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Review article

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Freiberg-Köhler disease: clinical manifestations, diagnostics, and treatment (literature review)

S.M. Gudi✉, M.D. Luchshev, V.V. Kuznetsov, L.K. Skuratova, F.V. Prokaev, I.A. Pakhomov

Novosibirsk Y.L. Tsivian Research Institute of Traumatology and Orthopaedics, Novosibirsk, Russian Federation

Corresponding author: Sergey M. Gudi, smgudinsk@gmail.com

Abstract

Introduction The paper deals with the main aspects of osteochondropathy of the heads of the II–V (small) metatarsal bones. The relevance of the problem of treating patients with Freiberg-Kohler disease can be explained by high incidence and poor results of traditional methods of its management. **Objective** The aim of this work was an attempt to generalize the available data and improve the understanding of approaches to the treatment of osteochondropathy of the heads of the II–V metatarsals. **Materials and methods** The publications obtained in various information systems (PubMed, eLibrary.ru, Google Scholar) were reviewed. **Results** Issues of history, etiology, pathogenesis, systematization and diagnosis of this disease were highlighted. The analysis of existing methods of treatment was carried out. Their advantages and disadvantages were assessed. **Conclusion** Despite more than a century of studies on Freiberg-Kohler disease, the available literature is scarce. Most works are case reports or series of cases describing small samples. This fact significantly reduces the scientific value. Thus, improving the methods of diagnosis and treatment of patients with this disease based on the foundations of evidence-based medicine is a task of current traumatology and orthopedics.

Keywords: Freiberg-Kohler disease, Kohler II disease, Freiberg disease, osteochondropathy of the small metatarsal heads, aseptic necrosis of the II–V metatarsal heads

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INTRODUCTION

Osteochondropathy of the heads of the 2nd to 5th (small) metatarsal bones is a disease, the description of which was first presented in the literature by A. Freiberg in 1914. For more than 100 years of studying this pathology, the researchers have accumulated a large amount of information regarding its risk factors, etiological and pathogenetic issues in the development of osteochondropathy of the metatarsal heads. Among all locations of osteochondropathy, aseptic necrosis of the heads of small metatarsal bones ranks fourth in prevalence. This disease affects mostly young people who are active in work and social life. However, its conservative treatment is ineffective. In most patients, degenerative changes in the head may advance and result in deforming osteoarthritis of the metatarsophalangeal joint, impairing the ability to work and the quality of life. To date, many options have been proposed for surgical treatment of osteochondropathy of the heads of the small metatarsal bones. However, in the professional trauma and orthopedic literature, most of the studies are a description of separate clinical cases or a series of cases with a small sample. It significantly reduces their scientific value. Thus, M. Alhadhoud analyzed 49 research papers on the treatment of Freiberg-Kohler disease in 2020. The author poited that only two works that are retrospective comparative studies of various methods have level of evidence III and grade C of recommendations [1]. At the same time, many authors note the need to conduct large prospective controlled comparative studies in order to state the advantages and disadvantages of every method of surgical treatment of patients with aseptic necrosis of small metatarsal heads.

The purpose of this work was to summarize the available data and deepen the understanding of approaches to the treatment of osteochondropathy of the heads of the 2nd to 5th metatarsal bones.

MATERIALS AND METHODS

This review of literature was conducted in April-May 2021 and was based on scientific studies from PubMed, eLibrary, Google Scholar databases. The search was carried out using the keywords: aseptic necrosis of metatarsal heads, osteochondropathy of metatarsal heads, Koehler II disease, Freiberg disease, metatarsal osteonecrosis, metatarsal head osteochondropathy, aseptic necrosis of metatarsal head. From the resulting studies, those articles were selected that corresponded to the subject of the study; preference was given to the works published from 2010 to 2021, inclusive. If necessary, earlier works were added to this review sample that revealed those aspects of the etiology, pathogenesis and treatment of Freiberg-Kohler disease that remain unchanged and relevant today. To clarify the main issues of the work and obtain additional

information, a targeted search for works was carried out using the links provided in the already found materials. Those works are cited on a general basis.

History and terminology

In 1914, the American orthopedic surgeon A. Freiberg (Fig. 1) reported the results of treatment of six patients with damage to the heads of the second metatarsal bones for the first time in the article "Infraction of the second metatarsal - a typical injury". Therefore, in the American literature, osteochondropathy of the heads of the metatarsal bones is called Freiberg's disease. The author associated the damage to the heads with their injury.

Fig. 1 Albert Henry Freiberg (1868–1940), American orthopaedic surgeon [2]

In the European literature, osteochondropathy of the heads of the metatarsal bones was first highlighted by the German founder of radiology A. Köhler (Fig. 2). In his book of 1915 “Grenzen des Normalen und Anfänge des Pathologischen im Röntgenbilde”, the author described 4–5 cases of damage to the heads of the metatarsal bones, linking them with a cancellous bone “infarction” resulting in aseptic necrosis. Nine years later, A. Köhler published an article in which he combined the available knowledge about this disorder. Moreover, the author pointed to an "incomplete" description of "aseptic necrosis of the heads of the metatarsal bones" in the works of A. Freiberg. In turn, A. Freiberg in 1926 recognized that acute trauma is unlikely to be the cause of aseptic necrosis and noted the fundamental nature of A. Köhler's research [3-6]. Therefore, in Europe, including Russia, osteochondropathy of the heads of the metatarsal bones became known as Koehler II disease. In our opinion, based on the chronology of publications and the invaluable contribution of both authors to the study of this disease, Freiberg-Kohler disease is the optimal eponym for osteochondropathy of the metatarsal heads.

Definition

Freiberg-Kohler disease is a degenerative necrotic disease of the osteochondropathy group that affects the heads of the metatarsal bones II–V.

Fig. 2 Albam Koheler (1874–1947), German radiologist [7]

Incidence

In the group of osteochondropathy, Freiberg-Kohler disease takes the fourth place in terms of frequency of occurrence after disorders of the hip, knee and ankle joints [8]. The incidence of this pathology in the population ranges from 0.01 to 0.22% [9]. Freiberg-Kohler disease is the only osteochondropathy that is more common in women than in men; the ratio is 5:1 [10]. The disease usually begins at the age of 11 to 17 years [11], but the consequences of aseptic necrosis of the heads of the metatarsal bones, of course, is observed at a later age. The disease is more common in the right foot than in the left. At the same time, it most often affects the head of the second metatarsal bone (68% of cases), followed by the third metatarsal bone (27%), then the fourth metatarsal (3%) while the head of the fifth is affected extremely rarely [12, 13]. Moreover, bilateral disease of both second metatarsal bones is observed in about 10% of cases, less frequently of the 2nd and 3rd on both sides, and very rarely, two adjacent heads on one foot [14, 15].

Etiology and pathogenesis

According to literature, the most popular theories of Freiberg-Kohler disease are traumatic and vascular. There are attempts to simplify the understanding of the pathogenesis of this disorder and to associate an “acute” injury with necrosis of the metatarsal head by comparing it how Columbus placed the egg vertically on the table and broke it from one end; therefore, in the English literature, the term “egg shell fracture” is found. However, the role of microtrauma to the epiphyses of the metatarsal bones is more frequently

discussed in the pathogenesis of this disease. Thus, A. Köhler suggested that the cause of aseptic necrosis is a barely noticeable repetitive mechanical impact [3, 5], which was confirmed in the works of other authors. Since this disease is more common in women, wearing fashion shoes may also be the cause, what is also reflected in the literature [16]. However, according to other researchers, there is no direct relationship with wearing of uncomfortable shoes and Freiberg-Kohler disease [17]. At the same time, it has been proven that diseases such as hallux rigidus and hallux valgus, Morton's foot, congenital or acquired shortening of the 1st metatarsal bone disrupt the normal biomechanics of the forefoot and lead to an increased load on the heads of the second, third and fourth metatarsal bones, which may be one of the reasons of the disorder [16]. The onset of Freiberg-Kohler disease in adolescence has prompted a speculation that there is a relationship between foot bone growth and the development of this disease [18]. The center of ossification of the second to fifth metatarsal bones appears in children aged 5 to 8 years, and the final fusion of the metaphyseal zone occurs at the age of 18 to 20 years. Trauma at any stage of its development can lead to a lack of blood circulation and the initiation of the disease [15]. Features of the blood supply to the heads of the metatarsal bones undoubtedly serve as an important link in the pathogenesis of the development of the disease. The heads of the small metatarsal bones have two vascular networks, one in the diaphysis and one in the epiphysis [19]. However, B. Rath found that in half of the cases, the distal branches of the arteries supplying the diaphysis of the second metatarsal bone do not participate in the blood supply to its distal epiphysis. In this situation, the head of the metatarsal bone is supplied with blood only by the vessels located at the site of attachment of the articular capsule of the metatarsophalangeal joint [20]. Thus, in traumatic edema, there occurs compression of the feeding arteries, which is probably triggers dyshemic disorders at the microvascular level in the metatarsal head [21, 22]. Vascular disorders can also result from mechanical destruction of blood vessels, blockage of the arterial or venous bed. It should be noted that the use of corticosteroids, alcohol consumption, and Gaucher disease cause impaired bone tissue perfusion due to an increase in intramedullary pressure [8, 22, 23, 24]. Regardless of the cause of the circulatory disorder, initial histological changes occur as early as the first week. In the second week, the death of hematopoietic cells, endothelial cells of capillaries and lipocytes of bone tissue occurs. Without the ability to self-repair, such avascular bone eventually collapses, becoming fragmented and sclerotic [25]. Thus, the disease begins with epiphyseal ischemia and then progresses to cancellous absorption of the head of the affected metatarsal bone, collapse of the articular cartilage, and, ultimately, mushroom-head deformity with pronounced arthritic changes in the metatarsophalangeal joint [25, 26]. The occurrence of aseptic necrosis of the head of the second metatarsal bone has been reported in twins, suggesting a possible genetic predisposition to this disease [27]. In their study, M. Myśliwiec et al. reported the association of violations of the coding sequences of the fifth chromosome with the formation of Freiberg-Kohler disease of the second metatarsal bones in two patients with Lejeune's syndrome [28]. The literature also describes a case of Freiberg-Kohler disease associated with epiphyseal dysplasia. Due to the underlying disease, the patient was diagnosed with bilateral lesions of the heads of the second metatarsal bones, femoral heads, and other bones, which the authors also associated with mutations in the genome [29].

Classification

A large number of classifications of Freiberg-Kohler disease have been proposed in the literature [4]. Most authors of the last century used the existing classifications of osteochondropathy when systematizing this disease, in particular Perthes' disease. In the pathogenesis of osteochondropathy, the following stages are distinguished: ischemia, infarction, necrosis, subchondral fracture with collapse of the articular surface, resorption and remodeling. In accordance with the well-known staging of the course of this group of diseases, professor R.R. Vreden proposed a classification of Freiberg-Kohler disease [30]. However, this classification does not consider the anatomical and morphological features of the heads of the metatarsal bones that was taken into account in the original classification by I. Smillie in 1967 (Fig. 3) [26]. The author identified 5 stages of the disease course: stage I is characterized by a "crack" of the distal epiphysis of the metatarsal bone and sclerosis of the cancellous bone; stage II is resorption of the spongy bone of the proximal fragment with the impression of the articular cartilage of the head of the metatarsal bone; stage III is progression of bone resorption of the distal epiphysis which leads to an impression fracture of the articular surface with the formation of paraarticular osteophytes; stage IV is characterized by the progression of the articular surface impression, osteophyte fractures with the formation of free intraarticular bodies, restoration of normal anatomy is impossible; stage V is osteoarthritis with flattening and deformity of the affected metatarsal head. This classification is the most cited and used as it reflects the dynamics of disease progression and allows the surgeon to determine treatment approaches. Attempts to improve the classification of Smillie in later works have not been widely adopted.

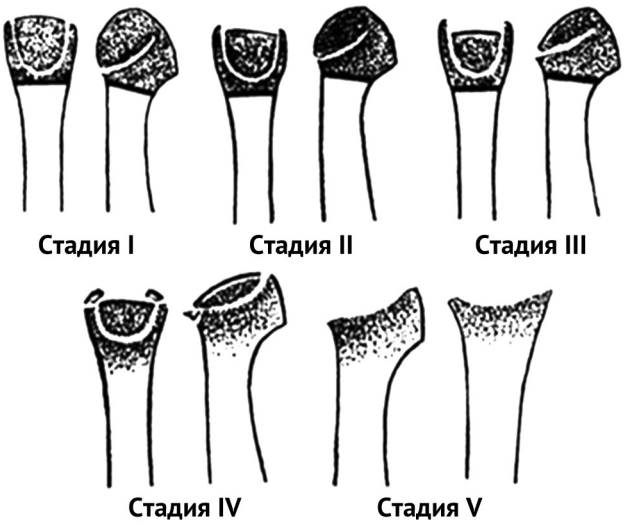

Fig. 3 Classification of Freiberg-Kohler disease according to Smillie (1967) [26]. Stage I: "crack" in the epiphysis, sclerosis of the cancellous bone. Stage II: resorption of the cancellous bone of the proximal fragment, depression of the articular cartilage. Stage III: progression of bone resorption, articular surface impression, formation of paraarticular exostoses. Stage IV: progression of the articular surface impression, fracture of marginal exostoses with the formation of free intra-articular bodies. Restoration of normal anatomy is impossible. Stage V: osteoarthritis with flattening and deformity of the metatarsal head

Thus, in 1979, G. Gauthier and R. Elbaz proposed a classification based on the changes in the morphology of the tissues of the affected metatarsal head and assessment of their regenerative potential [12]. The authors distinguished five stages of the course of aseptic necrosis. The zero stage is characterized by a "marching" fracture of the subchondral bone of the metatarsal head, which is not visualized radiographically and has a high ability to consolidate; the first stage is osteonecrosis of the head of the metatarsal bone without its deformity and the ability to recover is still preserved; the second stage is manifested by osteonecrosis of the head with deformity of its articular surface, but regeneration is still possible; the third stage is characterized by progressive damage to the cartilage of the metatarsal head, and restoration of tissue viability at this stage is no longer possible; the fourth stage is osteoarthritis of the metatarsophalangeal joint. Thus, this classification is largely based on the morphological picture and has blurred boundaries between the stages of Freiberg-Kohler disease, which limits its use in clinical practice. In 1987, F. Thompson and W. Hamilton proposed to systematize this disorder based on the choice of treatment method [29]. The authors identified four types of Freiberg-Kohler disease. Type I lesions are transient and do not manifest themselves as deformity of the articular surfaces of the metatarsal head. Its treatment, however, is limited to conservative methods. In type II, as a result of a pronounced ischemic lesion, an impression fracture occurs in the distal epiphysis of the metatarsal bone without damage to the articular cartilage, but with the development of periarticular osteophytes. Its surgical treatment is debridement of the metatarsophalangeal joint and cheilectomy. Type III Freiberg-Kohler disease is characterized by severe damage to the articular cartilage of the metatarsal head, as well as severe proliferative and degenerative changes in the metatarsophalangeal joint. The treatment option for this type is exclusively surgical, aimed at restoring the congruence of the articular surfaces. Type IV is extremely rare and is a form of epiphyseal dysplasia that affects the heads of several metatarsal bones. Its management is surgical and should correspond to the specific stage of the lesion in each metatarsal head. Due to the lack of standards of conservative and surgical treatment depending on the stage of osteochondropathy of the metatarsal heads, practical application of this classification is difficult and is not supported by an evidence base.

Summing up all of the above, from our point of view, the Smillie classification is the most convenient for practical and scientific work. At the same time, wide application of this classification and small samples of patients in the available works enable to combine data for systematic reviews.

Clinical manifestations

The initial clinical manifestation of the disease is patient's complaints of pain in the area of the affected metatarsophalangeal joint by walking, especially barefoot. The patient may report a "feeling of walking on a rock" or "bumps in the sock" [31]. As the disease progresses, the pain becomes more intense, the range of motion in the affected joint is lost. The area of the metatarsophalangeal joint becomes deformed, edema is noted, movements are accompanied by crepitus. In the later stages, the finger of the affected ray can shift to the position of the dorsal subluxation, with the formation of a hammer-shaped deformity and disorder of its support function, a positive modified Lachman stress test is noted, which indicates instability of the capsular-ligamentous apparatus. There are skin manifestations in the form of keratoses and hard calluses, both from the plantar and dorsal surfaces of the affected joint due to pathological conflict with footwear.

Diagnosis

Clinical manifestations can only suspect the disease. Features of the lesion and indications for surgical treatment are determined using instrumental methods. A standard radiography, which includes an assessment of foot X-rays in the frontal and lateral views, is uninformative in the early stages of Freiberg-Kohler disease. The earliest radiographic sign, joint space dilatation, is determined only 3–6 weeks after the onset of the first symptoms [33]. As the disease progresses, the x-ray pattern becomes clearer and more specific, with increased subchondral bone density and flattening of the metatarsal head. In the later stages of the disease, there is a decrease

in bone density, depression of the central fragment of the articular surface, free articular bodies, and sclerosis of the affected head. At the stage of the outcome of the disease, signs of osteoarthritis of the metatarsophalangeal joint are revealed against the background of mushroom-shaped deformity of the head and articular surface of the main phalanx of the toe. Thus, in the later stages, the radiographic symptoms of Freiberg-Kohler disease are pathognomonic and extremely characteristic, while in stage I, according to the Smillie classification, radiodiagnosis is difficult. At the same time, for the purpose of early diagnosis and assessment of the morphology of the lesion, additional research methods are successfully used, such as magnetic resonance imaging (MRI), multislice computed tomography (MSCT), scintigraphy and ultrasound (US). An MRI study assists to determine changes in the structure of the bone marrow in the early stages of Freiberg-Kohler disease [34]. Diffuse bone marrow edema gives low signal intensity on T1-weighted images and high signal intensity on T2, which allows detection of osteonecrosis signs in the heads before radiological manifestations.

At later stages of the disease, the osteonecrotic segment shows low signal intensity on both T1- and T2-weighted images [23, 35]. However, there is an opinion that the small size of the metatarsal head, as well as the signal enhanced due to bone marrow edema and articular effusion, may interfere with the assessment of the exact location and size of the focus of osteonecrosis on MRI. Computed tomography is more informative for detecting subchondral fractures, free intraarticular bodies, osteophytes, and disorders in the relationship of articular surfaces [36]. Thus, MSCT allows a detailed assessment of the area of osteonecrosis of the metatarsal head to determine the optimal tactics of surgical treatment [37]. The practical use of scintigraphy in the diagnosis of Freiberg-Kohler disease is currently limited. However, in separate works, the authors emphasize the unique possibilities of the method in the diagnosis of osteonecrosis at different stages. During the acute ischemic phase, the entire affected metatarsal head accumulates an isotope (technetium-labeled diphosphonate) due to increased osteoblast activity. In later stages, a delineated edge of increased activity surrounding the hypoperfused necrotic nucleus becomes apparent [38, 39]. Ultrasound diagnostics allows assessing the shape of the metatarsal head, its structure, the condition of hyaline cartilage, the presence of fluid and intraarticular bodies in the joint cavity [40]. The advantages of ultrasound are the absence of radiation exposure and the availability of the method. Moreover, ultrasound assesses the condition of the soft tissues around the joint, which helps to make a differential diagnosis of Freiberg-Kohler disease with Morton's neuroma.

Treatment

Conservative methods of treating patients with Freiberg-Kohler disease are effective only in the early stages of the disease, when the damage to bone and cartilage structures is not pronounced, and the potential for their recovery is preserved [41]. Due yo the etiopathogenetic features of this disease, the goal of conservative treatment, in addition to alleviating symptoms, is to slow down the rate of its progression. The intake of oral anti-inflammatory drugs, decrease in physical activity, unloading of the forefoot, and orthopedic appliances form the basis of conservative treatment [10, 42–44]. However, until the 1930s, this approach was the method of choice for the treatment of patients with Freiberg-Kohler disease [30]. Given the low effectiveness of conservative methods, especially in the treatment of patients in the late stages of Freiberg-Kohler disease, orthopedic traumatologists began to introduce surgical methods of treatment into clinical practice. In turn, the latter can be divided into three groups: "palliative", "joint-destroying" and "joint-preserving".

Palliative surgical methods for the treatment of patients with Freiberg-Kohler disease include debridement of the metatarsophalangeal joint, cheilectomy, bone decompression of the metatarsal head, distraction and unloading of the affected metatarsophalangeal joint in an external fixation device.

Debridement of the metatarsophalangeal joint

In 1914, A. Freiberg first proposed an original technique for debridement of the affected metatarsophalangeal joint with the removal of intra-articular bodies and para-articular exostoses that develop in the late stages of Freiberg-Kohler disease [45]. This technique has not undergone any special changes and has been currently used, but more frequently as one of the stages of surgical treatment. Nevertheless, there are still works in which the authors report satisfactory results of the application of this method alone [1]. Thus, in 2013, M. Erdil and colleagues used the debridement technique in the treatment of 14 patients in the late stages of the disease with an average follow-up of 40.2 months. The authors reported an improvement in the forefoot function and quality of life after surgery according to AOFAS and SF36 scores, respectively, which allowed them to conclude that the technique was effective [46]. The use of arthroscopic technique in the rehabilitation of the metatarsophalangeal joint enables to decrease the surgical invasiveness. In 2004, L. Carro et al. presented a case of arthroscopic debridement of the affected metatarsophalangeal joint with excision of the base of the proximal phalanx that achieved a good clinical result [47]. In turn, G. Maresca and K. Hayashi described the technique of arthroscopic debridement with osteoperforation of the affected head [14]. However, only a few clinical cases were presented in

these few works, and the authors themselves stressed the need for further study of the effectiveness of this method.

Cheilectomy

In the available literature, there are some works devoted to the evaluation of the use of cheilectomy in the surgical treatment of Freiberg-Kohler disease. thus, in 2010, M. Air and colleagues presented a clinical case of treating a patient with Smillie stage IV Freiberg-Kohler disease with the use of this method. A year after the operation, the authors observed an increase in the range of motion in the affected metatarsophalangeal joint from 30° to 90° and complete relief of pain. The patient was able to return to professional dancing [48]. However, the use of cheilectomy alone is aimed at increasing the range of motion in the joint and does not affect the pathogenetic mechanisms of the disease. At the same time, partial resection of the necrotic head can lead to dorsal subluxation of the toe and formation of its hammer-like deformity.

Osteoperforation of the head

According to current specialized in traumatology and orthopedics literature, bone decompression has been successfully used in the treatment of the pathology of the femoral head and talus to relieve intraosseous pressure associated with avascular necrosis. The intervention is aimed, among other goals, at revascularization of the necrosis area and stimulation of structural changes [49]. In 1995, A. Freiberg and R. Freiberg presented a case of successful decompression with a five-year follow-up in one patient [50]. In turn, M. Dolce published a report on the use of this technique in a 34-year-old patient, whose X-ray 12 weeks after the operation showed positive changes in the areas of sclerosis of the head, and after 14 months the patient noted a regression of all symptoms of the disease [49]. In both articles, decompression was performed by creating several holes in the head with a 1.1-mm Kirschner wire. In all the presented patients, osteochondropathy was diagnosed at an early stage without structural changes in the metatarsal head. The literature presents a number of works on the effectiveness of conservative methods of treating patients in the early stages of Freiberg-Kohler disease without bone decompression of the affected metatarsal head [44, 51, 52], which emphasizes the need for further study of the feasibility of using this method.

Transosseous compression-distraction method (TCDM) in the management of Freiberg-Kohler disease

According to the literature, in osteochondropathy, in particular for Perthes' disease management, a good effect is observed with various devices to unload the affected joint. However, in Freiberg-Kohler disease, this method, like other methods of hardware decompression, is almost never used. In 2005, A.V. Ralnikov reported on the successful treatment of 70 patients with Freiberg-Kohler disease using the TCDM method. The meaning of the operation proposed by the author in his dissertation work is to pass the wires through the distal metaphysis of the affected metatarsal bone and the main phalanx of the corresponding toe, followed by unloading in external fixation devices of various designs. According to the author, in the observation period of 9 to 12 months, all patients had good results: complete or almost complete cessation of pain [53]. The undoubted advantage of the proposed method, according to the author, is the possibility of its use in children with open growth zones. However, the presence of an external fixation device in the area of the forefoot with pins protruding onto the skin is accompanied by high risks of infectious complications, difficulties in observing the orthopedic regimen, and the need for constant monitoring of the patient in a medical facility. In 2011, Yu. G. Shekunova et al. presented retro- and prospective studies of patients treated for Freiberg-Kohler disease with distraction of the affected metatarsophalangeal joint. The aim of the researchers' work was to search for new minimally invasive techniques that improve neoangiogenesis and regeneration of the affected metatarsal head. The main group consisted of 34 patients treated with TCDM in combination with osteoperforation of the metatarsal head with high-intensity laser radiation; the comparison group consisted of 70 patients in whom the therapeutic effect was limited to TCDM. Based on ultrasonic osteometry, the researchers concluded that the use of laser osteoperforation can reduce the time needed to restore blood flow in the affected metatarsal head. According to the authors, the complex application of their method improves the results and reduces the treatment time for patients with Freiberg-Kohler disease [40]. However, we did not find confirmation of this conclusion by other researchers in the available literature.

The "joint-destroying" methods of surgical treatment of patients with Freiberg-Keller disease include arthroplastic resection of the affected metatarsal head, interpositional arthroplasty of the metatarsophalangeal joint. These methods are used in patients in the late stages of Freiberg-Kohler disease (Smillie stage IV-V), when restoration of the congruence of the articular surfaces is not possible.

Arthroplastic resection of the damaged head of the metatarsal bone

Resection of the affected metatarsal head is one of the most "radical" approaches in the treatment of Freiberg-Kohler disease. In the literature, there is a description of the operation technique according to the A. Denis method, which consists in resection of the head of the 2nd metatarsal bone followed by resection of the heads of the 3rd–4th metatarsal bones for alignment of the distal foot. Also known is the operation of G.V. Khondrikov, the method of osteoplastic surgery for Freiberg-Kohler disease which consists in resection of the affected

head of the metatarsal bone and transplantation of adipose tissue into the area of the formed defect [54]. J. Hoskinson published a report in 1974 in which he stated that 2 out of 4 patients who underwent resection of the affected metatarsal head were not satisfied with the treatment result [52]. N. El Mahboub in 2019 analyzed the results of treatment of 20 patients suffering from Freiberg-Kohler disease using the metatarsal head resection technique. The author reported that all patients had relief of pain and maintained the function of the forefoot [55]. However, the fact remains undoubted that the resection of the head is a "crippling" operation, while modern concepts in traumatology and orthopedics are organ-preserving techniques. A significant shortening of the metatarsal bone changes the biomechanics of the forefoot, what inevitably leads to poor long-term results. Moreover, this technique makes the use of subsequent reconstructive interventions extremely difficult [13].

Interpositional arhtroplasty

One of the methods for filling the defects of the affected head after its partial or complete resection is interpositional arthroplasty. Various donor sites have been suggested in the literature, such as extensor digitorum longus tendon, extensor digitorum brevis tendon, local pedicle tissue graft, palmar longus tendon, peroneus brevis tendon, etc. [1, 5, 6, 10, 13]. In a study by Y. Ozkan in 2008, the results of treatment of 10 patients with the method of interpositional arthroplasty with the extensor digitorum brevis tendon were presented. In the final assessment of the results, the author observed 4 excellent results, 5 good and one fair; the average AOFAS score increased from 58.3 (44–77) before surgery to 80.4 (67–100) after it [56]. In 2007, T. Lui presented the results of treatment of two patients (women, 45 and 60 years old) who used the technique of arthroscopic interpositional arthroplasty with the extensor digitorum brevis tendon. The author reported good results of treatment in both patients [57]. In turn, C. Liao and colleagues in 2015 presented a clinical example of the treatment of a 27-year-old man by the method of intrapositional arthroplasty with a Palmaris longus tendon graft. Three months after the operation, the level of pain on the VAS scale decreased from 6–7 to 0–1 points [58]. In 2016, domestic authors proposed an original method for the surgical treatment of Freiberg-Kohler disease by wrapping the affected head of the metatarsal bone with a graft taken from the fascia lata of the thigh. The authors of the patent presented a clinical example of a 14-year-old patient who had a good treatment result 3 months after the intervention. However, no long-term follow-up results were reported [59].

However, it should be noted that a frequent complication that a surgeon may encounter is the occurrence of a symptom of a "painful donor site" in the area of graft harvesting. In this regard, in 2018, W. Abdul et al. proposed a technique for interpositional arthroplasty with a periosteal graft of the proximal phalanx of the toer and the distal portion of the capsule of the affected metatarsophalangeal joint on the "fat pedicle". The results of treatment of 23 patients with Freiberg-Kohler disease by the proposed method were presented. The authors noted a significant reduction in pain according to the VAS scale and an improvement in the function of the forefoot according to the AOFAS questionnaire [60]. However, E. Stautberg and colleagues in 2020 expressed the opinion that the soft tissues surrounding the affected metatarsophalangeal joint are also subject to degenerative changes and can become an independent source of inflammation when used as an interposition material. In order to exclude this negative effect on long-term results, the author suggested using a frozen semitendinosus tendon allograft. According to the researcher, the advantage of using this technique is the stability of the frozen graft tissue and the absence of pain in the donor site. Retrospectively, 15 patients were assessed; the average pain score on the VAS scale decreased from 7 to 1 point. However, three patients required reoperation. One of them had migration of graft fixators which resulted in its instability and removal of the plastic material. The second patient underwent additional debridement of the joint due to the presence of residual para-articular exostoses. The third patient had progression of valgus deformity of the first toe, which required surgical correction [61]. It should be noted that the question of the body's immune response to the allograft, as well as the nature of its interaction with local tissues, remains open. At the same time, a common disadvantage of interpositional arthroplasty, regardless of the type of the graft, is the formation of fibrous ankylosis with impaired biomechanics and stability in the metatarsophalangeal joint.

The "joint-sparing" methods of surgical treatment of patients with Freiberg-Keller disease include osteotomy, osteochondroplasty of the head (capituloplasty) of the metatarsal bone, endoprosthetic replacement of the metatarsophalangeal joint. Modern orthopedic traumatologists emphasize the need to restore the congruence of the articular surfaces and the lost anatomy of the metatarsophalangeal joint for successful treatment of this group of patients.

Osteotomies of the metatarsal bone

Currently, a huge number of osteotomy variants of the distal metatarsal bone affected by Freiberg-Kohler disease are known. Two main types can be distinguished, shortening or "decompressive" and remodeling osteotomies. The purpose of shortening osteotomies is to decompress the joint and unload the pathologically altered metatarsal head [5]. In 1932, Professor R.R. Vreden proposed an original osteotomy of the metaphysis of the metatarsal bone. According to the author, this osteotomy creates optimal conditions for the regeneration of the necrobiotic area of the bone tissue, but it has not been confirmed in the literature.

The author reported that 18 out of 20 patients in whom the proposed technique was applied, fully returned to their professional activities, considering themselves to be completely healthy [30]. In 1991, T. Smith et al. published the results of treatment of 28 patients with Freiberg-Kohler disease. Fifteen of them underwent plate-fixating metatarsal shortening osteotomy. Only one patient was dissatisfied with the result due to a pronounced limitation of movement in the metatarsophalangeal joint [51]. However, it should be noted that metatarsal shortening after osteotomy violates the Lelièvre's parabola, which undoubtedly affects the biomechanics of the forefoot. Thus, shortening by more than 2 mm leads to an increase in the load on the adjacent metatarsal bones, the development of "transfer" metatarsalgia and the formation of the so-called "floating toe" (impaired adaptation of the toe to the surface) [62]. Moreover, a shortened toe may result in an unsatisfactory cosmetic outcome [10].

The remodeling osteotomy of the metatarsal head in Freiberg-Kohler disease is based on the rotation of its preserved plantar articular surface to the articular surface of the proximal phalanx. It is customary to distinguish between intra-articular and extra-articular osteotomies. The main difference between extra-articular osteotomies is that the distal osteotomy passes proximal to the area of necrosis of the head. Thus, the area of necrosis is not excised, but turned onto the dorsal surface of the metatarsal bone. In 1989, P. Kinnard and R. Lirette demonstrated the results of surgical treatment of 10 patients using the method of extra-articular osteotomy according to the "closed wedge" type [63]. The authors initially suggested fixing the osteotomy zone with a wire cerclage, but, faced with a complication such as toe extensor tendonitis. So, they began to use transosseous sutures with absorbable sutures. All patients were satisfied with the result of treatment; pain disappeared. At the same time, radiologically, the average shortening of the metatarsal bone was 2.3 mm. In 2013, H. Lee et al. presented the results of treatment of 13 patients after extra-articular osteotomy of the metatarsal bone of the "closed wedge" type with fixation with Kirschner wires. The authors observed positive results of treatment in all patients. Clinical and radiographic examinations were performed at 2 weeks, and months 1, 2, 4, 6 and 12 after surgery. At the final follow-ups, the average AOFAS score was 92.2 (92-100) points, the range of motion in the affected joint increased by an average of 6o (-10-25), and the pain syndrome on the VAS scale averaged 1 (0-4) after treatment, while the value before treatment was 7.5 (5–10) [64]. Similar results were obtained by K. Ikoma et al. in 2014 using extra-articular osteotomy of the "closed wedge" type in the treatment of 13 patients in the late stages of Freiberg-Kohler disease. The authors performed fixation of the osteotomy with transosseous polymeric material sutures. The authors reported a significant decrease in pain and an increase in the range of motion in patients [65]. The disadvantage of the extra-articular osteotomy technique is the preservation of the necrotic focus and its turning into the osteotomy zone, which, undoubtedly, can affect consolidation and increase the risk of non-union.

In 1979, G. Gauthier and R. Elbaz first proposed an original intra-articular osteotomy of the "closed wedge" type. In their work, the authors presented a report on 88 cases of affected heads of the metatarsal bones in 83 patients. Fifty-three osteotomies of the "closed wedge" type were performed through the area of subchondral necrosis. All 83 patients who underwent surgery were followed up from 3 to 82 months, with an average of 22 months. Out of 53 patients treated according to the proposed original technique, only one patient developed pain syndrome [12]. In 2015, M. Helix-Giordanino and co-authors published the results of treatment of 28 patients with osteotomy of 30 affected metatarsal heads using the G. Gauthier technique with fixation with one or two staples. After a mean follow-up of 6.5 years, 17 patients rated their results as good, 11 as fair, and 2 as poor [66]. Similar results of this osteotomy were obtained by B. Pereira and colleagues in 20 patients with Freiberg-Kohler disease. The authors used a wire loop to fix the bone fragments. The mean follow-up period was 23.4 years (range, 15 to 32 years). Thereby, 16 patients reported "excellent" results and 4 "good" results, with an increase in AOFAS scores up to 96.8 (91–100) [42].

An undoubted disadvantage of intra-articular osteotomies is the limitation of their use in patients in the late stages of Freiberg-Kohler disease with mushroom-like deformity of the head and destruction of the plantar articular surface. Equally important are the difficulties of fixing the distal fragment of the osteotomized head due to its small size and high risks of its fragmentation during fixation. The question of the possibility of restoring the blood supply to the distal fragment of the osteotomized head also remains open due to damage to the vessels supplying it during osteotomy. Edmondson et al. pointed to one more drawback of the classic osteotomy of the "closed wedge" type, the impossibility of complete resection of the affected area of the bone. Given these shortcomings, the authors proposed a modified Weil osteotomy in 2011, which is a combination of two techniques developed by G. Gauthier and L. Weil. By conducting a more horizontally oriented distal filing, the apex of the wedge is located more proximal than with a Gauthier osteotomy, which allows resection of a large area of necrosis and facilitates fixation with screws. Evaluating the results of the proposed osteotomy in 17 patients with Freiberg-Kohler disease, the authors noted an improvement in the function of the forefoot according to the AOFAS from 51.6 (5–87) points before surgery to 87.6 (37–100) after it. Thereby, 12 (71%)

patients achieved an "excellent" result, 1 (6%) "good", 2 (11.7%) fair and 2 (11.7%) were poor [67]. J. Kim's 2012 study presents the results of treating 19 patients with Weil's modified osteotomy. After a mean follow-up of 72.6 months (41–121), all but one of the patients were rated as excellent or good. The patient with a fair result had some limitation of movements in the metatarsophalangeal joint, but at the same time, pain according to VAS decreased from 8 points before surgery to 4 after the intervention [68]. Also, H. Lee in 2016 published a case series of 20 patients with Freiberg-Kohler disease who underwent the modified Weil osteotomy technique. Twenty-four months after surgical treatment, all patients had relief of pain. The average VAS score decreased from 7.2 to 2.1, the function of the forefoot according to the AOFAS increased from 52.4 to 78.2 points [69]. However, it should be noted that in the study of the vascular anatomy of the heads of the metatarsal bones on cadaveric material, B. Rath (2008) found that the classic Weil osteotomy causes damage to small arteries on the medial and lateral side of the head. The author also reported that there is a risk of damage to the plantar metatarsal arteries during filing [20]. Thus, the blood supply to the distal fragment of the osteotomized head affected by aseptic necrosis is compromised, and the possibility of its restoration is extremely doubtful.

Methods of osseocartilaginous defect management

In 2002, K. Hayashi et al. presented the technique of transplantation of an osteochondral autograft taken from the unloaded part of the femoral condyle for treatment of one patient suffering from Freiberg-Kohler disease. The patient, a 13-year-old girl, had a bilateral lesion of the heads of the second metatarsal bones. Stage IV (according to Smillie) of the lesion was established on the right, stage III on the left. The right foot was operated on using the technique proposed by the authors, while osteoperforation with arthroscopic control was performed on the left foot. One year after treatment, the patient noted relief of pain on both sides [70]. In a retrospective study conducted in 2015, W. Miyamoto and colleagues evaluated the mid-term clinical outcomes of bone-cartilaginous autograft transplantation from the unloaded part of the femoral condyle after more than five years in 13 patients. The hallmark of the study is a significant improvement in AOFAS forefoot function from 66.9 to 93.0 after surgery. MRI results showed consolidation of the transplanted autograft [71]. In 2019, D. Georgiannos and colleagues evaluated the results of treating 13 patients with Freiberg-Kohler disease using osteochondroplasty. A graft was harvested from the unloaded zone of the ipsilateral talus using arthroscopic technique in 9 patients. The authors reported that all patients were satisfied with the treatment outcome and returned to professional sports activities [72]. However, surgeons often face a complication of this method, a “painful donor site” syndrome. In a retrospective study, S. Kim et al. compared two surgical techniques: osteochondroplasty using an autograft from the non-load-bearing zone of the femoral condyle and dorsal osteotomy with a “closed wedge”. The first group consisted of 12 patients, the second one included 15. The authors reported that the AOFAS scores (71.4 before surgery versus 95.7 at the end of the observation) and VAS (7.5-1.3) in the group after osteochondroplasty were better than in the group after osteotomy (70.5–87.9 and 6.9–1.9, respectively). In addition, in the first group, one complication was observed (persistent pain in the area of the graft harvesting), while in the second group, 4 patients had complications (medial deviation of the toe, claw-like deformity, contracture of plantar flexion, recurrence of pain 73]. Taking into account the shortcomings of the method of harvesting an autograft from the femoral condyles, our clinic developed and introduced into clinical practice a method for harvesting an osteochondral autograft from the pre-Achilles zone of the calcaneus [74]. In the dissertation work Kuznetsov V.V. in the experiment confirmed the presence of articular cartilage in the proposed autograft and carried out a comparative assessment of the original method of graft sampling with the Hangoody method in the treatment of patients with aseptic necrosis of the talus block. The author reports good long-term results, but in the group in which the graft was taken from the femoral condyle, the "painful donor site" syndrome was more often observed [75]. The clinic has developed an original method for osteochondroplasty of the affected metatarsal head in patients with Freiberg-Kohler disease, and received a patent of the Russian Federation [76].

In the professional literature, the issue of endoprostheses in the surgical treatment of the affected metatarsophalangeal joint has been repeatedly considered, in particular in the later stages of the disease, when restoration of the articular surfaces is impossible. Historically, silicone implants have been used for the surgical correction of small toe pathology, with mixed short-term results and unsatisfactory long-term results. Osteolysis, loosening, tissue intoxication with silicone breakdown products, and synovitis usually lead to the need to remove the implant what results in extensive defects in the bones and soft tissues of the forefoot. Other materials such as titanium and ceramics have been used as arthroplasty material in limited case studies with conflicting results [13]. In a study by B. Brandao and colleagues, the effectiveness of a silicone implant was evaluated. Despite the improvement in the quality of life in the postoperative period, according to the change in the scores on the FAAM and MOXFQ scales, four out of six patients required a second operation which was Weil osteotomy [77]. M. Glazebrook et al. reported using the same implant in 5 patients. The authors reported good treatment outcomes. The

follow-up period was 15–38 months [78]. The use of ceramic implants was highlighted by D. Townshend and M. Greiss in 2007 in their study. The authors performed arthroplasty of the second metatarsophalangeal joint in six patients with Freiberg-Kohler disease. Among them, two patients had already been operated on previously (one had a wedge-shaped osteotomy, the second had a resection of the affected head). The study also included 3 patients suffering from metatarsalgia of another etiology. The authors rated the results of the treatment as follows: six excellent outcomes, two good and one bad. The poor outcome was associated with an intraoperative fracture of the proximal phalanx of the toe; thus, it wasa impossible to install the endoprosthesis, and the authors performed arthroplasty of the affected metatarsophalangeal joint. The median follow-up was 23 months (6–46) [79]. In 2021, N. Saragas and colleagues presented their own endoprosthesis of the "small" metatarsophalangeal joint. The design of the endoprosthesis includes three components: a phalangeal component, a metatarsal component and a polyethylene liner, "meniscus". The authors conducted mechanical-strength studies and trial implantations on cadaveric material, during which it was found that the design of the prosthesis allows for up to five million flexion-extension movements under load, and the tension of the soft tissues of the joint area ensures sufficient stability of the endoprosthesis. According to the researchers, Freiberg-Kohler disease is one of the indications for the use of this prosthesis. Obviously, arthroplasty is supposed to be used in the terminal stages of Freiberg-Kohler disease or for revision interventions. The authors did not introduce the implant into clinical practice, but it was approved for trials by the ethics committee [80].

DISCUSSION

The relevance of treating patients with Freiberg-Kohler disease is explained by its high incidence and poor treatment results obtained with traditional methods. The problem is exacerbated by the age and gender characteristics of this disease. Osteochondropathy of the heads of the metatarsal bones affects mostly females (ratio of 5:1) in their adolescence [10, 11]. However, patients start seeking help at advanced stages of aseptic necrosis with a clinical manifestations of deforming osteoarthritis at a later age. Chronic pain in the area of the forefoot, limited range of motion in the metatarsophalangeal joint, biomechanical disorders of walking with the formation of hyperkeratosis reduces labor, sports and even daily activities and negatively affects the quality of life of patients. These facts determine the medical and social significance of this problem. Thus, the goal of treating patients with Freiberg-Kohler disease is to eliminate pain, restore the range of motion in the affected metatarsophalangeal joint, reduce the periods of disability and the rehabilitation period, and achieve a satisfactory cosmetic effect.

We consider it necessary to note that the foot is a single complex of support. The disorders in the relationship of its parts entail a redistribution of the load on the overlying structures. It must be considered by planning a surgical intervention. In our opinion, from a practical point of view, the existing methods of treating patients with Freiberg-Kohler disease can be divided into three groups: "palliative", "joint-destroying" and "joint-preserving". Palliative methods, despite the lack of impact on the pathogenetic mechanisms of this pathology, are widespread among practicing orthopedic traumatologists. Thus, according to a number of authors, the use of bone decompression methods helps to relieve bone marrow edema and reduce ischemia of the metatarsal head [49, 50]. However, according to the literature, this method is effective only at the initial stage of Freiberg-Kohler disease, when the articular surface of the affected head is not yet deformed. There are works in the literature that testify to the high efficiency of conservative methods of treatment in early diagnosis of osteochondropathy [44, 51, 52]. However, it is necessary to note the difficulties in diagnosing the initial stages of Freiberg-Kohler disease on standard radiographs. MRI is the diagnostic method of choice in early stages of Freiberg-Kohler disease [23, 34, 35].

Cheilectomy and debridement of the metatarsophalangeal joint, proposed by A. Freiberg at the beginning of the last century, remains the most common treatment for patients with osteochodropathy of the metatarsal heads [45]. These interventions are symptomatic and allow a slight increase in the range of motion in the affected joint, but do not slow down the progression of the disease. The TCDM method has not been widely used due to the anatomical and physiological features of the forefoot, the difficulties of external fixator placement and the discomfort of patients in the postoperative period, although the results of single studies with a large sample are encouraging [40, 53].

"Joint-destroying" operations, according to some authors, are a radical method of surgical treatment of patients with Freiberg-Kohler disease. However, partial or complete resection of the affected metatarsal head leads to disruption of spatial relationships in the forefoot with redistribution of the load in almost all patients, which is manifested by "transfer" metatarsalgia. A huge problem of these interventions is the formation of an osteochondral defect in the resected head of the metatarsal bone, in which the method of choice for most researchers is interpositional arthroplasty with autografts or allografts [56–61]. Each of these methods has its advantages and disadvantages, however, regardless of the chosen implant, this operation often leads to the formation of fibrous ankylosis of the

metatarsophalangeal joint with the possible formation of a secondary deformity of the toe, which causes a problem for the patient when wearing shoes and disrupts the biomechanics of walking.

High demands of patients on the results of surgical treatment are reflected in the current trends in traumatology and orthopedics. They are based on the restoration of the lost anatomy and the preservation of the functions of the affected segment with minimal trauma and short rehabilitation periods. In this regard, practicing orthopedic traumatologists prefer "joint-sparing" operations in the surgical treatment of patients suffering from osteochondropathy of the heads of the small metatarsal bones. Taking into account the fact that most patients seek specialized care in the late stages of Freiberg-Kohler disease with severe metatarsophalangeal joint deformity, by which it is impossible to restore its normal anatomy, the defining aspect in the approach to treatment is the maximum possible preservation of joint function. Remodeling osteotomies of the metatarsal heads, according to the literature, are the most common group of surgical methods for the treatment of late stages of this pathology [1, 4–6, 10, 13]. However, in the advanced stages of Freiberg-Kohler disease with the formation of rough mushroom-like deformities and a large area of aseptic necrosis, the use of these osteotomies is technically difficult. Thus, the surgeons, in order to minimize the disruption of relationships in the forefoot, are forced to save part of the anesthetized tissue. Partial or complete preservation of the necrotic focus in the region of the metatarsal head increases the risk of nonunion and progression of the disease, regardless of the method of fixation. It should also be noted that by performing osteotomy, there is a risk of damage to the vessels supplying the head of the metatarsal bone [20].

This can exacerbate a compromised blood supply and lead to impaired consolidation of the osteotomy area. A characteristic complication of shortening osteotomies is a disorder in the spatial relationships in the forefoot with a violation of the metatarsal arch, the consequences of which were mentioned above. Undoubtedly, arthroplasty is an effective, and in some cases, the only method of surgical treatment of the pathology of various joints. However, at the present stage, the use of small metatarsophalangeal joint endoprostheses in forefoot surgery is limited and is associated with a high rate of complications [5, 6, 10]. In this regard, endoprosthesis application in the affected metatarsophalangeal joint in Freiberg-Kohler disease is not the operation of choice, and in the available literature there are single research papers with conflicting results [77–80]. The undoubted advantage of arthroplasty is the possibility of its implementation during revision interventions, even in conditions of a total defect of the metatarsal head. In our opinion, the optimal method of surgical treatment of patients with advanced stages of Freiberg-Kohler disease is osteochondroplasty. With the use of this treatment method, the sites of entry into the metatarsal bone of the feeding vessels of the head remain intact, and this fact has a positive effect on the consolidation of the graft. In addition, due to the processing of the autograft, it is possible to restore the lost anatomy of the head of the metatarsal bone while maintaining the congruence of the articular surfaces. It allows minimizing the violation of biomechanics in the forefoot. The question of choosing a donor site for autograft harvesting remains open [70–73, 76]. The classical method of graft harvesting from the unloaded zone of the condyle of the contralateral femur is associated with the frequent occurrence of the "painful donor site" syndrome. The method proposed in our clinic for harvesting an osteochondral autograft from the pre-Achilles region of the calcaneus has shown good clinical results in the treatment of patients with aseptic necrosis of the talus trochlea [74–76]. However, a clinical study is needed to prove the benefits of this method in the treatment of patients with Freiberg-Kohler disease.

CONCLUSIONS

Most of the works available in the literature on the surgical treatment of Freiberg-Kohler disease describe single cases or series of cases surgically treated by various methods. The analysis of the literature reveals not long follow-ups without long-term results of treatment and late complications. Thus, current treatment of patients with Freiberg-Kohler disease is a problem of high relevance. It is evident that there is a need to continue improving the methods of diagnosis and management of patients with this disease by offering new low-traumatic, pathogenetically substantiated methods based on the foundations of evidence-based medicine.

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Information about the authors:

1. Sergey M. Gudi – Candidate of Medical Sciences, smgudinsk@gmail.com;
2. Matvey D. Luchshev – M.D.;
3. Vasilii V. Kuznetsov – Candidate of Medical Sciences;
4. Liliya K. Skuratova – M.D.;
5. Fyodor V. Prokaev – M.D.;
6. Igor A. Pakhomov – Doctor of Medical Sciences.