



Follicular Occlusion Syndrome — a Possible Option of Follicular-Retension Origin of Pilonidal Sinus

Daria D. Shlyk, Maria N. Pikuza, Yuri E. Kitsenko*, Anna S. Pirogova,
Nina B. Paramonova, Ramin T. Rzaev, Natalia P. Teplyuk, Petr V. Tsarkov

I.M. Sechenov First Moscow State Medical University (Sechenov University)

Aim: to analyze and evaluate the clinical and morphological manifestations of pilonidal sinus disease (PSD) as a part of follicular occlusion syndrome (FOS).

Materials and methods. In the Clinic of Coloproctology and Minimally Invasive Surgery, 80 patients with PSD underwent surgeries from November 2018 to December 2019: 62 (77.5 %) patients — with primary PSD, 18 (22.5 %) — with recurrence of the disease.

Results. There were 80 patients, 6 patients (9.7 %) with primary and one (5.6 %) patient with recurrent cyst had concomitant manifestations of follicular occlusion syndrome. Thus, the frequency of combination of PSD with other variants of FOS course amounted to 8.8 %. Hidradenitis suppurativa of axillary and inguinal areas was found in 5 out of 7 patients. Acne conglobata, as one of the components of FOS, was noted in three patients. Dissecting cellulitis of the scalp was diagnosed in one patient. Follicular occlusion triad was observed in two patients. Follicular occlusion tetrad was not noted in any observation. All patients were treated with excision of the pilonidal sinus disease with local tissue-plasty of the defect. At present, no recurrences have been noted in any of the cases, and the mean follow-up time was 14 ± 5.6 months (6–27 months). PSD as a manifestation of follicular occlusion syndrome is characterized by a more cranial and more superficial location of the cavity in the sacrococcygeal region. According to the data of histologic examination of patients with FOS, the morphologic picture is identical with patients with isolated PSD. All patients with confirmed FOS have received pathogenetic local and conservative therapy. After the therapy remission of combined diseases is noted.

Conclusion. Deroofing of the lining of the cavity, often used in dermatologic practice, along with complex treatment within the framework of multidisciplinary (together with a dermatologist) management of patients with FOS, looks promising.

Keywords: follicular occlusion syndrome, epithelial coccygeal tract, colorectal surgery, dermatovenerology

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Синдром фолликулярной окклюзии — возможный вариант фолликулярно-ретенционного происхождения эпителиального копчикового хода

Д.Д. Шлык, М.Н. Пикуза, Ю.Е. Киценко*, А.С. Пирогова, Н.Б. Парамонова, Р.Т. Рзаев, Н.П. Теплюк, П.В. Царьков
ФГАОУ ВО «Первый Московский государственный медицинский университет им. И.М. Сеченова» Министерства
здравоохранения Российской Федерации (Сеченовский университет)

Цель исследования: анализ и оценка клинико-морфологических проявлений эпителиального копчикового хода (ЭКХ) как одного из проявлений синдрома фолликулярной окклюзии (СФО).

Материалы и методы. В Клинике колопроктологии и малоинвазивной хирургии в период наблюдения с ноября 2018 по декабрь 2019 г. были прооперированы 80 пациентов с диагнозом ЭКХ, из них 62 (77,5 %) обратились в клинику с первичным ЭКХ и 18 (22,5 %) — с рецидивом заболевания.

Результаты. Из 80 наблюдений у 6 (9,7 %) пациентов с первичным и у одного (5,6 %) пациента с рецидивным ЭКХ определялись сопутствующие проявления синдрома фолликулярной окклюзии. Таким образом, частота сочетания ЭКХ с другими вариантами течения СФО составила 8,8 %. У 5 из 7 пациентов встречался гнойный гидраденит подмышечных и паховых областей. Конглобатные акне как один из компонентов СФО отмечены у трех пациентов. Рассекающий целлюлит волосистой части головы диагностирован у одного пациента.

Триада фолликулярной окклюзии наблюдалась у двух пациентов. Тетрада фолликулярной окклюзии не была отмечена ни в одном наблюдении. Всем пациентам проведено лечение в объеме иссечения эпителиального копчикового хода с пластикой дефекта местными тканями. В настоящее время ни в одном наблюдении рецидивов не отмечено, среднее время наблюдения составляет $14,0 \pm 5,6$ мес. (6–27 мес.). ЭКХ как проявление СФО характеризуется более краниальным и более поверхностным расположением полости в клетчатке крестцово-копчиковой области. По данным гистологического исследования у пациентов с подтвержденным СФО морфологическая картина идентична с пациентами, у которых ЭКХ наблюдался изолированно. Всем пациентам с подтвержденным СФО назначена патогенетическая местная и консервативная терапия. На фоне проведенной терапии отмечается ремиссия сочетанных заболеваний.

Заключение. Наряду с комплексным лечением в рамках междисциплинарного (совместно с дерматологом) ведения пациентов с диагнозом СФО перспективным направлением выглядит применение более щадящих по объему вмешательств, направленных на разрушение эпителиальной выстилки полости и часто используемых в дерматологической практике.

Ключевые слова: синдром фолликулярной окклюзии, эпителиальный копчиковый ход, колоректальная хирургия, дерматовенерология

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Pilonidal sinus disease or pilonidal cyst is the fourth most common disease among inpatient stays in a coloproctological unit. Today the gold-standard treatment is surgical excision of the cavity followed by closure of the wound with sutures [1–3]. According to studies pilonidal disease can be seen in combination with hidradenitis suppurativa in 35 % of cases [4]. Therefore, hidradenitis suppurativa in combination with acne conglobata and dissecting cellulitis of the scalp represent follicular occlusion syndrome [5]. The basis of the pathogenesis of these diseases, including pilonidal, from the dermatovenerologists' point of view, is occlusion of the hair follicle, followed by local inflammation in the dermis and subcutaneous fat [6, 7].

Follicular occlusion syndrome (FOS) includes four separate diseases: hidradenitis suppurativa, acne conglobata, dissecting cellulitis of the scalp, and pilonidal disease. These diseases can occur either isolated or combined with each other making up follicular occlusion dyad, triad, or tetrad [8]. Despite different localization all skin manifestations are characterized by similar pathological characteristics [9]. The most common manifestation of FOS is acne conglobata, which in approximately 50 % of patients coexists with hidradenitis suppurativa, in 35 % — with pilonidal sinus disease and in 6–8 % — with dissecting cellulitis [12]. Four of these diseases together are seen extremely rare [9–11].

In addition to the natural difficulties associated with the diagnosis and subsequent treatment of FOS caused by the fact that these diseases belong to different medical specialties, the question of their joint treatment inevitably arises. In addition, it would be appropriate to search for

answers to such questions as the effectiveness of traditional methods of treating pilonidal sinus disease when it coexists with dermatological manifestations of FOS and the possibility of using traditional methods of surgical treatment of dermatological manifestations of the syndrome in the practice of treating pilonidal cysts.

Materials and methods

Eighty patients with pilonidal disease had been operated in the Clinic of Coloproctology and Minimally Invasive Surgery from November 2018 to December 2019, of those 62 (77.5 %) — with primary pilonidal disease and 18 (22.5 %) — with a recurrent cyst. The average age of the patients was 28.0 ± 3.8 years.

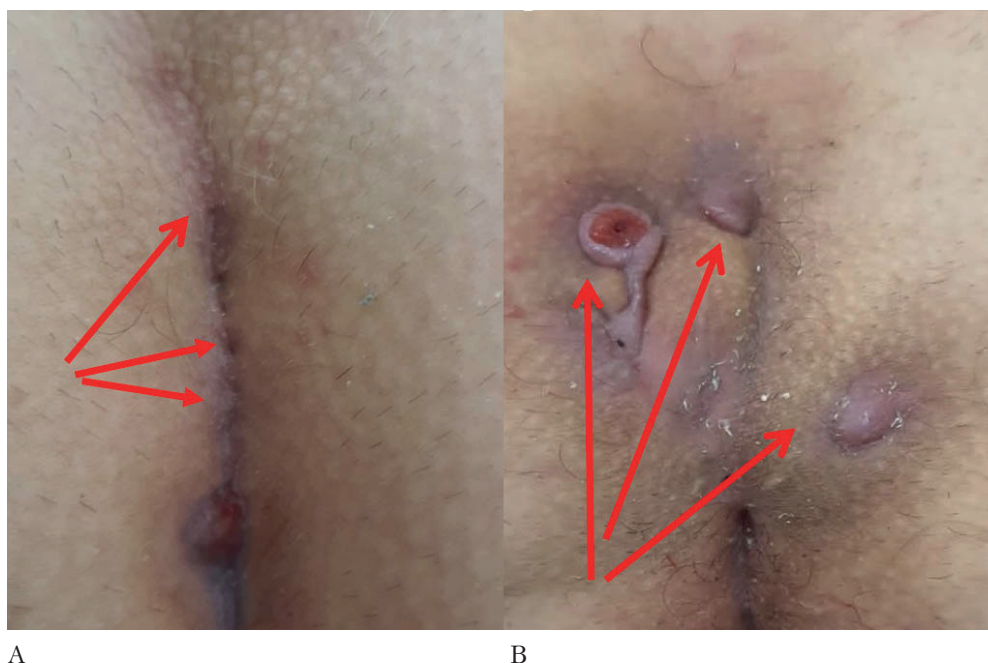
Clinical picture

The clinical picture in all cases of primary pilonidal disease was the appearance of mucoid or purulent drainage from primary and secondary openings located in the sacrococcygeal region, mainly in the projection of the natal cleft.

The appearance of secondary orifices, localized at some distance from the gluteal cleft, indicates the periodical exacerbations with the appearance of a painful induration on the skin with subsequent release of purulent discharge (abscess) and general improvement of the patient's condition.

Out of 80 observations, 6 (9.7 %) patients with primary and one (5.6 %) patient with recurrent pilonidal disease had concomitant manifestations of follicular occlusion syndrome (Table). Thus, the frequency of pilonidal sinus disease coexisting with other variants of FOS was 8.8 %.

In 5 out of 7 patients hidradenitis suppurativa of axillary and inguinal areas was identified, with



A

B

Figure 1. Clinical variations of pilonidal sinus: A — primary pilonidal sinus disease (arrows indicate primary orifices); B — recurrent pilonidal sinus disease with secondary orifices and leaks (arrows indicate secondary orifices located lateral to the natal cleft)

Рисунок 1. Возможные клинические варианты ЭКХ: А — первичный эпителиальный копчиковый ход (стрелками указаны первичные отверстия); В — эпителиальный копчиковый ход с множественными вторичными отверстиями и затеками (стрелками указаны вторичные отверстия, расположенные латеральнее межъягодичной складки)

which patients had previously sought emergency help in a surgical hospital (Fig. 2, 3). Three patients had previously undergone surgery with incision and drainage for acute lesions of hidradenitis suppurativa. Two patients were prescribed systemic antibacterial and symptomatic therapy by a dermatologist. Patients noted a clinically significant improvement in their condition during treatment in the form of decrease in pain and the amount of discharge. Acne conglobata, as one of the components of FOS, was noted in three patients (Fig. 4). Dissecting cellulitis of the scalp was diagnosed in one patient (Fig. 5).

Diagnostics

On the ultrasound examination of the sacrococcygeal region soft tissues (Fig. 6) pilonidal sinus is a hypoechoic cavity located directly under the skin, with hyperechoic inclusions (hair). The cavity is connected to the skin through sinus tracts opening with primary and secondary opening(s). The boundaries of the cavity are well-defined, without increased blood flow during doppler imaging.

In case of secondary pits patients are recommended to undergo pelvic magnetic resonance imaging (MRI) with intravenous contrast to exclude

additional leakage, as well as differentiate with anorectal fistula and presacral cyst.

On MRI pilonidal sinus can be visualized as an elongated tubular structure with a hypointense signal on a T2-weighted image with the presence of a cavity with a fibrous capsule located in the projection of the intergluteal fold directly under the skin in the subcutaneous tissue, which drains to the skin surface through sinus tracts (Fig. 7).

Treatment

Follicular occlusion syndrome was most often observed as the combination of hidradenitis suppurativa and pilonidal sinus disease. In turn follicular occlusion triad was indicated in two patients. Follicular occlusion tetrad was not observed (Table).

The extent of surgical treatment for all patients was excision of the pilonidal sinus, followed by closure of the resulting defect with local tissues by lateral displacement of the wound from the median intergluteal fold using Bascom II procedure (Fig. 8).

Postoperative period

Dressings with antiseptic solutions, antibacterial and symptomatic therapy were carried out daily in the postoperative period. One patient had

Table. Comparative characteristics of patients with follicular occlusion syndrome and pilonidal sinus disease
Таблица. Сравнительная характеристика пациентов с синдромом фолликулярной окклюзии и ЭКХ

Criteria <i>Критерии</i>	Patients / <i>Пациенты</i>						
	C., 2019	G., 2019	C., 2020	P., 2020	Ch., 2021	V., 2022	D., 2022
Age, years <i>Возраст, годы</i>	24	43	29	32	28	51	44
Diagnosis before surgery <i>Диагноз до операции</i>	Recurrence / <i>Рецидив</i>	Primary pilonidal sinus / <i>Первичный ЭКХ</i>					
Manifestation of follicular occlusion syndrome <i>Проявления СФО:</i>							
- acne conglobata <i>- конглобатные акне</i>	+	—	—	+	—	+	—
- hidradenitis suppurativa (HS) <i>- гнойный гидраденит (ГГ)</i>	+	+	+	—	+	—	+
- dissecting cellulitis <i>- рассекающий целлюлит</i>	—	—	—	+	—	—	—
- pilonidal cyst <i>- пилонидальная киста</i>	+	+	+	+	+	+	+
HS Hurley stages <i>Стадия ГГ по Hurley</i>	II	I	I		III		II
HS surgical treatment <i>Хирургическое лечение ГГ</i>	—	—	+		+		+
Histological examination after excision of pilonidal sinus <i>Гистологическое исследование после иссечения ЭКХ</i>	Granulation tissue <i>Грануляционная ткань</i>	Cavity lined with keratinized stratified squamous epithelium. In the dermis, single sebaceous and sweat glands, as well as hair follicles, are identified. Focal scanty inflammatory infiltration, represented by lymphohistocytic elements <i>Полость, выстланная многослойным плоским ороговевающим эпителием. В дерме определяют единичные сальные и потовые железы, а также волосные фолликулы. Очаговая скудная воспалительная инфильтрация, представленная лимфо-гистоцитарными элементами</i>					
Contacting a dermatovenerologist and carrying out conservative treatment of HS after surgical treatment of pilonidal sinus <i>Обращение к дерматовенерологу и проведение консервативного лечения ГГ после хирургического лечения ЭКХ</i>	+	+	+		—		—
Recurrence of pilonidal sinus after surgical treatment <i>Рецидив ЭКХ после хирургического лечения</i>	—	—	—	—	—	—	—

a divergence of the lower third of the wound on the seventh day after surgery, which was associated with recommended physical activity disorder.

In other patients no complications from the post-operative wound were noted. Suture removal terms were 13.0 ± 2.8 days.

Morphologic study of specimen

It was shown by morphologic study of specimen of primary PSD that the structure located

in the subcutaneous fat tissue was represented by a cleft cavity with multilayer squamous keratinizing epithelium of different thickness with the presence of areas of necrobiosis and necrosis of epitheliocytes (Fig. 9). A thin layer of granulation tissue with scattered and focal weak, in some places moderately expressed infiltration by polymorphonuclear leukocytes with admixture of lymphocytes, plasma cells and macrophages were also determined. Hair rods and their fragments



Figure 2. Hidradenitis suppurativa of the left inguinal and pubic region, Hurley stage II (medium severity): A — acute stage (limited not connected sinus tracts with purulent discharge); B — status localis after 1 month of conservative therapy (decrease the volume of purulent discharge, severity of inflammation, subcutaneous sinus tracts remain)

Рисунок 2. Гнойный гидраденит левой паховой и лобковой областей, стадия II по Hurley (средняя степень тяжести): А — стадия обострения (отмечаются не взаимосвязанные между собой подкожные свищевые ходы с гнойным отделяемым, воспалительные узлы); В — локальный статус через 1 месяц комбинированной терапии (отмечается уменьшение объема гнойного отделяемого, выраженности воспаления, подкожные свищевые ходы сохраняются)



Figure 3. Hidradenitis suppurativa of the right axillary region, Hurley stage II (medium severity): A — acute stage (a single subcutaneous sinus tract); B — status localis 12 months after conservative therapy (decrease of inflammatory changes, atrophic scar at the site of subcutaneous sinus tract)

Рисунок 3. Гнойный гидраденит правой подмышечной области, стадия II по Hurley (средняя степень тяжести): А — стадия обострения (отмечается наличие единичного подкожного свищевых хода и патогномоничного для заболевания шнуровидного рубца); В — локальный статус через 12 месяцев комбинированной терапии (отмечается уменьшение воспалительных изменений, атрофический рубец на месте подкожного свищевых хода)

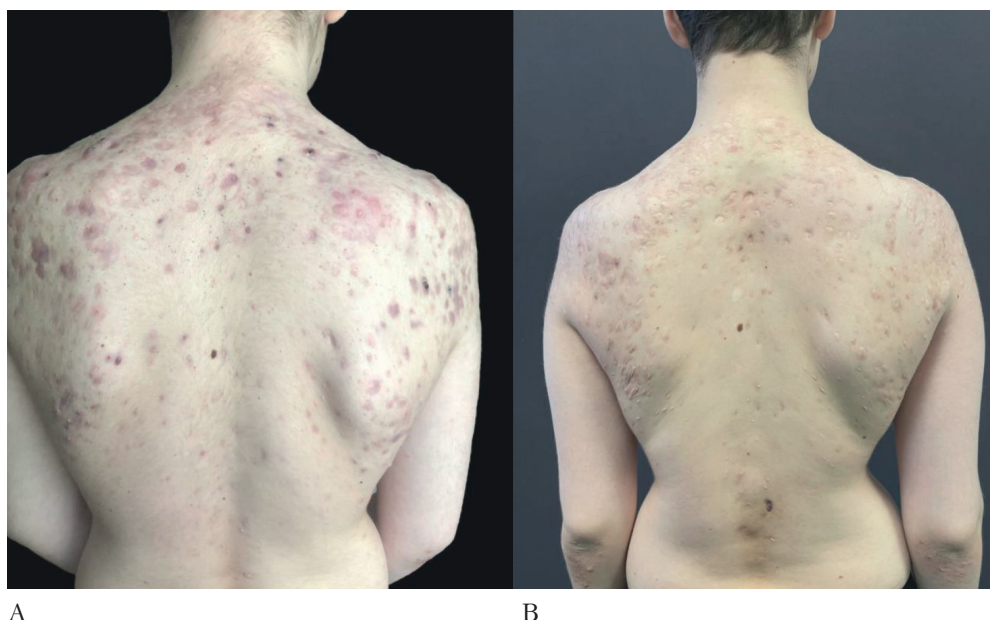


Figure 4. Acne conglobata: A — acute stage (multiple inflammatory nodules, open comedones, status before therapy); B — status localis 6 months after conservative therapy (multiple atrophic scars in the treatment outcome, single remaining inflammatory elements, dynamic monitoring of the patient continues)

Рисунок 4. Конглобатные акне: А — стадия обострения (отмечаются многочисленные воспалительные узлы, открытые комедоны, состояние до начала терапии); В — локальный статус через 6 месяцев комбинированной терапии (множественные атрофические рубцы в исходе лечения, единичные сохранившиеся воспалительные элементы, динамическое наблюдение за пациентом продолжается)



Figure 5. Dissecting cellulitis (Hoffman's perifolliculitis capitis abscedens et suffodiens) of the scalp: A — before treatment (focal hair loss in the area where the inflammatory nodule is located); B — status localis 1 month after conservative therapy (reduction of erythema and inflammation expression, partial resumption of hair growth)

Рисунок 5. Рассекающий целлюлит (абсцедирующий и подрывающий фолликулит и перифолликулит Гоффмана) области бороды: А — до начала лечения (очаговая потеря волос в зоне расположения воспалительного узла); В — локальный статус через 1 месяц на фоне комбинированной терапии (уменьшение выраженности эритемы и воспаления, частичное возобновление роста волос)

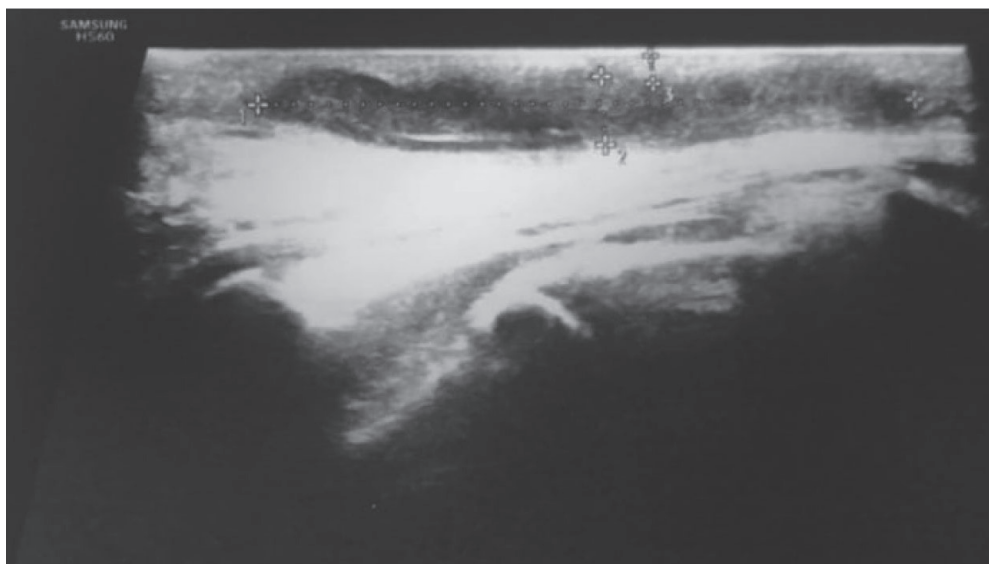


Figure 6. Cavity of pilonidal sinus according to ultrasound

Рисунок 6. Полость ЭКХ по данным ультразвукового исследования

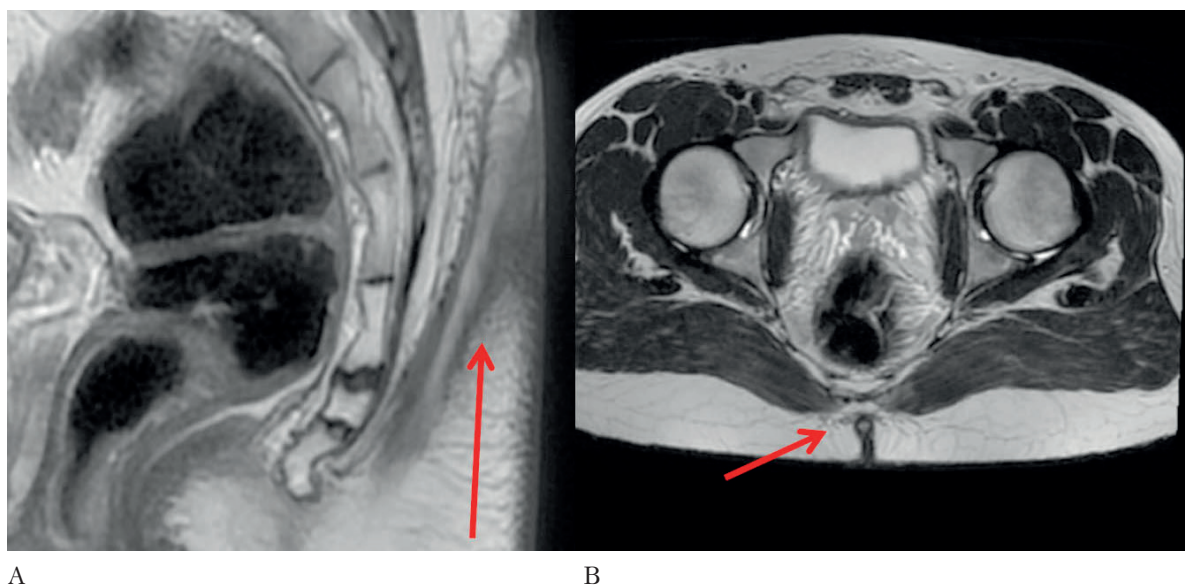


Figure 7. Magnetic resonance imaging of the pelvic organs with intravenous contrast: A — sagittal view T2-weighted image (the arrow indicates the cavity. A layer of subcutaneous fatty tissue was preserved between the cavity and sacral fascia); B — axial view T2-weighted image (arrow indicates cavity)

Рисунок 7. МРТ органов малого таза с внутривенным контрастированием: А — сагиттальная проекция Т2-ВИ (стрелкой указана полость ЭКХ. Между полостью и крестцовой фасцией сохранена прослойка подкожно-жировой клетчатки); В — аксиальная проекция Т2-ВИ (стрелкой указана полость ЭКХ)

were found in the lumen of the cavity in 4 out of 6 patients, besides, hair follicles were visualized in the walls of the cyst. Moderate hyperemia of vessels, focal edema, focal perivascular lymphohistiocytic infiltration were observed in the

preserved dermal tissue adjacent to the PSD. The recurrent cavity lining over a large length was represented by a wide layer of granulation tissue, represented by newly formed hyperemic vessels with hydropic walls, located in the hydropic

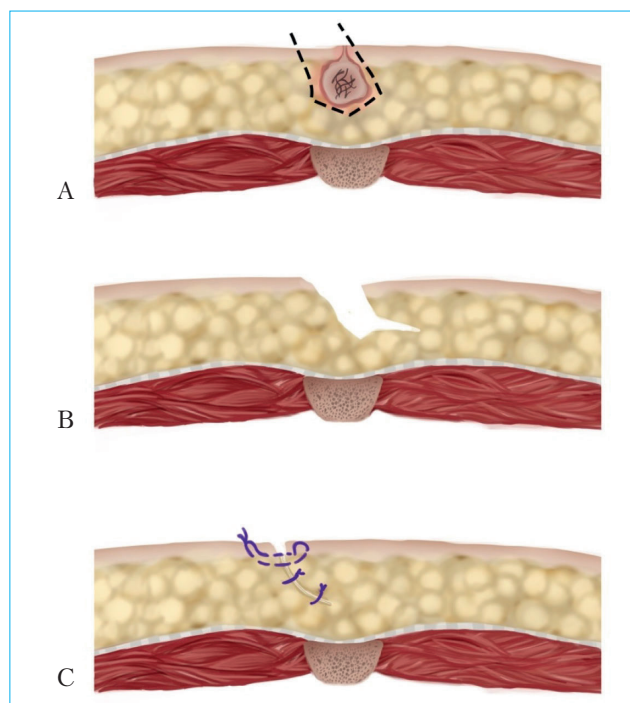


Figure 8. Operation scheme: A — the border of cavity resection is indicated by the dotted line; B — mobilized cutaneous-subcutaneous flap formed to close the defect formed after excision laterally the natal cleft; C — layer-by-layer sutured wound

Рисунок 8. Схема операции: А — пунктирной линией обозначена граница резекции полости ЭКХ; В — мобилизованный кожно-подкожный лоскут, сформированный с целью закрытия образовавшегося дефекта после иссечения; С — послойно ушитая рана

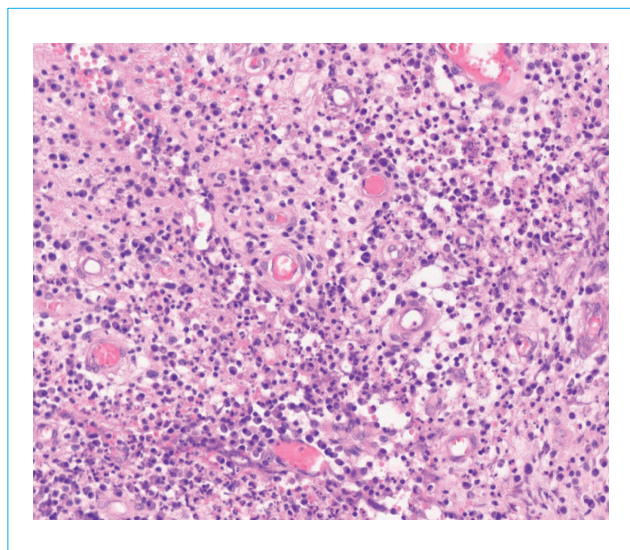


Figure 10. Recurrent pilonidal sinus disease. Granulation tissue. Hematoxylin and eosin staining, × 400

Рисунок 10. Рисунок 10. ЭКХ, рецидив заболевания. Грануляционная ткань. Окраска гематоксилином и эозином, × 400

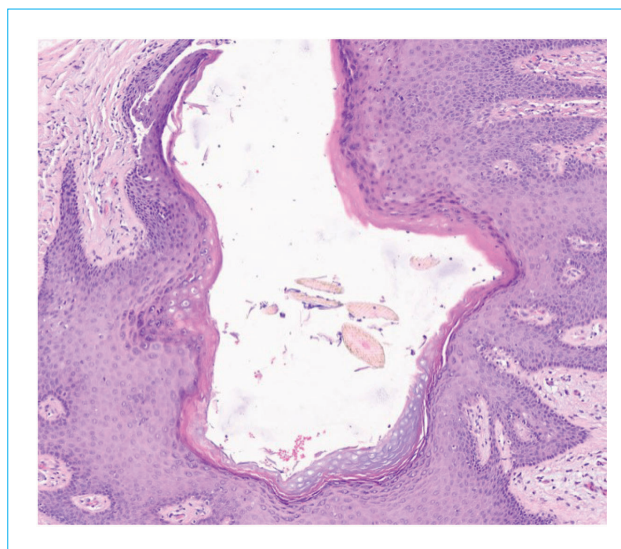


Figure 9. Primary pilonidal sinus with multi-layered squamous keratinous epithelium lining (hair rods are detected in the cavity). Hematoxylin and eosin staining, × 200

Рисунок 9. Первичный ЭКХ с выстилкой из многослойного плоского ороговевающего эпителия (в полости определяются волосные стержни). Окраска гематоксилином и эозином, × 200

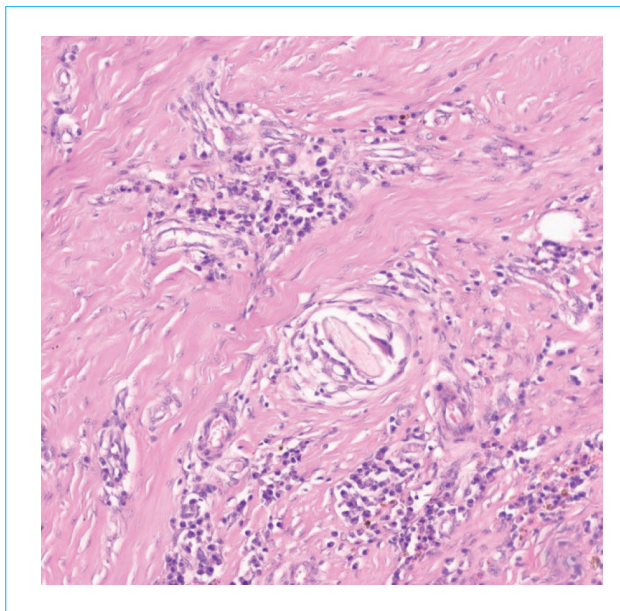


Figure 11. Recurrent pilonidal sinus disease. Site of sclerosis of pilonidal sinus, wall with a "walled" hair shaft; focal lympho-macrophage infiltration with the presence of giant multinucleated cells of foreign body resorption. Hematoxylin and eosin staining, × 200

Рисунок 11. ЭКХ, рецидив заболевания. Участок склероза стенки ЭКХ с «замурованным» волосным стержнем; очаговая лимфо-макрофагальная инфильтрация с наличием гигантских многоядерных клеток рассасывания инородных тел. Окраска гематоксилином и эозином, × 200

interstitial substance with abundant, predominantly diffuse lympho-plasmacytic infiltration with admixture of polymorphonuclear leukocytes and macrophages (Fig. 10, 11).

Long-term results

Currently, no recurrence have been noted in any observation; the average follow-up time is 14.0 ± 5.6 months (6–27 months). All patients lead an active lifestyle. During control examinations of the sacral region in all patients complete healing of the wound was determined without signs of relapse. In patients with concomitant manifestations of FOS on complex local and systemic conservative treatment under the supervision of dermatologist positive dynamics are observed including life quality improvement and long-term remission.

Discussion

In 1980, J. Bascom proposed the follicular-retention theory of the origin of the pilonidal sinus disease, which currently prevails in Western countries [13, 14]. According to this theory, the first link in the occurrence of PSD is acute purulent folliculitis, which is a “primary abscess” with minimal clinical manifestations. The appearance of fistulous outside the gluteal cleft is considered as a “secondary abscess” as a manifestation of trichogenic-pompous mechanism. Nine years later, A.V. Kulyapin described the results of histologic examination of PSD after excision and showed the obvious similarity of pilonidal disease with hidradenitis suppurativa in his thesis [14].

To be fair, it should be noted that a few years earlier, in 1975, G. Plewig, based on the similarity of pathologic manifestations, joined pilonidal disease to the triad of follicular occlusion syndrome, which at that time included hidradenitis suppurativa, acne conglobate, and dissecting cellulitis of the scalp [4, 15]. All these nosologic units can be seen alone and in combination with each other. Follicular occlusion triad has often been described in the literature, but references to the follicular occlusion tetrad which includes pilonidal sinus are found only in case reports [9, 10]. This can be explained due to the rarity of the combination of all four diseases and by the possibly of inadequate examination of the patient's skin in the sacrococcygeal region at a dermatologist's appointment and of the back, axillary and scalp regions by a coloproctologist. The last factor of follicular occlusion syndrome underdiagnosis is the lack of awareness among proctologists, surgeons specialized in purulent diseases and dermatologists who encounter each of these illnesses in their practice that the combination of follicular occlusion diseases is syndromic in nature and requires a multidisciplinary approach.

This is also important because within the framework of follicular occlusion syndrome development of these diseases, including pilonidal disease, is considered as a disruption of the pilosebaceous complex due to occlusion in the infundibulum of the hair follicle [5–7]. Subsequently, these processes lead to inflammation of the pilosebaceous unit, sinus tracts and abscesses formation and destruction of the sweat and sebaceous glands structure. The presence of opportunistic pathogens and biofilm formation provide a stronger and longer-lasting inflammatory response [7, 10]. According to some authors, dysregulation of the innate and acquired immunity also plays an important role in the development of pilonidal disease [16].

In contrast to the views of western colleagues, domestic surgeons give priority to the disorders of embryogenesis in pilonidal disease occurrence [14, 17, 18]. Indeed, pilonidal disease alone is usually located quite deep, connected to the anococcygeal ligament and has no more than 2–3 orifices located in the intergluteal cleft, which probably confirms the theory of disembryogenesis.

However, more and more often there are patients with pilonidal disease and follicular occlusion syndrome which are distinguished by a more superficial location of the pilonidal sinuses, several primary openings and, often, the presence of one or two secondary orifices located outside the intergluteal cleft. Despite the difference in location and probably origin, in all cases pilonidal disease has a similar morphological picture. Having analyzed the histological picture of removed coccygeal cavity, both in the pilonidal disease alone and as a part of follicular occlusion syndrome we can say that in all six observations the cavity of the primary pilonidal disease is lined with keratinized stratified squamous epithelium, which indirectly confirms the acquired theory of disease development. In inflammatory stages foci of necrosis and granulation tissue are observed in microslides, and the cavity almost always contains hair that are not attached to the epithelial lining. It is worth noting that despite an identical morphological picture, the clinical picture of pilonidal disease alone and pilonidal disease in follicular occlusion syndrome is different. The isolated congenital pilonidal disease is characterized by the presence of one or two primary orifices, which are located in the lower third of the intergluteal cleft, quite close to the anus.

According to MRI in most cases, “fixation” of the formed cavity to the coccygeal vertebrae is registered, defined in the form of heavy structures of hypointense MR signal on T2-VI, which is characteristic of fibrous tissue. If the formation of PSD is considered as an “acquired” form of the disease against the background of other manifestations of FOS, the primary orifice is located in the gluteal cleft more proximally or at a greater distance from the anus than in congenital PSD. From our

perspective, minimally invasive techniques such as laser destruction of pilonidal cyst (SiLaC) [19, 20], deroofing and curettage (LOCULA) [21] should be considered as the “operation of choice”. These procedures involve local destruction of the lining with desquamation of the multilayer squamous keratinizing epithelium and do not involve extensive excision of the surrounding and underlying tissues, making the treatment of PSD a “one-day surgery” or “lunch break surgery”.

At the same time, in dermatological practice, treatment of follicular occlusion syndrome is based on the use of symptomatic, antibacterial and pathogenetic therapy, and in the case of severe forms it comes to the use of immunosuppressive and genetically engineered biological agents [5, 8, 9, 11, 22]. Based on this, we suggest that an integrated approach to the treatment of patients with follicular occlusion syndrome including minimally invasive surgery for pilonidal disease and systemic drug therapy for combined nosologic units, can significantly shorten the postoperative period, reduce the

number of recurrence, and improve the quality of life of such patients.

Conclusion

The presented observations suggest that under the nosologic unit “pilonidal sinus disease” two different forms of this disease are hidden with an identical morphological picture. And it is not just a matter of creating a new classification system. From our point of view, this is a question of changing the approach to treating this disease. The limitation of this study is a small number of observations with a proven form of pilonidal disease combined with other manifestations of follicular occlusion syndrome and the lack of experience in treating this form using minimally invasive treatment methods. Despite this, the presented work makes it possible to change the view on the disease course and, as a result, to reasonably give preference to non-excisional, minimally invasive methods of treating this category of patients.

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Information about the authors

Daria D. Shlyk — Cand. Sci. (Med.), Associate Professor of the Department of Surgery, N.V. Sklifosovskiy Institute of Clinical Medicine, I.M. Sechenov First Moscow State Medical University (Sechenov University).
Contact information: shlikdarya@gmail.com;
119435, Moscow, Pogodinskaya str., 1, build. 1.
ORCID: <https://orcid.org/0000-0002-9232-6520>

Maria N Pikuz — Resident of the Department of Surgery, N.V. Sklifosovskiy Institute of Clinical Medicine, I.M. Sechenov First Moscow State Medical University (Sechenov University).
Contact information: Mashenka_pikuz@mail.ru;
119435, Moscow, Pogodinskaya str., 1, build. 1.
ORCID: <https://orcid.org/0000-0002-2680-9372>

Yuri E. Kitsenko* — Cand. Sci. (Med.), Associate Professor of the Department of Surgery, N.V. Sklifosovskiy Institute of Clinical Medicine, I.M. Sechenov First Moscow State Medical University (Sechenov University).
Contact information: yury@kitsenko.ru;
119435, Moscow, Pogodinskaya str., 1, build. 1.
ORCID: <https://orcid.org/0000-0002-4415-6141>

Anna S. Pirogova — Postgraduate of the Department of Skin and Venereal Diseases, I.M. Sechenov First Moscow State Medical University (Sechenov University).
Contact information: annese@mail.ru
119435, Moscow, Bolshaya Pirogovskaya str., 4, build. 1.
ORCID: <https://orcid.org/0000-0002-2246-1321>

Nina B. Paramonova — Cand. Sci. (Med.), Associate Professor of the Institute of Clinical Morphology and Digital Pathology, I.M. Sechenov First Moscow State Medical University (Sechenov University).
Contact information: paramonova_n_b@staff.sechenov.ru;
119435, Moscow, Abrikosovsky lane, 1.
ORCID: <https://orcid.org/0000-0001-5380-7113>

Сведения об авторах

Шлык Дарья Дмитриевна — кандидат медицинских наук, доцент кафедры хирургии Института клинической медицины им. Н.В. Склифосовского ФГАОУ ВО «Первый Московский государственный медицинский университет им. И.М. Сеченова» Министерства здравоохранения Российской Федерации (Сеченовский Университет).
Контактная информация: shlikdarya@gmail.com;
119435, г. Москва, ул. Погодинская, 1, стр. 1.
ORCID: <https://orcid.org/0000-0002-9232-6520>

Пикуза Мария Николаевна — ординатор кафедры хирургии Института клинической медицины им. Н.В. Склифосовского ФГАОУ ВО «Первый Московский государственный медицинский университет им. И.М. Сеченова» Министерства здравоохранения Российской Федерации (Сеченовский Университет).
Контактная информация: Mashenka_pikuz@mail.ru;
119435, г. Москва, ул. Погодинская, 1, стр. 1.
ORCID: <https://orcid.org/0000-0002-2680-9372>

Киценко Юрий Евгеньевич* — кандидат медицинских наук, доцент кафедры хирургии Института клинической медицины им. Н.В. Склифосовского ФГАОУ ВО «Первый Московский государственный медицинский университет им. И.М. Сеченова» Министерства здравоохранения Российской Федерации (Сеченовский Университет).
Контактная информация: yury@kitsenko.ru;
119435, г. Москва, ул. Погодинская, 1, стр. 1.
ORCID: <https://orcid.org/0000-0002-4415-6141>

Пирогова Анна Сергеевна — аспирант кафедры кожных и венерологических болезней им. В.А. Рахманова Института клинической медицины им. Н.В. Склифосовского ФГАОУ ВО «Первый Московский государственный медицинский университет им. И.М. Сеченова» Министерства здравоохранения Российской Федерации (Сеченовский Университет).
Контактная информация: annese@mail.ru;
119435, г. Москва, ул. Большая Пироговская, 4, стр. 1.
ORCID: <https://orcid.org/0000-0002-2246-1321>

Парамонова Нина Борисовна — кандидат медицинских наук, доцент Института клинической морфологии и цифровой патологии ФГАОУ ВО «Первый Московский государственный медицинский университет им. И.М. Сеченова» Министерства здравоохранения Российской Федерации (Сеченовский Университет).
Контактная информация: paramonova_n_b@staff.sechenov.ru;
119435, г. Москва, Абрикосовский пер., 1.
ORCID: <https://orcid.org/0000-0001-5380-7113>

* Corresponding author / Автор, ответственный за переписку

Ramin T. Rzaev — Cand. Sci. (Med.), Radiologist at the Department of Radiation Diagnostics, the University Clinical Hospital No. 2, I.M. Sechenov First Moscow State Medical University (Sechenov University).

Contact information: ramin-rz@mail.ru;
119435, Moscow, Pogodinskaya str., 1, build. 1.
ORCID: <https://orcid.org/0000-0002-6005-6247>

Natalia P. Teplyuk — Dr. Sci. (Med.), Professor of the Department of Skin and Venereal Diseases, I.M. Sechenov First Moscow State Medical University (Sechenov University).

Contact information: teplyukn@gmail.com;
119435, Moscow, Bolshaya Pirogovskaya str., 4, build. 1.
ORCID: <https://orcid.org/0000-0002-5800-4800>

Petr V. Tsarkov — Dr. Sci. (Med.), Professor, Head of the Department of Surgery, N.V. Sklifosovskiy Institute of Clinical Medicine, I.M. Sechenov First Moscow State Medical University (Sechenov University).

Contact information: tsarkov@kkmx.ru;
119435, Moscow, Pogodinskaya str., 1, build. 1.
ORCID: <https://orcid.org/0000-0002-7134-6821>

Рзаев Рамин Теймурхан оглы — кандидат медицинских наук, врач-рентгенолог отделения лучевой диагностики Университетской клинической больницы № 2 ФГАОУ ВО «Первый Московский государственный медицинский университет им. И.М. Сеченова» Министерства здравоохранения Российской Федерации (Сеченовский Университет).

Контактная информация: ramin-rz@mail.ru;
119435, г. Москва, ул. Погодинская, 1, стр. 1.
ORCID: <https://orcid.org/0000-0002-6005-6247>

Теплюк Наталия Павловна — доктор медицинских наук, профессор кафедры кожных и венерологических болезней им. В.А. Рахманова Института клинической медицины им. Н.В. Склифосовского ФГАОУ ВО «Первый Московский государственный медицинский университет им. И.М. Сеченова» Министерства здравоохранения Российской Федерации (Сеченовский Университет).

Контактная информация: teplyukn@gmail.com;
119435, г. Москва, ул. Большая Пироговская, 4, стр. 1.
ORCID: <https://orcid.org/0000-0002-5800-4800>

Царьков Петр Владимирович — доктор медицинских наук, профессор, заведующий кафедрой хирургии Института клинической медицины им. Н.В. Склифосовского ФГАОУ ВО «Первый Московский государственный медицинский университет им. И.М. Сеченова» Министерства здравоохранения Российской Федерации (Сеченовский Университет).

Контактная информация: tsarkov@kkmx.ru;
119435, г. Москва, ул. Погодинская, 1, стр. 1.
ORCID: <https://orcid.org/0000-0002-7134-6821>

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