul ombilical adiacent. Material pentru examinările histomorfologice au servit probele tisulare (1,0×1,0×0,5 cm) prelevate intraoperator și din piesele anatomo-chirurgicale din zonele menționare ale omfalocelului, în medie 6-7 probe. La etapa de colorare s-au utilizat metodele: *hematoxilină-eozină* (H&E) şi selectiv *Van Gieson* (VG), pentru evidenţierea țesutului conjunctiv, și *Azur-Eozină* (A&E), pentru testarea prezenţei florei micotico-bacteriene.
În funcție de zonele cercetate ale omfalocelului și țesuturile din zona limitrofă, inclusiv în bioptatele musculaturii abdominale, a fost observată o variație structurală a componentei tisulare și vascularizării. În probele prelevate din vecinătatea zonei omfalo-abdominală, începând cu regiunea limitrofă cu structurile tisulare abdominale (nivelul de rezecţie), în peretele omfalocelului în limitele a 1,5 -2 cm, pe unele arii până la 3 cm, spre bolta sau zona apicală a omfalocelului au fost depistate structuri mozaice asemănătoare arhitectural cu cele abdominale, zona internă *muco-conjunctiv-vascularizată* fiind bogată în fibre și dotată cu o rețea vasculară mult mai dezvoltată.
Rezultatele studiului a permis de a conchide că structura morfologică a omfalocelului reprezintă o membrană muco-conjunctiv-epitelială dimorfă, determinată de componenta mezotelio-fibro-epidermală, cu prezența țesuturilor predecesorii fibro-musculare și vasculare mixte sanguino-limfatice și folicular limfoide protectoare locale, care în perioada extrauterină au un impact semnificativ în evoluția proceselor reparative. În omfalocelul necomplicat există unii factori de risc major în evoluția rupturilor spontane, proceselor inflamatorii prin contaminarea complexului placentar, riscul sindromului tromboembolic placento-fetal și în peretele omfalocelului în perioada intranatală cu repercusiuni evidente hemoragice, neonatal precoce, la nivel de omfalocel. Au fost identificate unele particularități etiopatogenetice ce confirmă parțial ipoteza înaintată de unii autori că omfalocelul se dezvoltă secundar unor dereglări de migrare și de maturizare a mioblastelor în peretele abdominal.
CONCLUZII. În omfalocelul de dimensiuni majore necomplicat, cu disproporție viscero-abdominală semnificativă, ținând cont de structura arhitecturală a sacului, există condiții favorabile pentru procesele reparative, cu epitelizarea treptată a sacului și transformarea lui în hernie ventrală. Rata scăzută a letalității în lotul studiat justifică conduita conservativă în formele date ale malformației. În afectarea intraamnională există riscul de penetrare a componentului infecțios-inflamator a omfalocelului, îndeosebi în cazurile rezolvate cu întârziere, cu o posibilă contaminare postnatală.

Cuvinte cheie: omfalocel, disproporție viscero-abdominală, hernie ventrală, investigații morfologice

Abstract

The purpose of the study was to elucidate the morphostructural and morphofunctional features of the wall and regional area of uncomplicated omphalocele in estimating the prognosis of evolution of local complications and the choice of treatment tactics.

The study group included 18 new-born babies, 37- 40 weeks of gestation, with birth weight 2850-3780 g, of which 4 (22.2%) with major omphalocele and 14 (77, 8%) with minor and medium uncomplicated omphalocele, predominantly medium-sized one.

The morphological examination was performed on the adjacent region of the abdominal wall, the omphalocele domewall and the adjacent umbilical cord areas.Tissue samples (1.0×1.0×0.5 cm) of anatomical-surgical specimens taken intraoperatively from the mentioned omphalocele areas, on average 6-7 samples, were used as material for histomorphological examinations.

At the staining stage the following methods were used: *hematoxylin-eosin* (H & E) and selectively *Van Gieson* (VG) to highlight the connective tissue, and *Azur-Eosin* (A & E) to test the presence of mycotic-bacterial flora.

Depending on the investigated areas of the omphalocele and the tissues in the adjacent area, including abdominal muscle biopsy, a structural variation of the tissue and vascularization component was observed. In the samples taken nearby the omphalo-abdominal region, starting with the region adjacent to the abdominal tissue structures (resection level), in the omphalocele wall within 1.5-2 cm, in some areas up to 3 cm, towards the domor apical area of omphalocele, mosaic structures similar architecturally to the abdominal ones were found, the internal muco-connective-vascularized area being rich in fiber and supplied with a much more developed vascular network.

The results of the study allowed to conclude that the morphological structure of omphalocele represents a dimorphic muco-connective-epithelial membrane determined by a mesothelial-fibro-epidermal component has been established, with the presence of the predecessor fibro-muscular tissue and mixed vascular blood-lymphatic and protective local follicular-lymphoid, tissues which during the extrauterine period have a significant impact on the evolution of reparative processes.In the uncomplicated omphalocele there are some major risk factors in the development of spontaneous ruptures, inflammatory processes by placental complex contamination, the risk of thromboembolic placental-fetal syndrome during the intranatal period with obvious hemorrhagic and early neonatal repercussions on the omphalocele level.Some etiopathogenetic features have been identified which partially confirm the hypothesis advanced by some authors that omphalocele develops secondary to migration and maturation disorders of myoblasts in the abdominal wall.

Thus, in the uncomplicated major omphalocel, with significant viscero-abdominal disproportion, taking into account the architectural structure of the sac, there are favorable conditions for reparative processes, with the gradual epithelization of the sac and its transformation into ventral hernia. The low rate of lethality in the studied group justifies conservative behavior in the given form of malformation. In intra-amnional involvement there is a risk of penetration of the infectious-inflammatory component of the omphalocele, especially in delayed cases, with possible postnatal contamination.

Keywords: omphalocele, viscero-abdominal disproportion, ventral hernia, morphological examination

Correspondence to: E-mail: ionnegru@yahoo.com; mob. +37361055112

Introduction

Omphalocele (*exomphalos*) represents a medial defect of the anterior abdominal wall at the base of the umbilical cord, covered by an amnio-peritoneal membranous sac, through which the abdominal organs are extruded [1, 23]. The omphalocele dimensions and location have important implications in the therapeutic conduct of this malformation. The defect size ranges from 2 cm to 12 cm, and the location can be centered on the upper, middle or lower abdomen [13, 17]. The membranous sac, on which the umbilical cord is implanted, consists of the amnion, Wharton`s jelly and peritoneum [16, 27]. The omphalocele contents include: intestinal loops, often liver portion and occasionally other organs, such as the spleen [18].

The type of omphalocele is determined by the embryonic fold involved in the pathological process. Thus, epigastric omphalocele resulting from the cephalic envelope defects, was found in the pentalogy of Cantrell (epigastric omphalocele, anterior diaphragmatic defect, sternal defect, pericardial defect, and associated intracardiac defects). The lateral fold defects cause the development of the *"classic"*omphalocele, while the caudal fold defects cause hypogastric omphalocele, bladder and cloacal exstrophy.

Omphalocele may be a part of various genetic syndromes, indicating that this malformation does not have a specific etiology [8, 9, 17]. Animal studies, as well as human gene research, have highlighted several genes incriminated in the development of this defect, including:Rock-1, Pitx2, IGFR-1, FGFR1 and FGFR2, FOXA2 and Hoxb4, Alx-4, MSX1 etc. [19]. Familial cases of omphalocele have also been described [21].

The management of omphalocele, which aims at closing the defect, depends mainly on its dimensions [2, 7], the reduction of abdominal viscera, the presence of complications, the degree of pulmonary and cardiovascular insufficiency, associated anomalies [10,20].

Practice has shown that only in 30% -50% of cases the defect can be primarily closed, in most cases alternative correction strategies being needed [5].

In the literature special attention is paid to surgical techniques based on the macroanatomic peculiarities of omphalocele and less on the structural and morpho-functional ones, including the adjacent umbilical-abdominal area, being important in the medical-surgical management of this condition, which justifies the approach of this issue in the present study.

The purpose of the study was to elucidate the morphostructural and morphofunctional features of the wall and regional area of uncomplicated omphalocele in estimating the prognosis of evolution of local complications and the choice of treatment tactics.

Material and Methods

The study group included 18 new-born babies, 37-40 weeks of gestation, with birth weight 2850-3780 g, of which 4 (22.2%) with major omphalocele and 14 (77, 8%) with minor and medium uncomplicated omphalocele, predominantly medium-sized one.

The morphological examination was performed on the adjacent region of the abdominal wall, theomphalocele domewall and the adjacent umbilical cord areas (fig. 1). In 5 (27.8%) cases,the placental complexes were examined morphoanatomically. Tissue samples (1.0×1.0×0.5 cm) of anatomical-surgical specimens taken intraoperatively from the mentioned omphalocele areas, on average 6-7 samples,were used as material for histomorphological examinations.In 7 (38.9%) cases, the intraoperative abdominal muscle biopsy (0.3x0.6 cm) was taken. There were taken 5-6 samples from the placental complexes, including annexes.

The tissue samples were preserved in Formol 10% for 6-12 hours, then processed according to the standard protocol for histological investigations with the automatic histoprocessor (Diapath) and the staining network (Raffaello) of the histomorphological tests. At the staining stage the following methods were used: *hematoxylin-eosin* (H & E) and selectively *Van Gieson* (VG) to highlight the connective tissue, and *Azur-Eosin* (A & E) to test the presence of mycotic-bacterial flora. The histological examination was performed using the following microscopes: Nikon Labophot-2 and Carl Zeiss with *oculars× 10, lenses × 2.5; × 10; × 20; × 40.*

Results and Discussions

The omphalocele diameter varied between 3.5 cm and 12 cm, having a non-uniform translucent wall (fig. 1). In 11 (61.1%) cases, the presence of the vascular network, radially or spirally traced in the wall area was confirmed, more frequently at the domeand adjacent to the proximal cutaneous resection area. In 7 (38.9%) cases, the vascular network was more compact, embedded in a gelatinous cord (proper umbilical cord), with increased density, with or without vascular varicosities, passing continuously into the umbilical cord. In the basal area, the omphalocele wall is unevenly thickened, matte or whitish. Some vessels, along the trajectory, have a purple shade of varying intensity due to the gelatinous tissue, including the umbilical cord in the omphalocele area.

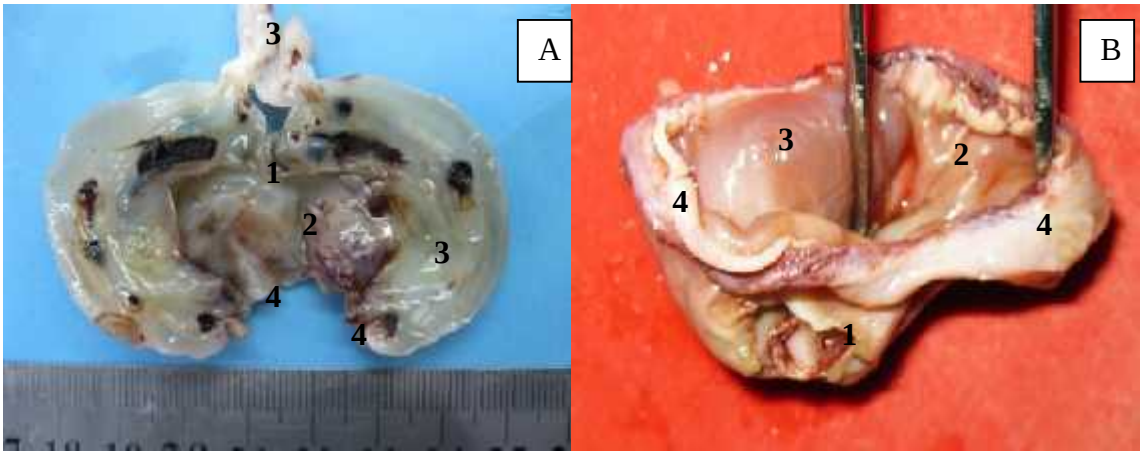

Fig. 1. Anatomo-surgical specimens.Excisionedomphalocele.Hernialsac - omphalocele: 1 - apical-umbilical area; 2 - gelatinous membranous wall; 3 - umbilical cord in the hernial wall area; 4 – adjacent cutaneous area

Another characteristic of omphalocele, found in 4 (22.2%) cases, was the presence of adhesion structures. In one case, a partial herniation of an intestinal segment through the double orifice was determined through the adhesion. This phenomenon, in our opinion, deserves attention because it reflects the pathogenesis of omphalocele and indicates the preexistence of a major risk of strangulation of the mentioned intestinal segment.

The membraneous wall of omphalocele had a gelatinous consistency due to the predominance of the fundamental extracellular substance compared to the cellular one, the fibrillary component being less present. From the outside, omphalocele was circumscribed by amniotic or pavemental epithelium, andunistratified directed to the adjacent area. The inner omphalocele surface, in all cases and throughout the whole area, is represented by mesothelium. Thus, there are two layers in the omphalocele structure: *external muco-connective*, avascular, lax, where the fundamental extracellular substance prevails, and *internal muco-connective-vascularized*, much more compact and hypercellularized, with pseudofibrillary aspect and the presence of the vascular component (fig. 2A). In the umbilical cord only the muco-connective tissue is present with a dispersed discrete cellular component (fig. 2B). In the samples taken from the omphaloceledome, the internal area is much more diminished, sometimes merging with the epithelial structures of the mesothelium and amnion, determining itsdimorphic muco-connective-epithelial membranous structure. Depending on the investigated areas of the omphalocele and the tissues in the adjacent area, including abdominal muscle biopsy, a structural variation of the tissue and vascularization component was observed. In the samples taken from the omphalo-abdominal proximal region, starting with the region adjacent to the abdominal tissue structures (resection level), in the omphalocele wall within 1.5-2 cm, in some areas up to 3 cm, towards the domeor apical area of omphalocele, mosaic structures architecturally similar to the abdominal ones were found, the internal muco-connective-vascularized area being rich in fiber and supplied with a much more developed vascular network.

Microarhitecturally, closely to the defect, between the muco-fibro-vascular and muco-connective components myocytic and fibrotic elements were found, being dispersed chaotically and ordered in bundles, forming a lax fibro-muscular layer to the omphalocele dome. At the median level, the fibrillary-myocytic component was more orderly, on some areas with fascicular appearance. Later on, the presence of a comparatively dense fibromuscular fascicular layer, predominantly of connective-fibrillary or fascicular origin, was found. Throughout the internal area, mesothelium, regardless of its volume, was present.A lax connective tissue layer supplied with an abundant vascular network was present towards the periphery (fig. 3A). The gelatinous component was circumscribed towards the periphery, in different proportions, by fibrocytic-fibroblastic cellular elements, among which small capillaries and angioblasts could be found. Externally, a squamous pavemental epithelial layer was found, which, towards the omphaloceleapex, transformed into a pluristratified or unistratified layer of amniocytes.

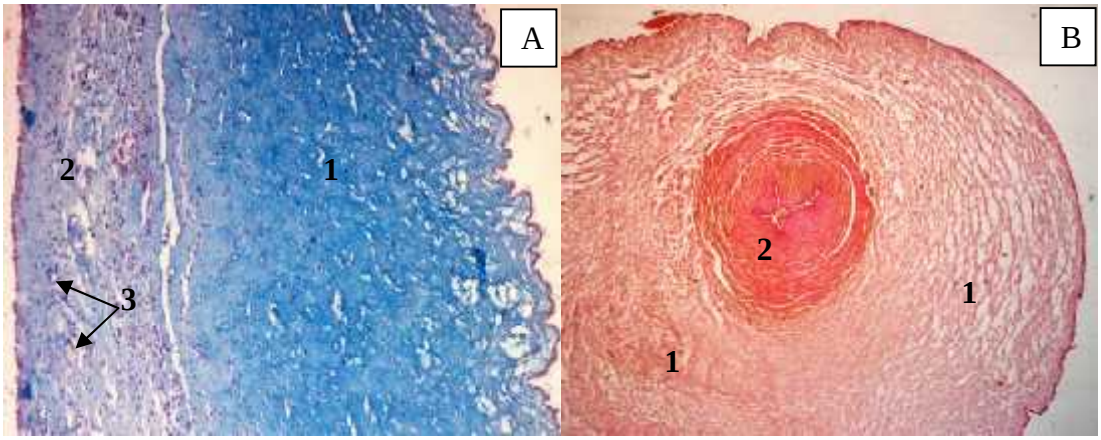

Fig. 2. Muco-connective component in omphalocele and umbilical cord. A-omphalocele: 1-external muco-connective area; 2 - internal muco-connective-vascular area; 3 - venous capillaries × 75. *Alcian blue staining*; B - umbilical cord: 1- muco-connective component; 2 - umbilical artery × 25. *Hematoxylin-Eosin Staining*

Vascular structures, especially those in the umbilical cord, exhibited dysplastic processes of the vascular tunica muscularis. The musculo-fibro-vascular component sometimes had a chaotic fibro-cellular appearance, consisting of fibrocytes, myocytes, and mucocytes with small gelatinous accumulations, including the area adjacent to the omphalo-abdominal aperture (fig. 3B).

In other areas,the deficiency of fibro-cellular and fibrocytic-muscular components with ordered or lax aspect, especially at the apical level of the omphalocelewas observed, as well asaltered and hydropic degenerations, with the formation of serous accumulations(fig. 3C). In 8 (44.4%) cases, nodular and follicular lymphoid structures (fig. 3D) were found in the orifice of the hernial sac (omphalocele), arranged in chain on the perimeter of the orifice, sometimes in the gelatinous area, in the form of lymphocyte depletion of sinusoid type. Lymphoid structures could also be observed throughout the vascular network, with a varied depletion of the lymphocyte component.

In some cases the presence of amniotic adhesions was found in omphalocele on the internal surface, with connective-vascular mesothelium-coated cords and supplied with small and medium capillary and vascular network, sometimes with small lymphocytic pseudo-follicular structures (fig. 4A). In 2 cases, solitary myocytes were detected. Adhesion changes point to another omphalocele genesis, which differentiates it ontogenetically from the umbilical hernia itself. The presence of lymphoid structures at this level may, in our opinion, can be the consequence of the amnio-liquid content action that can penetrate the gelatinous layer, as well as the lymphoid-cellular barrier function of protecting the eventrated intestinal loops or the abdominal cavity.

In the exploration of the abdominal muscle area in the immediate vicinity of the omphalocelic orifice, an obvious disruption of the muscular connective and cellular-adipose tissue components manifested by an amorphous and irregularly structured appearance was confirmed (fig. 4B, C, D). Muscle tissue is composed of bundles of hypoplastic and hypertrophied muscle fibers that are oriented disorderly between themselves and in relation with cell-adipose tissue and fibrillary connective tissue (fig. 4B). In some samples, a correlation between identical tissues is preserved except for the hypotrophy of myocytes (fig. 4C), some areas being more hypotrophic or as myocyte islets, embedded in the connective tissue mass (fig. 4D).

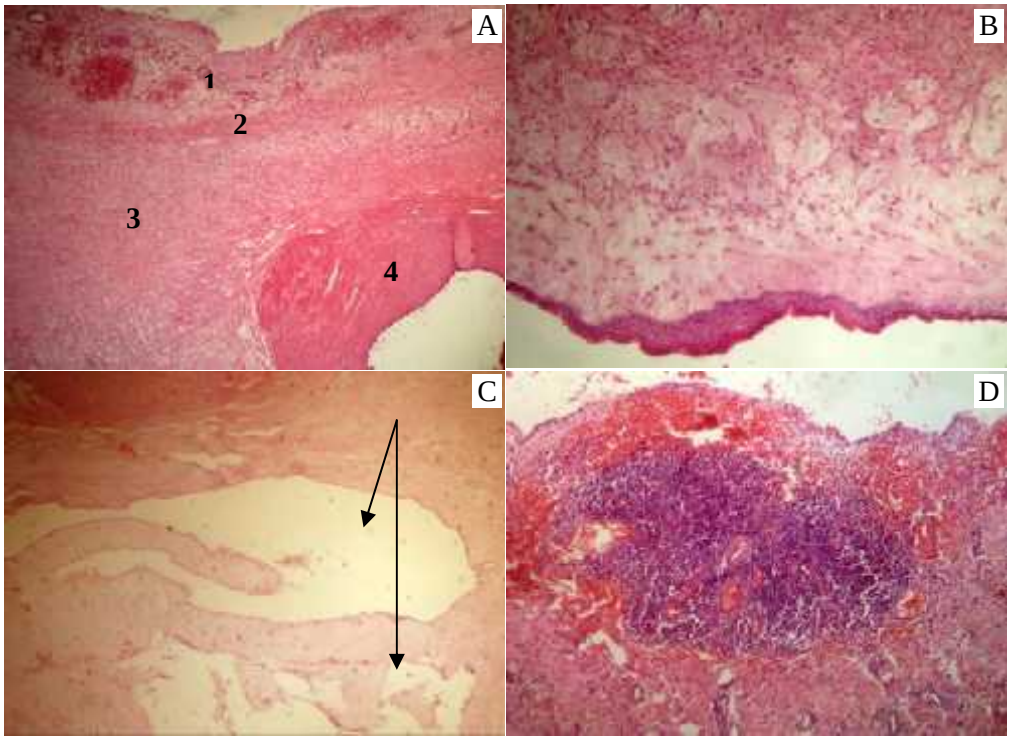

Fig. 3. The overall histopathological appearance of the omphalocele. A - internal structure of the omphalocele wall: 1 - vascular network; 2 - fibromuscular layer; 3 - gelatinous substance; 4 - umbilical vessel with the tunica muscularisdysplasia; B - Peripheral area of the omphalocelic wall: 1 cellularized gelatinous mass with fibrocytic-fibroblastic cells; 2 - multilayered squamous pavemental epithelium; C - gelatinous area with hydropic degeneration with serum fluid accumulation; D - follicular lymphocellular structures in the area adjacent to the resection.

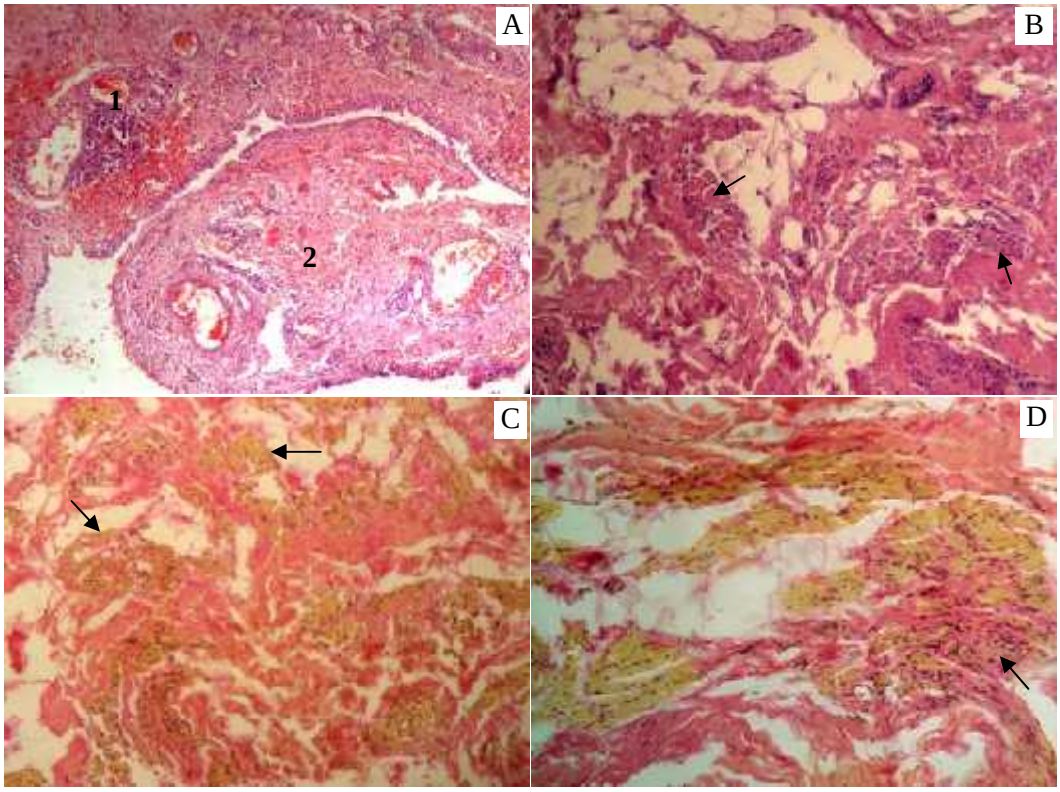

Fig. 4. Histopathological aspects of the abdominal muscle component A - internal structure of the omphalocele wall: 1 - fibro-vascular adhesion with vascular network coated with mesothelium; 2 - the perivascular lymphocytic pseudo-follicle in the omphalocele wall in the adjacent area × 25; B - disordered muscular bundles with a hypertrophic appearance in the fibrillar-connective and cellular-adipose tissue component. × 75; C - slightly ordered abdominal tissue plate with hypertrophied muscle fibers × 75; D - islets of muscle fibers and myocytes with intumescence aspectand atrophy of connective tissue masses. Colors H-E (A, B) and VG (C, D)

In 7 (38.9%) cases the presence of an inflammatory process in omphalocele structures was found (fig. 5A, B), especially in cases where the inflammatory processes involved the placental complex structures. The inflammatory process had different intensity, especially in the middle and apical areas of the omphalocele, being more pronounced in cases resolved surgically 3-5 days after birth. A pronounced inflammatory process was concomitantly detected on the internal side, being less pronounced in intermediate or superficial areas. In these cases, a visceral intestinal peritoneum reaction could also be observed.

At the same time, vascular dysplasia was confirmed both in the umbilical cord area included in omphalocele and in its branches. In 5,6% (1) of cases, vascular aneurysm was detected, which ruptured during the first hours after birth (fig. 5C), the muco-fibrillary component being invaded with red blood cells masses, resembling hemorrhagic accumulations (fig. 5D).

Histological examinations of placental complexes in neonates with omphalocele (10 cases) allowed changes to be observed in both the villous chorion and the amniotic membranes.

In 27.8% (5) of cases there were maturation changes of the villous chorion corresponding to the term of gestation, minimal adaptive compensatory processes, predominantly in the presence of nonspecific inflammatory lesions, confirmed in 16.7% (2) of the cases. Calcification foci, marked fibrosis and dilated-varicose angiopathy characterized by small and large varicosities, especially of venous vessels, including truncal villosities (fig. 6A, B), have been reported. Some aneurysms of impressive size, compared to villosity, and very thin walls, advocate for vascular dysplasia that occurs at the embryonic or embryonic-fetal stage (fig. 6B).

In placental angiopathies there was found the presence of thromboembolism in the lumen of venous vessels, indicating the risk of a placento-fetal thromboembolic syndrome and the evolution of vascular thromboembolic lesions in the omphalocele (fig. 6B). Following the exploration of the vascular network of omphalocele and the placental complexes, there was found an omphalo-umbilical and villous chorion vascular dysplasia, i.e. the fetal vascular network. Therefore, its evolution begins at the stage of the fetal concept and continues until the first 3-5 weeks of intrauterine development, which corresponds to the embryonic period when the blood circulation installs with repercussions, especially on the placental-allantoid one.

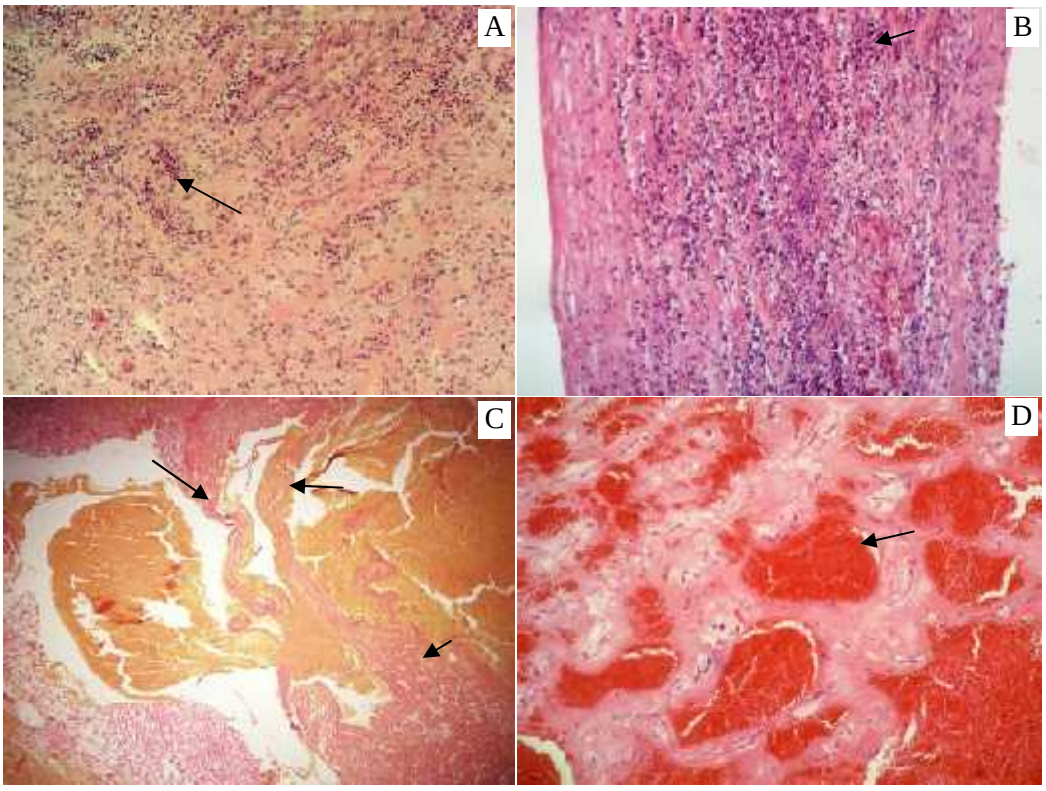

Fig. 5. The appearance of pre-existing lesions, accidentally evolving in omphalocele. A - microfocal and dispersed lympho-leukocytic inflammatory process in the proximal area of the resection × 50; B - micro-abscessed leukocytic inflammatory process in the middle area of the omphalocele wall × 75; C - ruptured aneurysm with the invasion of the musculo-fibrillary component of omphalocele. D - blood accumulations in the muco-fibrillary tissue of the omphalocelic umbilical segment. × 75. H-E staining (A, B, D); VG (C)

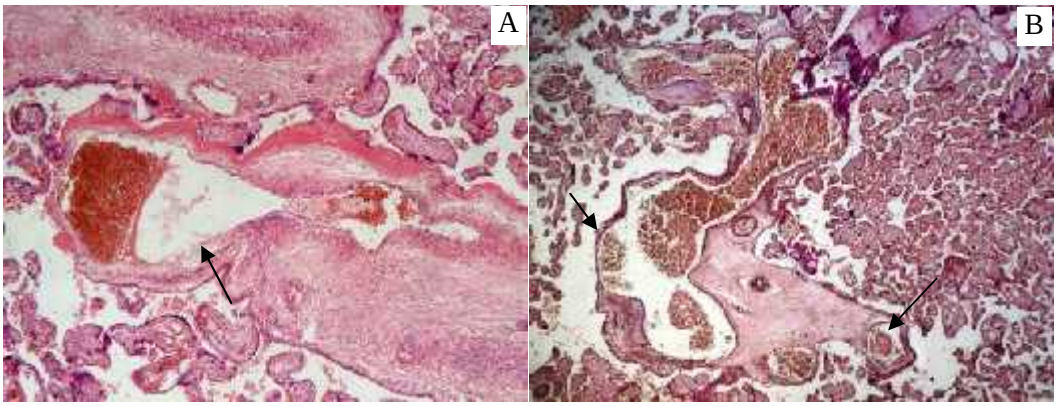

Fig. 6. Histopathological aspects of placental villous chorion vascularization in omphalocele. A - varicose dysplasia of the venous villous truncal vessel × 25; B - Varicose veins of a villous chorion tree with the presence of thromboembolus in the lumen × 25. *H-E staining.*

At the level of the villous chorion, a delayed discronism was identified with aspects of deficiency of the stromal connective component of the intermediate and terminal villosities, which determined their moderate monstrous appearance (fig. 7B). At this level, the syncytial epithelium was attenuated, poorly differentiated and with less proliferation in the form of buds. This could suggest a correlation between reduced cellular-syncytial activity and deficiency of connective density of the villosity stroma.

In some areas, varicose ectasia (fig. 7C) involved the entire venous vascular network of terminal, intermediate and truncal villosities. This phenomenon could be the consequence of hypertension in the venous trajectory, possibly, also at the level of the great vessels of the placental chorio-amnional plaque and umbilical cord.

In 8 cases there were some changes in the villous chorion arterial network, manifested by hypertrophic-stenotic arterial angiopathies in the truncal and intermediate villosities, unevenly zonally or segmentally distributed (fig. 7D). At the level of the basal decidua membrane, lymphocyte infiltration, more frequently focal one, of variable intensity, was determined, ranging from discrete to moderate. Concomitantly with the inflammatory process, significant fibrinoid deposition was detected at this level in the inter-villous spaces (fig. 7A). The morphopathological changes of the membranes comprised discrete and moderate micro-macrofocal leukocyte infiltration, in some areas associated with edema and amnion dystrophy (fig. 7B).

In the case of an amniotic adhesion, the amniotic epithelium manifested an active proliferation of the amniocytes, more pronounced in the membranes and at the level of the amniotic adhesion (fig. 7C).

On the basis of the revealed findings, we can conclude that, in the case of omphalocele, in the placental complexes there is a dysplasia of the villous chorion associated with varicose arterio-venous angiopathy and hypertrophic-stenotic arterial angiopathy. Vascular alterations found at this level, in our opinion, may be present from the very beginning along with the deficiency of the stromal connective component, or may be a consequence of omphalocele. Depending on the size, it may induce the hypertensive phenomenon in the umbilical vein with reflection on the chorionic vascularization, causing the hypertrophic spasm of the arterial network.

At present, according to histomorphological features and etiopathogenesis, omphalocele is a hernial sac in the omphalo-umbilical abdominal segment. However, over time, this malformation has been treated as an umbilical cord hernia in the structure of umbilical cord malformations or the abdominal supra-umbilical region caused by lack of aponeurosis and herniation of the peritoneal membrane at the umbilical fossa level.

The earliest description of omphalocele belongs to Ambroise Parè [15] and the first success in its surgical treatment were recorded by Hey (1803) and Hamilton (1806). Scarpa (1812) drew attention to the association of omphalocele with other malformations, which determines the fatal prognosis of omphalocele. Ahlfeld (1899), applying alcohol dressings on the hernial sac, was the first to treat omphalocele without surgery [15, 24]. At the moment, theetiopathogenesis of omphalocele is not fully elucidated [14].

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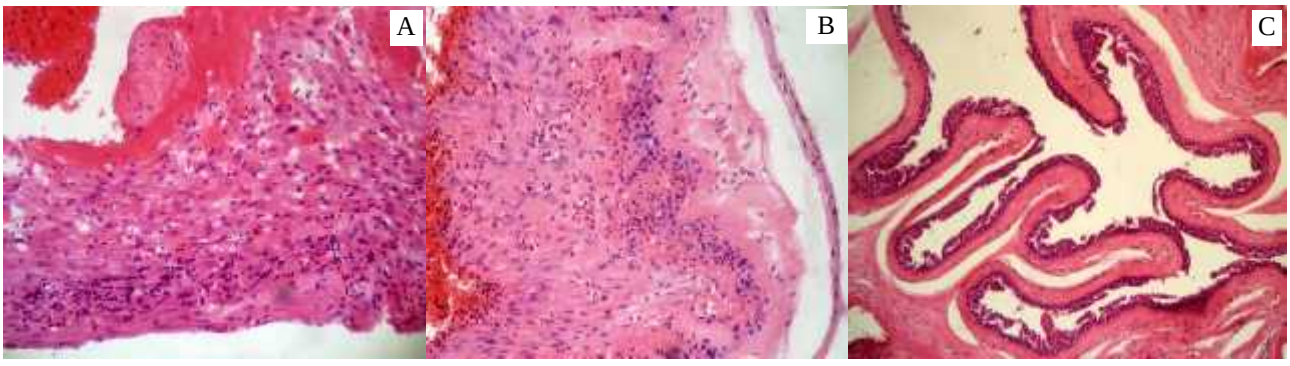

Fig. 7. A - moderate lymphocyte basal deciduitis in the focus. B – amniotic membrane, focal serous- leukocyte membranitis× 25. C - Papillary proliferation of amniocytes at membrane level. × 100. H & E staining

According to the study results, omphalocele is a vicious defect, triggered in the embryonic period in the abdominal wall, especially the omphalo-umbilical region. The deficiency and structural disturbances of the abdominal wall muscles in the areas underlying the omphalo-umbilical orifice as well as functional muscular dischronism serve as evidence. The latter is characterized by muscle fiber atrophy and hypertrophy, muscle fragmentation and disordered muscle fiber orientation in relation to fibrillar-connective tissue and cellular-adipose tissue, which causes a considerable muscle deficiency at this level.

Determination of the muscular, fibrillar-connective and epidermal-epithelial elements in omphalocele contradicts the assertions that omphalocele includes only the peritoneum elements and the amniotic membrane of the umbilical cord. The presence of epidermal fibro-muscular elements and vascularization may accelerate reparative and epithelialization processes of the muco-fibro-epithelial sac of omphalocele

Based on the results obtained, we can conclude that omphalocele is not a simple umbilical hernia, which is not a serious problem for pediatric surgery, but a defect of the abdominal muscles separation in the omphalo-umbilical region. The insignificant visual defects in the orifice are associated with major regional abdominal disturbances of the rectilinear musculature, being a problem in the medical-surgical management of this malformation and in the application of the umbilical cord ligature.

In this regard, some researchers believe that the midgut disturbances of return into the abdominal cavity underlie the malformation development during the embryonic period, after the normal embryonic herniation in the umbilical cord in the 6th-12th week of the intrauterine development, and the central migration of the mesodermal fold [12, 25, 26]. The liver does not migrate out of the abdominal cavity and, therefore will not be present in the physiological hernia contents. However, some authors state that if the lateral folds do not close, then there is a major defectof the abdominal wall, through which the abdominal cavity contents, including the liver, will herniate [11].

According to some studies, the differentiation of the abdominal wall muscles simultaneously occurs with the process of intestine rotation and return of the intestinal loops into the abdominal cavity. The above mentioned plays a central role in the development of omphalocele [6]. The primary abdominal wall is made up of ectoderm and the lateral mesodermal plate(somatopleura) which extends laterally and coalesces along the ventral medial line around the navel, forming the abdominal cavity. Myoblasts, migrating from the myotome to the primary wall, form secondary structures such as muscles and connective tissues. Thus, when the secondary abdominal development is completed, the abdominal wall is composed of four pairs of muscles (*oblique externe, oblique interne, transvesus abdominae and rectus abdominae*), surrounding connective tissues and skin. The orientation of the myofibrils in a particular muscle is unidirectional and distinct from the adjacent ipsilateral muscles and symmetrical to the muscle pair on the contralateral side [19, 22]. Omphalocele results from staged developmental disturbances of the secondary abdominal wall, caused by the failure of the migration process of myoblasts to thedestination point, with the alteration of spatial relations between the oblique and transverse muscles [19]. In giant omphalocele there are two distinct correction strategies, including surgical closure of the abdominal defect after multiple operations and delayed nonoperative closure by measures that contribute to the sac epithelialization [4, 5].

Soave F. (1963) considered that prognosis in omphalocele, especially in giant forms, could be improved if the conservative treatment were considered by surgeons with more confidence [24]. Currently, this strategy remains a safe and effective alternative to the treatment of omphalocele [3].

Conclusions

1. The morphological structure of omphalocele as a dimorphic muco-connective-epithelial membrane determined by the mesothelial-fibro-epidermal component has been established, with the presence of the predecessor fibro-muscular tissue and mixed vascular blood-lymphatic and protective local follicular-lymphoid, which during the extrauterine period have a significant impact on the evolution of reparative processes.
2. The presence of major risk factors of uncomplicated omphalocelein the evolution of spontaneous rupture, inflammatory processes by placental complex contamination, the risk of thromboembolic placental-fetal syndrome and in the omphalocele wall during the intranatal period with obvious hemorrhagicand early neonatal repercussions at the omphalocele level.
3. In 22.2% of cases, anatomic abnormalities of omphalocele, determined by adhesion aspects and adhesions, have been found, which can contribute to the strangulation of loops, herniated organs with irreversible complications, especially the intestinal segments.
4. Some etiopathogenetic features have been identified which partially confirm the hypothesis advanced by some authors [19] that omphalocele develops secondary to migration and maturation disorders of myoblastsin the abdominal wall.
5. In major uncomplicated omphalocele, with significant viscero-abdominal disproportion,

taking into account the architectural structure of the sac, there are favorable conditions for reparative processes, with gradual epithelization of the sac and its transformation into ventral hernia. The low lethality rate in the studied group justifies the conservative behavior in the given forms of malformation.

6. In intra-amnional involvement, there is a risk of the infectious-inflammatory component to penetrate omphalocele, especially in cases that are resolved with delay, with a possible postnatal contamination.

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Reviews

Contemporary concepts in reconstructive surgery of the diaphragmatic defects

Eremia V.

Laboratory of surgical infections in children of NicolaeTestemitanu SUMPh

Abstract

Concepte contemporane în chirurgia reconstructivă a defectelor diafragmatice

Defectele diafragmatice congenitale şi dobândite continuă să fie o problemă majoră în chirurgia pediatrică. În ultimii ani, tratamentul chirurgical al defectelor diafragmatice congenitale a evoluat de la o abordare urgentă la intervenții chirurgicale amânate, efectuate după stabilizarea stării copilului. Autorul descrie mai multe opinii conform cărora intervențiile chirurgicale de urgență rămân preferabile în herniile Bochdalek sau herniile diafragmatice majore asociate cu alte afecțiuni patologice: volvul gastric acut, perforația stomacului cu pneumotorax, peritonită perforativă, etc. În articol sunt descrise avantajele şi dezavantajele a diferitor aborduri deschise (thoracic, abdominal), strategiile de tratament miniinvaziv, cât şi diverse procedeie tehnice de închidere a defectelor diafragmatice. Autorul conchide, că deşi au fost înregistrate unele rezultate promițătoare în tratamentul herniilor şi eventrațiilor diafragmatice congenitale, tratamentul consensusal al acestor malformații rămâne eluziv, dovezile clinice în sprijinul mai multor modalități terapeutice fiind limitate, impunându-se necesitatea unei evaluări atente cu estimarea realistă a potențialelor riscuri.

Cuvinte cheie: defecte diafragmatice, hernie diafragmatică, eventrație diafragmatică, tratament chirurgical, grefe biologice

Abstract

Congenital and acquired diaphragmatic defects continue to be a major problem in pediatric surgery.Last years, the surgical treatment of congenital diaphragmatic defects has evolved from an urgent approach to delayed surgery after the child's condition has been stabilized. The author describes the opinion of several authors that emergency operations remain preferable in Bochdalek hernias or major diaphragmatic hernia associated with other pathological conditions: acute gastric volvulus, perforation of the stomach with pneumothorax, peritonitis, etc.
There are described the advantages and disadvantages of thoracic and abdominal approach, the miniinvasive treatment strategies and the techniques of diaphragmatic defect closure. The author concludes that although some promising results have been recorded in the treatment of diaphragmatic hernias and eventrations, the consensus treatment of these malformations remains elusive, clinical evidence supporting several therapeutic modalities being limited, requiring a careful assessment with the realistic estimation of potential risks.

Keywords: diaphragmatic defects, diaphragmatic hernias, diaphragmatic eventration, surgical treatment, biological patch

Correspondence to: e-mail : eremiavictor1@gmail.com; mob. +373 69346613

Diaphragmatic defect correction resulting from trauma or congenital origin remains a surgical problem, a major cause of morbidity and mortality in Pediatric Surgery [68, 95].

Over the past few years, the surgical treatment of congenital diaphragmatic defect evaluated significantly from emergency operation until the delayed intervention with the elective diaphragmatic defect, made after the stabilization of the condition of the child and the cardio-respiratory functions [81, 119, 125]. Surgical intervention strategy "delayed" in association with a gentle ventilator and, occasionally, with use of ECMO support, guarantees the best results [58, 125]. Emergency surgical interventions are preferred only Bochdalek hernias or major diaphragmatic eventrations, associated with other pathologic: acute gastric volvulus, perforation of the stomach with antenatal development pneumotorax and perforative peritonitis, etc. [55].

The purpose of preoperative diaphragmatic defect is "stabilizing" the patient: acceptable oxygenation (PaO2 > 40 mmHg) and CO2 (with arterial pressure growing < 60 mmHg) pulmonary pressure (50% of the pressure < systemic) [124].

The objectives of the operations have in diaphragm defects surgery aimed at reducing herniated content with closing the defect, thus resulting in the correction of the disfunction of ventilation and lung protection [25]. In this context, the choice of the technical process depends on several factors, the size of the defect, precise anatomical details or type directly influencing its prognosis of these malformations [1, 110].

Surgical treatment of diaphragmatic defect can be done through an open thoracic or abdominal approach [127, 137], and with the miniinvasive treatment strategies [139]. In most cases (91%) is applied to an abdominal subcostal incision [25, 67, 128]. Some authors consider that a midline longitudinal laparotomy is a good approach for exposing enough of diaphragmatic defect [131]. Laparotomy offers several advantages in comparison with open thoracotomy approach through: reducing easier viscera from the thoracic cavity, the possibility of mobilizing the posterior edge of the diaphragm, intestinal rotation malformations and avoidance of musculoskeletal posttoracotomic sequelae [127].

Transtoracic appproach in diaphragmatic hernia, was promoted by Truesdale P.E. (1921) and Everett Koop and Johnson J. (1952), it was considered beneficial by the rapidity of the thoracic cavity drain, especially in children with respiratory difficulties, and through better exposure of the diaphragm, subsequently challenged Gross R.E. (1953) [137]. Some authors consider that the thoracotomic approach is the best in posttraumatic diaphragmatic hernias, laparotomy being preferred in patients with abdominal injuries [37]. The literature is described and a combined abdomino-thoracic approach to diaphragmatic hernias [120].

Miniinvasive surgical techniques, both laparoscopic [57, 60] and thoracoscopic [32, 83,108], are considered safe alternative approaches in reconstruction of the diaphragmatic defect [33]. Clinical efficiency of endoscopic techniques in the treatment of diaphragmatic defect is comparable with open surgical intervention that could be the favorite in the treatment of these pathological states [76].

Thoracoscopic approach in congenital diaphragmatic defect was used for the first time in 1995 by Cristian M.L. et al. and represents a feasible and effective option in patients with diaphragm defect [92], providing the advantage of facilitating the diagnosis and treatment of pulmonary lesions associated with [75], being useful in cases of recurrence [61]. According to some studies, the thoracoscopic approach in congenital diaphragmatic hernias has a significantly higher rate of relapses, but also surviving rate [66, 128]. Thoracoscopic method is recommended and the diaphragmatic defects acquired, because it avoids many potential difficulties existing in transabdominal approaches [65].

Morgagni hernia correction surgery is performed immediately after diagnosis, even in asymptomatic forms, due to the potentially fatal complications, such as strangulation and volvulus, intestinal obstruction, ischemia and necrosis [36, 72, 93]. It has not reached a consensus concerning the abdominal approach. Achieved through upper midline incision or upper transverse abdominal approach allows easily and control access to herniate simultaneously repairing bilateral hernias, diagnosis and correction [6].

One of the scope of surgical intervention in congenital diaphragmatic defects is the excision of the hernial sac which remains a controversial issue [4, 10, 78]. While some authors argue the necessity of its excision because of the risk of pneumomediastinum, deterioration of mediastinal structures and cardio-respiratory complications, others would maintain in situ with good results [10, 14, 82, 118].

Opinions were divided, and with regard to how the closing of the hernia defect: either by using a primary repair with sutures or applying a continuous or nonresorbable interrupted, either through the use of prosthetic material. Transabdominal approach with absorbable interrupted sutures remain the preferred option [6, 14], including with the use of laparoscopic techniques [129].

Intrapericardiale congenital hernias can be solved by simple surgical primary closure and use of prosthetic patch, in clear diagnosis to recourse to the abdominal approach. In chronic cases, when there are endorsements between liver, pericardium, heart, the thoracic approach seems more secure. It is recommended, and the questionable diagnosis, because the intrapericardial

formations, such as cysts, teratomas and others can be removed in this way [15, 55].

Surgical treatment of congenital diaphragmatic defect closure aimed at them by bringing the edges and their fixation with absorbable interrupted sutures [25,102]. Clinical practice has shown that the major-sized diaphragm defects or hemidiaphragm agenesis in reconstruction methods with the application of direct suture are less effective [84].

Using perfused muscle flaps has certain advantages in the diaphragmatic reconstruction tension free, but not in primary repair [80, 90, 122].

Simpson J.S. and Gossage J.D. (1971) in congenital diaphragmatic hernias with major dimensions, through the subcostal approaches, used a pediculate muscle flap from anterior abdominal wall, which consists of the internal oblique muscle and transverse abdominal, which was sutured stump at the edges of the defect, achieving a satisfactory closure without significant deformation of the coast or abdominal wall. According to some opinions, this technique allows large diaphragmatic defect reconstruction tension free, without a potential risk of infection [20], the risk of relapse being comparable to that of the primary reconstruction and significantly lower than when using the patch [12].

According to the communication, using only the transverse abdominal muscle is a safe and proper technique in repairing congenital diaphragmatic major defects [8]. Bianchi A. et al. (1983), in reconstruction of recurrent diaphragmatic hernias or hemidiaphragm agenesis, has proposed using the flap from m. latissimus dorsi, the main advantage being safe feeding blood vessels with perforators. Some authors recommend that combination of m. latissimus dorsi with anterior m. serratus anterior fascia for this intervention, patients being selected with caution [109, 116].

Flap of the fascia Toldt, also called Gerota fascia consisting of diaphragmatic muscle of medial remenescence, peritoneum and fascia Toldt, connective tissue retroperitoneal space, can be a way of solving the major diaphragm defects. [34, 94].

Implantation of autologous tissue is quite complicated and invasive, requiring a long time with a high risk for bleeding, necrosis and other severe complications [138].

In major diaphragmatic defects, multiple authors indicate the use of bioprotetical patches, there is controversy regarding the type of material (or synthetic), expansion or non-resorbable, which depend directly on the results of treatment. In the literature are offered several types of prosthetic materials, without being identified as ideal [13].

Currently, the use of synthetic patch sites is the method of choice in the diaphragm defects reconstruction of major dimensions [87]. For the first time, diaphragmatic nylon patch was used experimentally by Adler and R.H. and FirmeC.N in 1957.Subsequently to this they have been tested on animals and other synthetic materials, including: Teflon by Harrison J. (1957), Ivalon by Cooley J.C. et al., (1957), Dacron by Dalton M.L. et al., (1966).

Currently they proposed several prosthetic synthetic materials in surgical treatment of diaphragmatic defect, their choice being random. The most common absorbable prosthetic materials used in reconstructive surgery, including diaphragmatic, are polypropylene (Brenda), polytetrafluoroethylene (Gore-Tex), expanded polytetrafluoroethylene (composite mesh with two faces), and place of polyethylene terephthalate (Dacron) [47, 62, 112].

Polytetrafluoroethylene is the most common biosynthetic material used in reconstructive surgery (81%) [132] including diaphragmatic [13]. This polymer formed by nature inert monofilament, produces an unstressed inflammatory response, it does not integrate the diaphragmatic tissue but not being elastic, does not ensure adequate diaphragmatic movements [38].

Synthetic patches of this material do not grow with the patient, repair processes are determined by the growing edge of tissue that can ensure diaphragmatic scope and its adaptation. If these processes do not occur, the patch as child rearing, can ward off the chest wall or edge with the development of recurrence, or chest wall may choose the synthetic material, patch deformation, causing chest wall [105].

Although the synthetic prostheses materials are durable, with angiogenesis properties can be easily modeled after the defect requiring diaphragmatic dissection and mobilization of tissues and minimum time of preparation, they remain in situ for life and constitute a microbial colonization potential, instead of requiring review at certain time periods [16,127].

For over three decades experience of use of synthetic patch showed that the main disadvantage of the method is increased recurrence rate, which can reach up to 50% of cases [86, 121, 126].

To minimize the risk of recurrence in diaphragm defects must be taken into account different aspects: the type and shape of the implant, suture material used, surgical technique, the patient type and comorbidities the dimensions of the diaphragmatic defect [121].

Recurrences can be bimodal, early in the first few months after surgery, and late that develops a few years later. Early recurrences are determined, most likely adhesions, inadequate or scar tissue it in large defects, with small portions of muscle.

Synthetic implants tend to decrease with diaphragmatic scarring, which leads to the development of late recurences in rising child requiring technical processes of reconstruction [127]. Some authors, in order to enable the extension in time, suggested fitting synthetic net in the shape of a Double Cone fixed

(double-sided) [77]. It was described and the use of synthetic patch used at all combined with two faces, made of Gore-Tex, on the one hand Marlex [104]. Regarding the surgical technique, several authors emphasize how the application of sutures and fixation of the prosthetic patches, eventually crossing the sutures around the ribs with intercostal muscles, involvement being used, typically, thread that is not absorbed [35, 83, 107].

While initially a pediculate muscle flaps and use of prosthetic synthetic absorbable materials seemed a good solution, in many studies, there has been an increased incidence of late complications, including: intestinal obstructions, need splenectomy, of the chest wall and abdominal deformities [97, 127]. According to some studies, propylene is causing most of the grips, their formation being related to pore size. The macroporous polypropylene mesh facilitates adhesion formation rather than the expanded polytetrafluoroethylene microporous [45, 91]. In children operate for major diaphragm defects, scoliosis was found in 4%-50% of cases and in 14% deformity of pectus-80%, no statistical difference observed between these two ways of surgical correction [90, 106].

The use of biological materials in the repair of congenital and acquired diaphragmatic defects is limited to small series and case reports [5]. Janes R.M. (1931) proposed the use of fascia lata graft in the treatment of post-traumatic diaphragmatic hernia, and in 1968 the use of this graft was reported in the surgical treatment of hiatal hernia by transthoracic approach, this method remaining current at present [19], including in the treatment of recurrent congenital diaphragmatic hernias [121]. Unique cases of use of human dura mater grafts, fascia lata and bovine peritoneum in diaphragmatic defects are reported [11, 50, 77, 103].

At present, considerable efforts are being made to develop alternative ways of closing both, congenital and acquired diaphragmatic defects [35, 113]. Progress in stem cell biology and tissue engineering has led to the creation of more bioengineering tissues that can be incorporated into host tissues by regenerating natural tissues with the ability to grow with the patient [28, 56]. The decellularization process allows the removal of resistant cells from donor tissues by special technologies, thus obtaining an extracellular three-dimensional matrix while preserving the native biochemical architecture including the maintenance of microvascular networks, which can be reclaimed with new progenitor or composite cells. The drastic reduction in DNA amount, significant cell depletion while preserving several properties of the treated tissue is a crucial outcome in decelularization methods, with the aim of avoiding any immune rejection of the graft [74, 98].

In the past decades, several biological grafts have been created, some of which are approved in clinical practice, including human cadaveric dermatitis, submucosa of the pig's small intestine, bovine pericardium. These materials have successfully demonstrated the ability to support human tissue repair [18]. There have also been described several acellular biological grafts as an option in the reconstruction of diaphragmatic defects [127], among which Alloderm [23], Surgisis [48], Permacol [86].

The human acellular dermal matrix is an allogeneic material obtained from human cadaveric skin (AlloDerm; LifeCell Corporation, Branchburg, NJ) with the special removal of cellular components of the dermis and epidermis. Several studies have found that the Alloderm graft exhibits biomechanical properties comparable to those of the abdominal wall or synthetic mesh fascia, and its early revascularization increases resistance to infection and contamination. The advantages of this material against synthetic prosthetic materials are: development of a minimal adhesion process, remodeling with autologous vascular tissue and resistance to infections [9].

Surgisis (SIS, Cook Biotech Inc., Cook Deutschland GmbH, Monchengladbach) is an extracellular matrix of porcine acellular intestinal submucosa, which is a type 1 collagen structure with active growth factors which after implantation undergoes degradation processes and remodeling and collagen replacement of the host [38].

Permacol is an acellular chemically-bonded dermal derived porcelain collagen material that produces an unprompted inflammatory response consistent with the normal wound healing process that allows for good incorporation. Welding of lisin and hydroxylisin residues from Permacol's collagen fibers gives the material greater resistance to collagenation and higher durability, and is considered by some authors to be a safe alternative to synthetic nets [86, 102].

In order to overcome the disadvantages and limitations of known prosthetic materials and to create optimal materials for closing the diaphragmatic defects, the feasibility of the use of cross linked porous collagen grafts obtained by tissue engineering techniques and composite or hybrid meshes resulting from the combination of several prosthetic materials, including polyester/collagen grafts [132], poly (ε-caprolactone)/collagen [138], a combination of several prosthetic materials, including polyester/Vicryl/ Colagen [21]. The results obtained are at the dizerate stage.

In severe forms of congenital diaphragmatic hernia, closure of the abdominal wall after the reconstruction of the diaphragmatic defect can be quite difficult due to a potential inconsistency between the volume of the viscera, which must be reduced, and the size of the abdomen, insufficiently developed [68]. Loss of pulmonary compliance may be due to an internal defect of the lung parenchyma or restricted mobility of the tho-

racic and abdominal cavity. Restrictions induced by these two sources can cause significant reduction in respiratory volume, causing worsening of adequate ventilation capacity of the diaphragmatic hernia. In this context, some authors hypothesized that applying a silo bag to the abdominal wall would greatly reduce the decline in abdominal wall compliance, associated with the primary closure of the abdominal wall after repairing the diaphragmatic defect [100].

With regard to abdominal wall enlargement techniques, there are reports of delayed closure of the abdomen after reconstructive interventions in children with congenital diaphragmatic defects [68, 111]. The phenomenon can be determined by the potential consequences of abdominal cavity reduction after the diaphragmatic defect closure, namely the sudden increase in intra-abdominal pressure and the development of compartment syndrome [81].

Chylothorax is a potentially severe complication after diaphragmatic hernia reconstruction, with an incidence of 5.5% -28% [73, 85]. This complication generates nutritional deficiency, hypoproteinemia, electrolyte disturbances and compromises immunity [69]. Although the exact etiology of chylothorax in the diaphragmatic hernia remains unknown, several theories have been proposed, including: lymphatic vessels dividing into hernia sac, direct traumatic lesion of diaphragmatic lymphatic vessels, and pressure phenomenon of visceral lymphomas resulting in small rupture of the thoracic canal [85]. Effective management of this complication continues to be a challenge, the initial treatment consisting of tube thoracostomy and total parenteral nutrition. The effectiveness of using a continuous perfusion with octreotide, as adjunctive therapy, remains controversial. The surgical treatment of chylothorax is indicated after a period of 1-3 weeks of conservative therapeutic measures, or in cases where more than 15 ml/kg of fluid is eliminated through the thoracic drainage [39, 69]. Numerous aspects of surgical treatment in congenital diaphragmatic defects are still questionable, especially the criteria for selecting surgical techniques [125] and the increased recurrence rate, which varies between 3% and 50% [99]. These patients frequently complain of vomiting, intestinal obstruction and abdominal pain (58%) or pulmonary pain (17%) [49].

Indications for surgical treatment in diaphragmatic events are: pulmonary compression with tachypnea without any improvement after conservative treatment, cardiac compression with arrhythmias, two or more recurrent pneumonia, severe life-threatening pneumonia which endangers the patient's life with failure to disconnect from mechanical ventilation, respiratory distress associated with paradoxical diaphragm movements [71, 135]. Surgical treatment of diaphragmatic eventration in children is indicated not only in symptomatic patients [40], but also to seemingly asymptomatic patients with major disorders of ipsilateral pulmonary function, found in the ventilation and infusion assessment [134, 135]. Diaphragmatic eventration associated with spontaneous diaphragm rupture [43], intestinal obstruction [3], symptomatic gastric volvulus, or suspected perforations are surgical emergencies where the most frequent recurrence to a subcostal or transversal abdominal incision. This approach makes it possible not only diaphragm bladder, but also anterior gastric fixation by gastropexia or gastroductomy [79, 117].

Regardless of the congenital or acquired origin, the classic thoracic approach through an incision in the intercostal space 7 or 8 is considered the optimal option in the exposure of diaphragmatic eventration [7]. The aim of surgical correction is to restore the diaphragm's topographic location, to provide the lung with a larger volume of expansion, and to restore satisfactory ventilation [41, 135]. Immobilization of the diaphragm through the envelope contributes to the reduction of the paradoxical movement and the contralateral movement of the mediastinum organs. The thoracic approach is the only possible placement of the right-handed eventration, and in the left-hand position in the central and anterior regions an abdominal approach can be used [124].

The open transabdominal approach provides access to both sides of the diaphragm and does not require selective ventilation, and laparotomy is a less morbid incision than thoracotomy [41]. The correction of diaphragmatic eventrations incidence using miniinvasive techniques, including laparoscopy [51, 133] and thoracoscopy [29, 52, 59, 63] are an acceptable alternative and along with assisted video surgeries are safe methods with satisfactory results in repairing the diaphragmatic event.

Several surgical techniques have been proposed to repair the diaphragmatic eventration, including excision and suture, diaphragmatic bladder and prosthetic patches [7]. The most commonly used technique in diaphragmatic eventration and paralysis is diaphragm epithelium [71, 101, 135], which aims to reduce dysfunctional caudal diaphragm excursion during inspiration [7]. For the first time application of this procedure to adults was described in 1923 by Morrison J.N.W. [41], although it was proposed by Wood in 1916 [71]. In children, the first diaphragmatic bladder was carried out in 1947 by Bisgard to a child with respiratory insufficiency [123].

Several intact or sectioned diaphragm processes are described [31, 130]. The envelope "in the overcoat" provides for the return of the diaphragmatic dome into tension, making a flame-shaped front flap directed towards the pleural cavity maintained at the base by a row of U-shaped non-resorbable sutures. Then the re-

tained portion is folded in front, anterior or posterior after is sutured with diaphragm with interrupted sutures, thus creating three layers superimposed on the thin part of the diaphragm [71]. The inverse inversion folding procedure involves suturing tight diaphragm edges by positioning the excess length to the peritoneal cavity. The first non-resorbable wire in surget is applied to close the depression at the periphery of the diaphragm. Subsequently, a second surget is applied [29, 88].

The central nesting technique consists in the application of several linear rows of horizontal non-resorbable sutures through the weakened part of the diaphragm. With sutures they are tightened in folds and weak tissues, creating a tense diaphragmatic surface [96].

The diaphragmatic enveloping techniques used have proven to be safe, simple and appropriate in the surgical correction of diaphragmatic eventrations incidence in children with good outcomes. They allow the reduction of most preoperative symptoms immediately after surgery and amelioration of pulmonary function [71].

Postoperative complications of diaphragmatic bladder include: atelectasis, pneumonia, pleural effusion, abdominal compartment syndrome, and spleen lesions, usually in the abdominal approach [44].

There are also unique communications for the successful use of Gore-Tex synthetic meshes in the correction of diaphragmatic eventrations [24, 53].

Despite the improvement in the survival rate and the change of the basic management paradigms, the consensual treatment of congenital diaphragmatic defects remains elusive, clinical evidence in support of several therapeutic modalities being limited. Although advances in the treatment of congenital diaphragmatic defects are quite promising, the results of a new treatment approach require a careful assessment with a realistic estimate of potential risks before the recommendation in practice [127].

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Subtalar arthroereisis in the treatment of flat-foot deformity: the pros and the cons

Korolkov O.I., Rakhman P.M.

Sytenko Institute of spine and joint pathology national Ukrainian Academy of Medical Sciences, Kharkiv, Ukraine

Abstract

Artrodeza subtalară în tratamentul deformității piciorului plat: pro și contra

Lucrarea prezintă analiza literaturii referitor la starea actuală a problemei utilizării artrodezei subtaliare în tratamentul deformității piciorului plat la copii și adulți. Sunt expuse informații despre indicații și contraindicații în acest tip de tratament și potențialele complicații.
Autorii conchid că în literatura de specialitate poate fi observată existența unor opinii contradictorii în această problemă, fiind subliniate atât eficiența acceptabilă și avantajele evidente ale acestui procedeu, cât și riscurile unor complicații care impun necesitatea intervențiilor repetate sau dezvoltarea unor stări ireversibile ale piciorului. Prezintă interes utilizarea acestei metode la copii și adulții cu afecțiuni neuromusculare, având ca obiectiv limitarea indicațiilor pentru utilizarea metodelor chirurgicale non-fiziologice.

Cuvinte cheie: artrodeză subtalară, deformitate de picior plat, tratament chirurgical

Abstract

The article presents the analysis of the literature on the current state of the problem of the use of subtalral arthroeresis in the treatment of flat foot deformity in children and adults. Information on indications and contraindications in this type of treatment and potential complications are exposed. The authors conclude that contradictory opinions can be observed in the specialized literature, emphasizing both the acceptable efficacy and the obvious advantages of this procedure, as well as the risks of complications that require the necessity of repeated interventions or the development of irreversible states of the foot.It is of interest to use this method in children and adults with neuromuscular disorders, aiming at limiting the indications for the use of non-physiological surgical methods.

Keywords: subtalral arthroeresis, flat foot deformity, surgical treatment

Correspondence to: e-mail: korolkovmd@gmail.com; mob. +380506347105

Introduction

Over the last decades there has been an increase in the number of children and adolescents with feet abnormalities. According to different authors, the plano-valgus deformity cases number equals to 35-50% of all the cases of children feet abnormalities [25, 67, 68]. It shall be noted that the plano-valgus deformity develops slowly and can be asymptomatic and painless for a long time, but in future pain and lameness may appear, as well as sensitivity disorders and trophic disturbances; feet joints mobility may be limited; arthritic phenomena may develop in the feet joints with the corresponding progression of dysfunction [25, 27, 38, 63]. There are few approaches in treatment of plano-valgus deformity:

- conservative way (used in 90-95% of cases)
- surgical treatment is advised in cases of persistent pain, progression of deformity and/or dysfunction of limbs, existence of trophic disturbances and failure of conservative treatment [25, 40, 48, 55].

One of the modern variants of surgical treatment of plano-valgus deformity of children and adults is the so-called subtalar arthroereisis solely or in combination with other types of treatment [24, 53, 59]. According to the encyclopedic dictionary of medical terms, the term "arthroereisis" (lat. athroereisis – arthron – joint + ereisis – lifting) means lifting of the joint[78]. Consequently, subtalarar throereisis is the type of surgical treatment aimed at limiting the excessive foot eversion and pronation of calcaneus while maintaining supination range of motion in the subtalar. Arthroereisis is performed by installing a special implant (of different designs, forms and materials) in the sinus tarsi.

Furthermore, all the authors emphasize that the implants for subtalararthroereisis are installed within the sinus tarsi and canalis tarsi where there is no chondral layer, i.e., this area is not articular, so the treatment is extra-articular. Sinus tarsi is an anatomical formation limited with ankle and calcaneus bones, ankle-calcaneus-navicular joint in front and rear facets of subtalar at the rear. A sinus tarsus is represented by 3 separate articular facets separated by canalis tarsi: the front and the middle facets lie distally, and the rear facet lies proximally from the canalis tarsi [73].

The term "subtalar arthroereisis" was introduced in the early XX century by Putti V., and in native medicine the term "lateral artroris" was introduced by Turner G.I. in 1930. Under this term the operation for limiting the mobility of subtalar by artificial bone barrier was meant. The method itself was originally developed for treatment of children flatfoot; however, later the indications were extended to adults. Del-Torto (Italy, 1927) was one of the first surgeons who performed arthroereisis using this type of treatment for correction of deformities in children aged 5-17 with complete or partial paraplegia (Tamoyev S.K., 2012) [49]. In 1946, Chambers E.F. announced the implementation of extra-articular subtalararthroereisis using a bone graft [8]. He tried to limit the foot eversion by removing a small portion of the anterior process of the calcaneus with bone grafting of the defect. This reduced range of motion in the subtalar and contributed to setting the foot in a functional position.

Grice D.S. (1952) used autograft from tibia bones of the patient to correct valgus at paralytic platipodia. He set the autograft in the sinus tarsi and called it an "extra-articular" subtalararthrodes, referring to integrity of articular surfaces of the subtalar. According to the literature, this operation is performed rarely due to the high risk of degenerative process in the subtalar [15].

Later various authors proposed different variants of implants for subtalararthroereisis: Vilandot [12] – used to use fibular part for arthroereisis later Subotnick S.I. (1974) described installation of inert silicone implants (elastomer) into the sinus tarsi; Smith S. (1975) first used the UHMW-polyethylene "STA-peg" implants; Valenti G. and Langford J. et al developed a helicoid structure with cylindrical shaped cutting (1987); Maxwell J. et al (1997) developed a titanium screw implant; many other scientists worked at this topic as well[13, 32, 35, 44, 59].

Study purpose: to present the current state of problem of use of subtalar arthroereisis in treatment of planovalgus deformity in children and adults, information about indications and contraindications to this type of treatment and possible complications based on analysis of the literature.

Material and Methods

During the study the data from the literature over the past 10 years were analyzed, References were made to previous works on application of subtalararthroereisis in children and adults with planovalgus deformity.

Results

The analysis of literature data shows that at present special attention is given to the biomechanical role of location of ankle bone relative to tibia and orientation of the axis of subtalar in the context of planovalgus deformity. Particularly, the concept of rotational equilibrium of foot relative to the axis of subtalar is being considered. Normally subtalar is placed obliquely, deflected from the horizontal by 42° in the sagittal plane and by 16° from the median longitudinal foot axis in the lumbar plane. Thus, the subtalar is a simple joint with the axis of rotation behaving like an "... oblique door hinge" [20, 34, 41, 69].

The purpose of subtalar arthroereisis is restoration of normal anatomic proportions in the joints of the hindfoot and optimization of static-dynamic loads pattern. Roth S. et al. claimed that at installation of an implant in subtalar the correction is achieved by stimulation of proprioception, which provides for permanent

nature of correction [41]. It is also believed that by limiting external pronation moments and increasing supination moments in subtalar not only the planovalgus deformity can be eliminated, but also the biomechanics of walking can be changed by changing the spatial location of the heel-ankle axis [46].

Considering the large number of options and different functional roles of implants for subtalararthroereisis, Vogler H.M. (1987) [51] suggested their classification:

a) implants functioning as a self-locking wedge;
b) implants altering the position and arrangement of the heel-ankle axis;
c) implants functioning due to direct compression action on the lateral process of ankle bone.

Indications for subtalar arthroereisis and additional procedures at planovalgus deformity

Most authors agree that subtalar arthroereisis alone can be considered as an option of treatment of "flexible" planovalgus deformity of I-II degree in children and adults. The results show that treatment of patients with planovalgus deformity by the method of subtalar arthroereisis is less traumatic and highly effective and promotes rapid and adequate restoration of supporting ability. However, the question of advisability of performing subtalar arthroereisis still remains. Some authors indicate that subtalar arthroereisis can be performed in patients aged 6-8, the other consider starting age to be 10-12 [10, 28, 52].

Many of the works point to the need to consider at the treatment of planovalgus deformity both the degree of deformation and its possible combination with other foot pathologies (dysfunction of posterior tibial muscle, additional navicular bone, forefoot pathology, etc.) [14, 48, 70, 74]. Based on the concomitant feet pathology combined treatment is used, including surgery on soft tissues (including achilloplastic operation, channeling of chorda of posterior tibial muscle) and bones (medialized calcaneus osteotomy, arthrodesis of talus-navicular joint, correction arthrodesis of calcaneus-cuboid joint, the so-called "+/-" osteotomy of cuboid and the 3 sphenoid bone and reconstruction of forefoot – by indications, etc.) [16, 30, 56, 77, 79].

Analysis of literature shows that in pathogenesis of planovalgus deformity, especially in adults, considerable attention is paid to dysfunction of posterior tibial muscle. In this regard a special tactic of examination was developed (clinical tests to determine the posterior tibial muscle dysfunction), classification (four-grade by Johnson K.A. and Storm D.E.) and methods of surgical treatment of this pathology [21, 39, 57, 59].

At the initial stages of widespread use of implants for subtalar arthroereisis the surgical treatment process was developed for use primarily for children, as the researchers hoped that with the child's growth the arthroereisis will help to prevent the development of secondary signs of excessive movement in hindfoot; that is, the earlier the subtalar complex becomes stable, the sooner the excessive load on the muscular system will be eliminated [2, 5, 23]. Currently most researchers agree that subtalararthroereisis in children and adults is advisable in case of persistent pain, feet dysfunction and failure of conservative treatment, but this intervention does not exclude other more traumatic and complex operations, if necessary [1, 4, 54].

Separately the discussion shall be considered which is held by supporters and opponents of subtalar arthroereisis in cases of paralytic or spastic planovalgus deformity. According to several review articles and recent thesis works it becomes clear that this problem is far from its final solution. First and foremost, this is due to the mechanistic approach to treatment of children with planovalgus deformity on the background of neurological disease and underestimation of neurophysiological mechanisms underlying the formation of feet deformities. There is also a question about the advisability of early surgery treatments with the aim of eliminating pathological settings and contractures in the lower limbs joints in connection with their possibly low efficiency and high recurrence rate [22, 33, 43, 65].

For example, Kenis V.M. in his works compared the effectiveness of planovalgus deformity treatment in children with infantile cerebral paralysis, by performing osteoplastic subtalar arthroereisis (34 patients, 56 feet) on the one hand and with subtalararthroereisis implants (46 children, 74 feet) on the other. The author comes to the conclusion that "...clinical indications for the osteoplastic arthroereisis is the pronational variant of foot deformity: children aged 5-7 with hindfootvalguse for more than 30° - as the main method; in children aged 8 - 11 with hindfootvalguse for more than 20° - as the main method. Indications for implant arthroereisis is the presence of pronation variant of deformity in children aged 5-7 with hindfootvalguse for up to 20° as the variant method"[23, 61, 66]. The author successfully used implants of various designs, including those of biodegradable material, in the treatment of mobile planovalgus deformities in children with infantile cerebral paralysis [62].

At the same time Ryzhikov V.D., 2011, in his candidate's dissertation proposed and validated diagnostic and treatment algorithm of choice of an optimal method of treatment of equinoplanovalgus deformity in children with infantile cerebral paralysis based on the patient's age, level of motor skills, severity of foot deformity, but does not even mention the subtalar arthroereisis [42].

Umnov D.V. (2010) showed that for elimination of planovalgus deformity in patients with infantile cerebral paralysis the corrective osteotomy of the calcaneus is the optimal method of surgical treatment, as is not

accompanied with a loss of function of the subtalar, in contrast to extra-articular subtalar arthrodesis operation, considering the comparable clinical and radiographic outcomes of both surgeries. The identified comparability of reconstructive capacities and efficiency of these two variants of treatment leaves, according to the author, the ability to use technically easier (in comparison with osteotomy) surgical operation in the form of extra-articular subtalar arthrodesis in a limited group of patients with overweight and questionable prospect of movement without assistance [50].

Bolotov A.V., 2015 proved that the use of minimally invasive methods of surgical treatment with use of submerged implants during the execution of subtalar arthroereisis in patients with myelodysplasticplanovalgus deformity promotes early activization of patients and improves the quality of life of patients [7]. Despite many developed methods of surgical treatment of flatfoot in children with the effects of neurological diseases, it can be stated that still need to be clarified age and clinical indications for the use of extra-articular stabilizing procedures and corrective osteotomies of the bones of the foot in children with this pathology [18, 58, 60, 64, 71, 76].

Complications and contraindications to subtalar arthroereisis

The analysis of literature data shows that the most common complications when performing subtalar arthroereisis are the following: syndrome of the ankle sinus; persistent pain in sinus tarsi; spasm of the peroneal muscles; the wrong choice of an implant leads to hypo- or overcorrection of the deformity; fracture of the subtalar implant; reaction to a foreign body; migration of the implant; limitation of motion of the foot joints [6, 17, 30, 37, 47, 75]. Rarer, but yet possible complications can be the following: superficial or deep infection; avascular necrosis, cystic changes or fracture of talus/calcaneus; synovitis, bursitis, capsulitis; need for additional operations; increasing pain in the knee and/or hip joint and lumbus [26, 45]. However, the literature data speak of a small number of certain complications and usually their description is limited to reports of several cases. In addition to it, some complications, such as complaints on pain in the operated foot during walking, do not require repeated interventions and can be corrected by the patient himself during 2-3 months after walking pattern change and normalization of load on foot or removed by local treatment and introduction of anti-inflammatory drugs in the subtalar area.

Many orthopedic surgeons stress the fact that if between the moment of setting the implant in the subtalar and the moment of possible occurrence of need of its deletion is more than 1-1.5 years, then the foot stays in the normal position, i.e., planovalgus deformity relapse does not occur [17, 43, 75].

One of the major drawbacks of this type of treatment is the existence of certain limitations in physical activity in case of presence of a subtalar implant in the title, for example jumping and contact sports activities are undesirable [32].

During the period since 1990 to 2004 De Pellegrin M. performed subtalararthroereisis in 152 children (82 boys and 70 girls) in 74 cases with bilateral pathology, in a total of 226 feet; the mean age was 10.6 +/- 1.9 years. The author obtained good results in 95.4% of cases, with complications in 4.6% of cases [11].

Contraindications to subtalar arthroereisis performing can be divided into surgical and orthopedic ones. The first group includes: age less than 8, high risk of development of suppurative processes in place of surgery and presence of specific infections in the body (tuberculosis, etc.), mental illnesses and severe somatic condition. The second group includes significant post-traumatic and congenital planovalgus deformity, rigid foot deformity with significant arthritic changes in joints and overweight [11, 26, 45, 75].

Analysis of the results of subtalar arthroereisis in treatment of planovalgus deformity

It shall be emphasized that the works on biomechanical modelling of subtalar arthroereisis with the use of implants of different design and studies of change of position of the talus and subtalar in cadavers show the change in contact stresses in subtalar and ankle joint after subtalararthroereisis. Martinelli N. et al. (2012) [31] studied fresh frozen cadaveric specimens for distribution of contractual pressure in feet joints of the foot in normal condition, at planovalgus deformity and after setting the Kalix implant in the sinus tarsi and it is shown that subtalararthroereisis restores the normal intra-articular pressure in subtalar. In a cadaver study J.C. Christensen and his colleagues found that changing the position of talus affects the location of other foot bones, which subsequently was confirmed in special X-ray studies [3, 9, 36].

Husain Z.S. and Fallat L.Y. revealed quantitative changes in the degree of restriction of movements in subtalar depending on the implant size. They found that the range of motion in subtalar was reduced by 32.0 – 44.8 – 58.0 – 65.5 and 76.8% when using implants with a diameter of 6, 8, 9, 10 and 12 mm, respectively [19]. Most of the analyzed works containing explanation of late results of subtalar arthroereisis in treatment of planovalgus deformity from a perspective of the evidentiary medicine are at the 3 and (what is more often) at the 4 level of evidence, i.e. the medium- and long-term retrospective studies of a certain group of operated patients are held, sometimes the results of subtalararthroereisis are compared in two age groups of patients, more rarely there is a comparison of results of subtalar arthroereisis with other types of operations

(such as medialized calcaneus osteotomy or Evans osteotomy) [6, 11, 47].

Loskutov O.A. in 2015 presented the results of treatment of 126 patients (204 feet) aged from 5 to 39 with planovalgus deformity and dysfunction of posterior tibial muscles using subtalar arthroereisis in the period from 2 to 5 years. Revision surgery was performed in 5 patients, removal of the implant – in 1 patient. The author came to the conclusion that subtalararthroereisis is less traumatic and highly effective and promotes rapid and adequate restoration of supporting ability [28]. According to Tamoev S.K., 117 patients aged from 18 to 32 (193 feet) in a period of 4 years after subtalar arthroereisis the following results were obtained: excellent – after 144 operations (74.61%); good – after 39 operations (20.20%); satisfactory – after 7 operations (3.62%); unsatisfactory – after 3 operations (1.55%) [49]. According to our research, De Pellegrin M. et al. had the greatest experience: since 1990 to 2012 he collected data on 485 patients who underwent subtalararthroereisis (247 –two sides arthroereisis, 238 – monolateral arthroereisis). The average age of patients was 11.5 ± 1.81 (range 5.0-17.9). Based on more than 20 years of experience, the authors believe that subtalar arthroereisis is the best method for correction of planovalgus deformity. However, they emphasize the necessity of setting precise indications for this type of treatment [11].

Conclusions. As can be seen from the above, the analysis of literature data on treatment of planovalgus deformity using subtalar arthroereisis showed the existence of contradictory views on this issue. On the one hand, most authors emphasize sufficiently high effectiveness of this operation (60-95.4% of excellent and good results) together with other obvious advantages – less traumatic and minimally invasive treatment, relatively low cost, early verticalization and timely rehabilitation, which meets modern requirements in orthopedics. Also one of the advantages of this procedure is that later it is possible to perform more complicated and traumatic operations if it is required according to the clinical situation.

On the other hand, this method can pose some complications not excluding repeated procedures and development of irreversible states in the hindfoot.

The question of combination of arthroereisis with other operations on different parts of foot and different soft tissue and bone structures depending on the patient's age and degree of foot deformity remains open. Another promising direction of research is the development of indications and contraindications to arthroereisis in children and adults with neuromuscular diseases. Further implementation of minimally invasive preventive procedures (including arthroereisis) will provide an opportunity to prevent formation of severe multiplanar feet deformities in children with infantile cerebral paralysis and other neuromuscular diseases and will also allow to limit the indications for complex non-physiological surgery.

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Case Report

The sylvian fissure arachnoid cysts: diagnosis and endoscopic treatment

Pleşco A., Bajurea A.

PMSI Mather and Child Institut , Chisinau , Republic of Moldova
Public Institution State University of Medicine and Pharmacy "Nicolae Testemiţanu", Chisinau ,Republic of Moldova

Abstract

Chistul arahnoid al fisurii silviene: diagnosticul si tratamentul endoscopic

Chistul arahnoid al fisurii silviene (SAC) este o localizare bine recunoscută pentru chisturile arahnoide intracraniane la copii. Aceste tipuri de formațiuni chistice sunt depistate destul de frecvent la examenul imagistic intracranian la copii. Au fost raportate mai multe modalități de tratament, inclusive cele endoscopice. Raportam rezultatele clinice şi radiologice ale fenestratării endoscopice ale acestor chisturi.

Cuvinte cheie: Chisturi arahnoide, fisuri silvice, tratament chirurgical, copii.

Abstract

A Sylvian fissure arachnoid cyst (SAC) is a well-recognized location for an intracranial arachnoid cyst in the pediatric population. They are a frequent finding on intracranial imaging in children. Several treatment modalities have been reported. We report clinical and radiological outcome of fenestration of these cysts by endoscopy.

Keywords: arachnoid cysts ;Sylvian fissure,surgical treatment, children.

Correspondence to: e-mail: bajureaala@mail.ru; mob. +373 69187148

Introduction

The term arachnoid cyst is a congenital cerebrospinal fluid-filled lesion that arise during development from splitting of arachnoid membrane [5]. The first case of an intracranial arachnoid cyst was described by Bright in 1831 [10].

Arachnoid cysts can be found anywhere in the brain, but in 50-60 % occur in the Sylvian fissure [4, 6]. Cysts in the middle cranial fossa are found more frequently in males than females, they occur predominantly on the left side. Most arise as developmental anomalies [4]. Galassi and associates (1982) have pro-

vided a useful classification of the arachnoid cysts of the Sylvian fissure based on computed tomographic appearances. Type I: cyst is a small, biconvex, located in anterior temporal tip. No mass effect. Communicates with subarachnoid space on water-soluble contrast CT cisternogram (WS-CTC). TypeII: involves proximal and intermediate segments of Sylvian fissure. Completely open insula gives rectangular shape. Partial communication on WS-CTC. Midline displacement, if any, is minimal. Type III: involves entire Sylvian fissure. Marked midineshift. Boni expansion of middle fossa. Minimal communication on WS-CTC [1]. Clinical symptoms depend from the location of the cyst. Sylvian cysts may cause seizures and hemi-syndromes, headache [5].

Clinical case: The 5 years old patient, domiciled in Chişinău, district Ghidighici, was hospitalized on 15th June 2015, in the neurosurgical department of the " Institute of Mother and Child". The causes of his hospitalization were headaches complain, idiopathic intracranial hypertension, emesis, nausea, focal convulsive seizures, paresthesia of the right hand and the presence of a cyst in the Sylvian fissure, as presented on the MRI. It had been performed endoscopic cystocisternostomy on 25th December 2015.

The skin incision was performed 2cm upwards from the zygomatic process and the temporal muscle was incised. It was, also, done an incision of the menix, through a hole burr, by drilling the parietal capsule of the cyst cavity. Right after, the endoscope was inserted, the cyst had been inspected. It could be viewed the cortex with the cortical blood vessels and its anatomical features, such as the cerebral carotid artery, the optic nerve and the oculomotor nerve. With a propulsive force of the endoscope towards the prepontine cistern, it was performed a fenestration between the oculomotor nerve and the carotid artery. Therefore, it was achieved a way of communication with the prepontine cistern. Through the prepontine cistern it could be visualized the basilar artery, the oculomotor nerve and the abducens nerve.After the surgical treatment the neurological signs regressed.

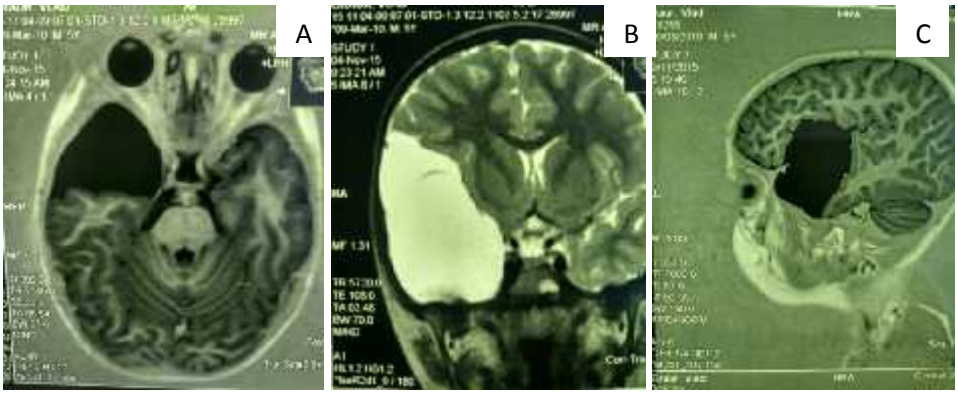

Fig. 1. Pre-operative MRI, the cyst appears with significant compression of the brain and midline displacement

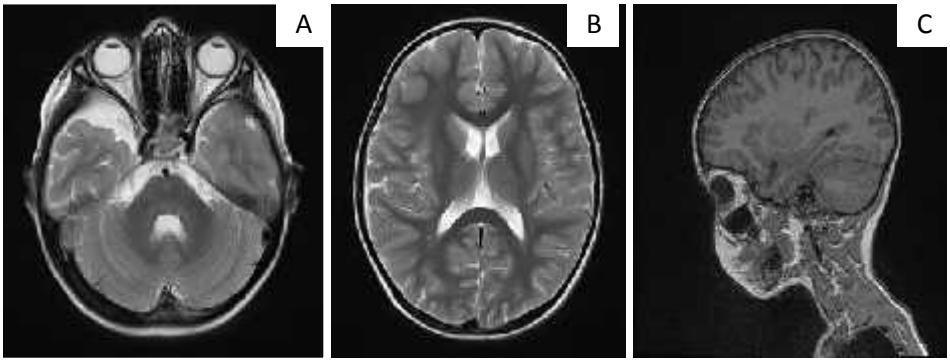

Fig. 2. Post-operative MRI, no mass effect without midline displacement

Discussions

Arachnoid cysts cause a wide spectrum of clinical symptoms. Often it is difficult to clearly establish the relation between the presence of an arachnoid cyst and problems reported by the patient, which may, although not necessarily, be due to chronically elevated intracranial pressure. In recent years, increasing attention is paid to cognitive disorders accompanying arachnoid cysts, such as memory disorders, attention disorders, or language function disorders [9]. Many authors recommend not treating arachnoid cysts that do not cause mass effect or symptoms. The surgical indication for asymptomatic arachnoid cysts remains controversial [2, 8]. In children however, asymptomatic cysts with a significant mass effect that may hinder the normal development of the adjacent brain tissue should be treated surgically [2, 3]. Symptomatic arachnoid cysts are an indication for surgery. They usually show a mass effect on MR imaging with flattening of cerebral gyri, midline shift, and/or ventricular compression. The optimal treatment for Sylvian arachnoid cysts is still under discussion. Surgical treatment options for arachnoid cysts are:

- Drainage by needle aspiration or burr hole evacuation. The advantages are simple and quick, but disadvantages are a high rate of cyst and neurologic deficit.
- Craniotomy, excising cyst wall and fenestrating it into basal cisterns.
- Endoscopic cyst fenestration through a burr hole.

The second and third treatment options have the next advantages: permits direct inspection of cyst, loculated cysts treated more effectively. The disadvantage is subsequent scarring that may block fenestration allowing re-accumulation of cyst.

- Shunting of cyst into peritoneum or into vascular system.

The advantage is low rate of recurrence and disadvantages - patient becomes “shunt dependent” and there is a risk of infection of foreign body (shunt).

Routine evaluation with CT or MRI is the procedure of choice because of its ability to demonstrate the exact location, extent, and relationship of the arachnoid cyst to adjacent brain. Further evaluation with CSF contrast or flow studies are necessary for the diagnosis of midline suprasellar and posterior fossa lesions: suprasellar cyst from dilated third ventricle, interemispheric cyst from porencephaly, posterior fossa cyst from Dandy-Walker malformation, cyst associated with isodense tumors [5, 9]. In Sylvian arachnoid cysts, the success rate is approx. 60–90% [7, 8]. Severe complications resulting in mortality and permanent morbidity are fortunately very rare (in most reports in the literature). Subdural hematomas (10%), CSF leaks (5%), and meningitis (5%) were the most frequently reported complications [7, 8].

Conclusions

The endoscopic treatment of the sylvian fissure arachnoid cysts is the most effective option and it should seriously be considered as the best choice in therapy. If the endoscopic procedure fails,craniotomy, excising cyst wall and fenestrating it into basal cisterns is the second line treatment. Shunting should be avoided when-ever possible.

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Pontocerebelos angle tumors: diagnostic and tactics of surgical treatment

Litovcenco A., Sula M., Scutaru V.

PMSI Mother and Child Institute , Chisinau , Republic of Moldova

Abstract

Tumorile unghiului pontocerebelos: diagnosticul și tactica de tratament chirurgical

Autorii prezintă un caz clinic a unui copil cu tumoră cerebrală în care sunt elucidate particularitățile evoluției clinice și unele aspecte de diagnostic imagistic și tratament, rezultatele obținute fiind confruntate cu datele literaturii. În lucrare se menționează că în cazul tumorilor cerebrale la copii, managementul este bazat exclusiv pe lucrul în echipă în care diferiți membri trebuie să fie familiarizați în amănunte cu toate aspectele patologiei date, precum și prezența unei experiențe de lucru, impunându-se necesitatea unui studiu continuu a problemei abordate. Autorii conchid că recurența tumorilor cerebrale la copii este determinată de rezecția incompletă, subtipul histologic fiind un factor important.

Cuvinte cheie: tumori cerebrale, tumoră pontocerebelară, meningioma, tratament chirurgical, copii

Abstract

The authors present a clinical case of a child with brain tumor in which are related the particularities of the clinical evolution and some aspects of imaging diagnosis and treatment, the results being confronted with literature data. The paper mentions that in the case of brain tumors in children, management is based exclusively on teamwork in which different members need to be familiar with all aspects of the pathology as well as the presence of a work experience, which implies the need for a continuous study of a problem addressed. The authors conclude that the recurrence of childhood brain tumors is due to incomplete resection, the histological subtype being an important factor.

Keywords: brain tumor, cerebello-pontine angle tumor, meningioma, surgical treatment, children

Correspondence to: e-mail: litovcenco@mail.ru; mob. +373 60009777

Introduction

Children brain tumors of the central nervous system (CNS) constitute (2,5 – 3,5 per 100 000 children annually), and therefore contribute to a major part of daily practice in pediatric oncology [3]. Brain tumors are very heterogeneous with regard to tissue, location, pattern of spread, clinical, and age of occurrence from the neonatal period to the adolescence. These tumors also vary in their surgical outcome and carry a different risk of post-operative complications.

At the moment remains the descriptive classification based on the histopathological examination the basic element for the adequate management of child brain tumors.While the localization and cell differentiation is the basis of the diagnostic system,remained the tumor classification "histogenetic. "The last edition

of the "WHO Classification of Tumors of the Nervous System" presents 127 entities, reflecting the wide variety of CNS cellular constituents. Theoretically, all these types of tumors can develop at the child. The number of histological types of brain tumors with particular importance at the child is however significantly lower (table 1) [7]. The majority of pediatric CNS tumors can be classified into five types: meduloblastoma, pilocytic astrocytoma, diffuse astrocytoma, ependymoma and craniopharyngioma.CNS tumors occurring in childhood have important characteristics that differ from those of tumors with other localizations and which deeply influence their behavior:- high degree of invasiveness, even when the histological degree of malignancy is low - heterogeneous structure with mixed tumor areas of different malignancy degree - heterogeneous structure with mixed tumor areas of different malignancy degree - disseminate frequently on the CSF way. Irrespective of the degree of malignancy - tumors can turn, moving from a low degree of malignancy to a high degree.Compared to adult tumors prevails the localization at the posterior brain fossa level: almost half of the pediatric tumors are located at this level [7].

Clinical case:

The 9 years-old patient is hospitalized in the department of neurology with pains of increased intensity in the right hemiface,sensation of benumbness in this part, repeated vomits,double dvisual field.

The detailed anamnesis reveals the appearance of the first clinical signs for about a month.It follows medical treatment for trigeminal neuralgia with transient effect.

The neurological status reveals facial nerve paresis (House-Brackmann III-IV) and abducens on the right part.Trigeminal neuralgia on the right part. Swallowing disorders. Coordination tests with bilateral dysmetria.Unstable in the Romberg test. Pronounced hypotonic in limbs. Taking into consideration the medical discipline and the ineffectiveness of the treatment it is decided to be investigated by cerebral CT (fig.1), by what was detected an expansive process in the region of the pontocerebelos angle on the right part with a mass effect on the cerebral trunk.

Table 1. Brain tumors of special importance for children: the most frequent locations, the according to WHO malignancy grade, and the approximate percentages that they represent reported to all brain tumors at children (modified after Pollack) [7]

Type of tumor	The most frequent locations	Percent of all brain tumors	WHO malignancy grade
Pilocytic astrocytoma	Cerebellum, Hypothalamus, Optic pathways	12-18% 4-8%	
Anaplastic astrocytoma, glioblastoma	Cerebral Hemispheres Brain stem	6-12% 3-9%	III-IV
Pleomorphic xanthoastrocytoma	Brain stem Superficial cerebral Hemispheres	3-6%	II
Oligodendroglioma, mixed glioma	Cerebral Hemispheres anaplastic oligodendroglioma	2-7%	II, III
Ependimoma, anaplastic ependimoma	Lateral and third ventricle Fourth ventricle	2-5% 4-8%	II, III
Choroid plexus papilloma carcinoma	Lateral and Fourth ventricle	2-4%	I, IV
Neuroblastoma	Cerebral Hemispheres		IV
Medulloblastoma	Cerebellum	20-25%	IV
Other primitive neuroectodermal tumors(PNET)	Whole neuroaxis	1-5%	IV
Atypical teratoid/rhabdoid tumor	Infra and supratentorial		IV
Germ cell tumors	Pineal region Hypothalamus	0,5-2 %	I-IV
Craniopharyngioma	(supra) selar	6-9%	I

For the confirmation of the diagnosis, for the discussion of a differential diagnosis as well as for the appreciation of the surgical treatment tactic the patient was investigated by Contrast cerebral MRI-3Tesla.

During the examination in the cerebral MRI is confirmed the presence of a tumor mass located in the posterior cerebral pore, namely in the pontocerebelos right angle with a diameter of about 5 cm,with a moderate capture of the contrast substance and a dislocation of the anatomical structures from the average line. The relatively diffuse contour, T2-hypertense regime (fig. 2). There is a secondary obstructive hydrocephalus expressed by the marked periventricular edema and the obstruction of the adjacent to the tumor licvorian pathways (fig. 3).

The detailed analysis of the clinical and paraclinical data has determinated the establishment of an expansive intracranial process with a pontocerebelous angle. Secondary obstructive hydrocephalus. According to the national and international protocols it was decided to carry out the surgical intervention that had the aim of the tumor ablation, the decompression of cerebral structures, of cranial nerves, of circulatory pathways of the cephalorachidian fluid as well as for histological analysis of the tumor that would determine the subsequent tactic of treatment.

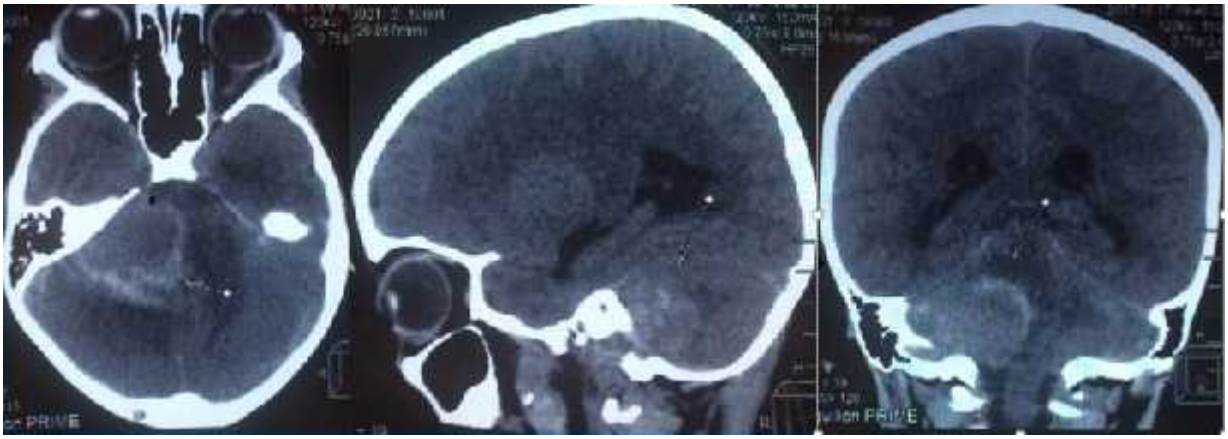

Fig. 1. Pacient L., 9 years. Preoperative CT scan. Explication in text

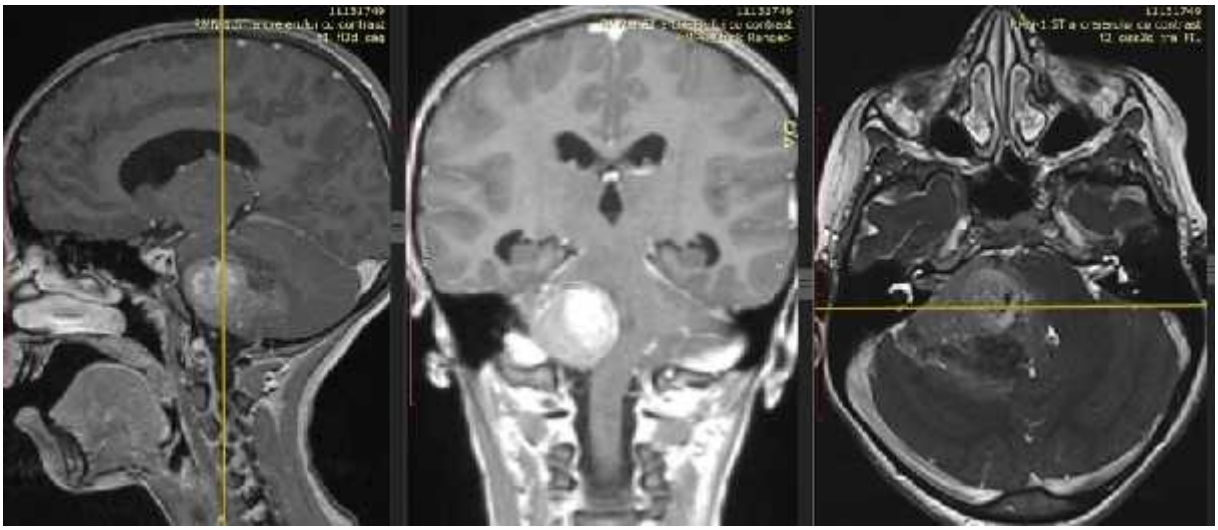

Fig. 2. Pacient L., 9 years. Preoperative MRI: UPC tumor

After a preoperative preparation, the patient was underwent surgery.Surgery with patient positioning in Park bench (patient in ¾ oblique position or almost ventral decubitus, with a roll under the shoulder and the head almost horizontal). Intraoperative the tumor grey-gray color, consecvent tough, weakly vascularized. It was performed the total tumor excision through the retro sigmoid (laterally suboccipital) approach, what was confirmed by the cerebral control CT scan on the second day after the surgery (fig. 4).

It is observed postoperatively a positive dynamic given by the regression of intracranial hypertension syndrome, of trigeminal pain, and the regression of facial nerve paresis (House-Brackmann II).

The histopathological examination denotes the presence of a meningioma meningoteliomatos WHO grade I-II.It was performed the pediatric oncologist consultation with repeated histological examination – it is confirmed the diagnosis and the subsequent treatment tactics that do not require adjunctive therapy.

Two months postoperatively the patient addresses at the emergency department with pronounced headache and repeated vomits that continue for several days. Neurological status aggravated by facial nerve paralysis (House Brackmann IV), abduction nerve paralysis on the right side, bulbar disorders, investigation by cerebral contrast MRI-relapse of expansive process with pontocerebelous angle on the right side with clivus and sellar region invasion (fig. 5).

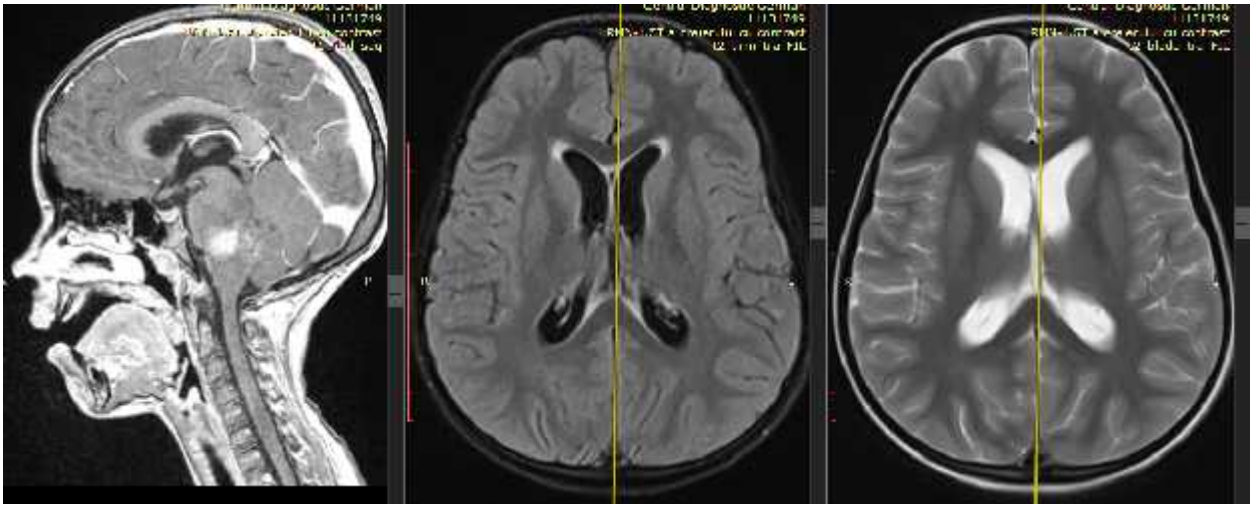

Fig. 3. Pacient L., 9 years. Preoperative MRI: periventricular edema and the obstruction of the adjacent to the tumor licvorian pathways

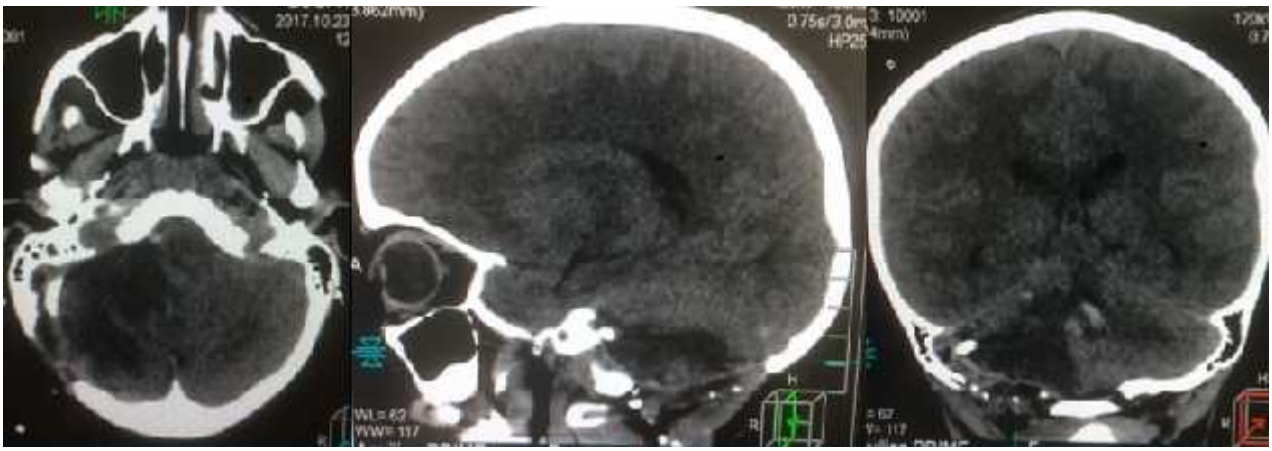

Fig. 4. Pacient L., 9 years. Postoperative CT scan: total tumor excision.

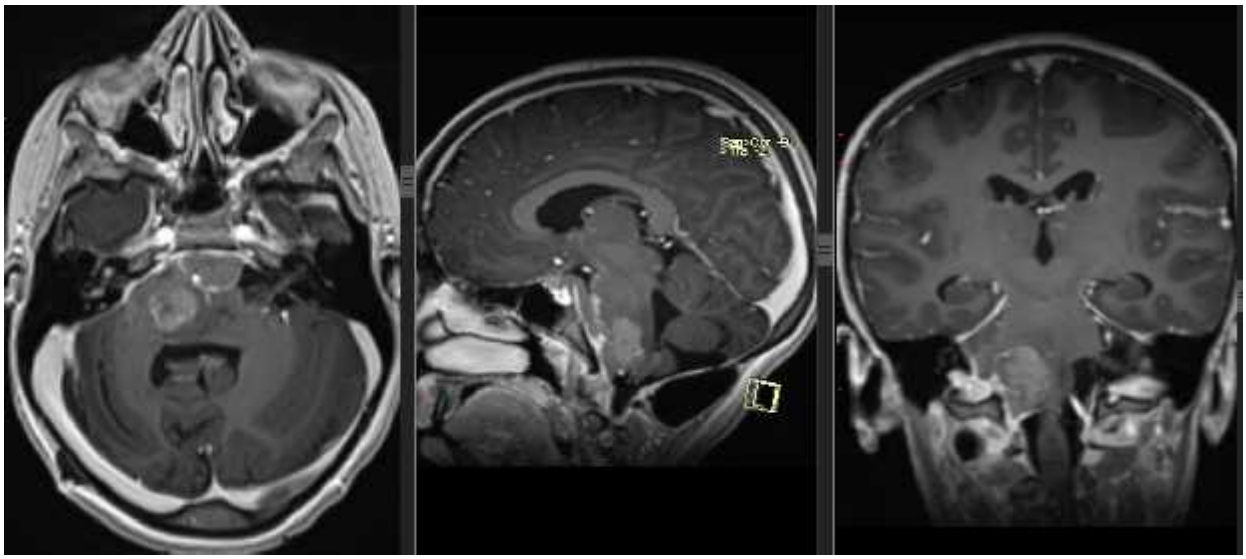

Fig. 5. Pacient L., 9 years. Postoperative MRI scan. Explication in text

Discutions

Meningiomas are uncommon neoplasms in the pediatric age group and differ in various clinical and biological aspects from meningiomas at the adult population [1].

The surgical treatment of posterior pit tumor at children, namely with pontocerebelos angle, has still great difficulty in diagnosis appreciation, performing the surgical step and the histopathological diagnosisas soon as possible.The amount of residual tumor tissue after surgery remains anyway a determining factor for the relapse rate [4, 6].Radical surgery is not possible in most cases due to tumor diffuse boundaries.Extensive tumor resection is useful for thereduction of tumor volume in limited spaces, life quality improving, remission duration increasing and is contributing substantially to the increasing of life expectancy [8]

About 90% of the meningiomas are slow-growing tumors (low-grade tumors), but even they are considered "biologically malignant" because the surgical treatment is often impossible due to localization [5]. Different series in the literature have shown a recurrence rate of approximately 13% [2].

Conclusions

Recurrence seems to be strictly related to incomplete resection and/or histologic subtype of the meningioma. Atypical, aggressive show a higher rate of recurrence and higher incidence of skull base location. Furthermore, there seem to be higher proportion of these tumors in pediatric age with WHO grades II and III.

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