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Utility of transperineal and anal ultrasonography in the diagnostics of hidradenitis suppurativa and its differentiation from a rectal fistula

Użyteczność przezkroczowej i przezodbytniczej ultrasonografii w diagnostyce i różnicowaniu zapalenia gruczołów apokrynowych z przetoką odbytu

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Authors' Contribution:
A Study Design
B Data Collection
C Statistical Analysis
D Data Interpretation
E Manuscript Preparation
F Literature Search
G Funds Collection

Summary

Introduction: The pathogenesis of hidradenitis suppurativa (HS) is not fully understood. There exist several theories, in which mechanical factors, genetic factors, as well as immunological dysfunction of lymphocytes are suspected. Clinically, this entity is frequently mistaken for anal fistula with consequently wrong treatment.

We aim to determine the utility of transperineal ultrasound (TPUS) and anal ultrasound (AUS) in the diagnosis of HS and its differentiation from an anal fistula.

Material/Methods: Retrospective analysis was performed on 51 patients (5 females, 46 males) aged 20–71 years (mean age 47.5), who were operated on in the years 2006–2011 for HS in the area of the anus and perineum, and pre-operatively had been imaged with TPUS and AUS. Sixty-seven operations were analyzed, as 11 patients were operated on more than once due to HS recurrence.

Results: In 66 out of 67 cases (98.5%), the pre-operative TPUS and AUS were in accordance with the intraoperative findings. Only in 1 patient was a pilonidal cyst diagnosed intraoperatively. In all 67 patients, the TPUS showed typical fluid-solid changes localized in the subcutaneous adipose tissue. In 6 out of 67 cases of HS (8.9%) AUS showed an anal fistula coexisting with the HS. In 2 cases (2.9%) a skin malignancy coexisting with HS was found.

Discussion: TPUS is an accessible imaging method, which confirms the typical localization of changes of HS, and together with AUS it allows for the proper differentiation of HS from an anal fistula or an abscess.

Key words: **hidradenitis suppurativa • anal fistula • pilonidal cyst • anal ultrasound • transperineal ultrasound • squamous cell carcinoma**

Streszczenie

Wstęp: Patogeneza zapalenia gruczołów apokrynowych nie jest dobrze poznana. Jest wiele teorii, w tym teorie mechaniczne, teorie podkreślające czynnik genetyczny, oraz teorie immunologiczne biorące

pod uwagę dysfunkcje limfocytów. Klinicznie choroba często jest mylona z przetoką odbytu i w konsekwencji źle leczona.

Cel pracy. Określenie przydatności ultrasonografii przezkroczowej (TPUS-transperineal ultrasound) oraz przezodbytniczej (AUS – anal ultrasound) w różnicowaniu zapalenia gruczołów apokrynowych (HS-hidradenitis suppurativa) z przetoką odbytu i ropniem.

Materiał/Metody: Ocenie retrospektywnej poddano 51 pacjentów (5 kobiet, 46 mężczyzn) w wieku 20–71 lat (średnia wieku 47,5 lat) operowanych w latach 2006–2011 z powodu zapalenia gruczołów apokrynowych okolicy odbytu i krocza, u których wykonano TPUS i AUS. Analizie poddano 67operacji, gdyż 11 pacjentów było operowanych więcej niż 1 raz z powodu nawrotu choroby.

Wyniki: U 66 z 67 pacjentów (98,5%) przedoperacyjne badanie TPUS I AUS było zgodne z obrazem śródoperacyjnym. Tylko u 1 pacjenta stwierdzono śródoperacyjnie torbiel włosową. U wszystkich 67 pacjentów obraz TPUS przedstawiał typowe zmiany płynowo-lite zajmujące podskórną tkankę tłuszczową. U 6 z 67 pacjentów z HS (8,9%) AUS przedstawiał przetokę odbytu współistniejącą z HS. W 2 przypadkach (2,9%)stwierdzono nowotwór skóry współistniejący z HS.

Wnioski: TPUS jest dostępną metodą obrazową, która potwierdza typową lokalizację zmian w przebiegu HS, umożliwiając ich pewne zróżnicowanie z przetoką lub ropniem odbytu.

Słowa kluczowe: **zapalenie gruczołów apokrynowych • przetoka odbytu • przetoka włosowa • ultrasonografia transrektalna • rak odbytu**

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Abbreviations: **AUS** – anal ultrasound; **HS** – hidradenitis suppurativa; **TPUS** – transperineal ultrasound.

Introduction

Inflammation of apocrine glands around the anus (hidradenitis suppurativa, HS) is a chronic condition that is difficult to treat and often recurs. It most often appears after puberty, in the 2nd–3rd decade of life, three times more often in females than males. The changes may be of variable severity, meaning they can be single or multiple, and may be distributed in the axillary, groin, gluteal, perineal and other regions of the body where apocrine glands are located [14,17,21]. The entity was first described by Velpeau in 1839, in a patient with multiple suppurative changes in several locations. In 1854, based on a series of patients, Verneuil described inflammation of the apocrine glands in detail, and was the first to associate it with sweat glands; the disease is also known under his surname (Verneuil's disease). Only in 1922 did Schiefferdecker, through histopathology, confirm the possible association of the disease with apocrine glands [11,14,23].

The pathogenesis of HS is not fully understood [22]. There exist several theories, in which an infective factor is rarely mentioned. Most often mechanical factors are suspected, meaning occlusion of the hair follicle (much more rarely the apocrine gland, only in 5% of HS cases), with subsequent stasis of secretions and their infection [3,11,12]. There are also theories involving a genetic factor, which is found in up to 26% of cases [7,9,11,22]. An interesting hypothesis presented by Boer and Weltevreden states that due to an immunological dysfunction of lymphocytes, the inflammation spreads from the hair follicle to the apocrine glands, causing an inverse inflammation of the hair follicles (so-called *acne inversa*) [4,20].

HS accounts for approximately 5% of pustular changes of the anorectal area [15]. The anorectal localization of HS is more common in males, and the disease may be manifested by pain, swelling, purulent discharge, pruritus or bleeding and can mimic several common problems, such as furunculosis, anal fistula, pilonidal disease, perianal abscess or Crohn's disease [14]. Due to the presence of external openings to the skin, the inflammatory changes often resemble an anal fistula; thus patients are frequently referred for surgery with such a diagnosis (Fig. 1). Rarely, a fistula to the anal canal may coexist with HS; in such cases it extends only into the lower portion of the anal canal, below the dentate line [6,14].

Aside from the proctological examination, anal ultrasound (AUS) and transperineal ultrasound (TPUS) imaging play a decisive role in the preoperative diagnosis of HS [5,16]. AUS confirms the presence or absence of

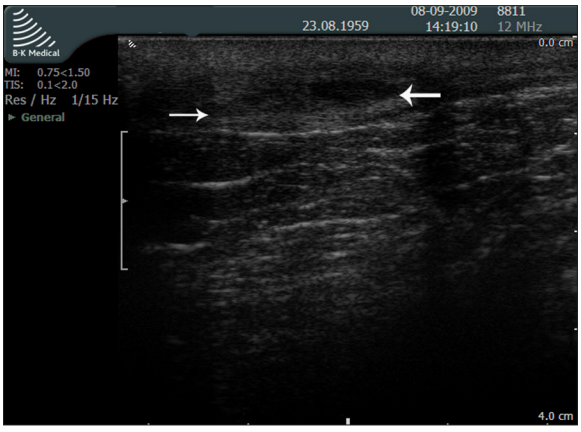

Fig. 1. Transperineal ultrasound: fluid-solid lesion within the subcutaneous tissue of the perianal area, which had developed within the course of HS (between arrows)

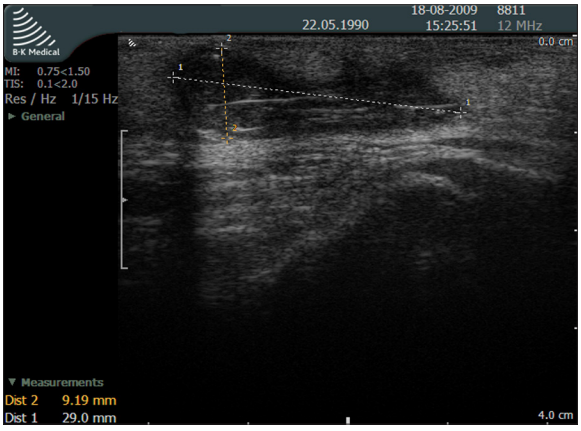

Fig. 2. TPUS showing a lesion with picture similar to HS, measuring 29×10 mm (between crosses) – histopathology showed the lesion to be a pilonidal cyst

continuity between the inflammatory changes and the anal canal, while TRUS confirms the typical subcutaneous location of changes.

The aim of this study was to analyze, on the basis of a retrospective review of own material, the utility of TPUS and AUS in the diagnosis of HS and its differentiation from anal fistula.

Materials and Methods

Retrospective analysis was performed on 51 patients (5 females, 46 males) aged 20–71 years (mean age 47.5), who were operated on in the years 2006–2011 for HS of the anus and perineum, in a reference center dealing with diagnosis and treatment of proctological diseases. None of the patients suffered from Crohn's disease. All patients had undergone a proctological examination, rectoscopy, TPUS and AUS imaging pre-operatively. The ultrasounds were performed using a BK Medical scanner, Profocus 2202, with a 8811 linear 6–12 MHz and 2050 type 3D mechanical volumetric endoprobe of 10–16 MHz frequency. On the basis of these 51 patients, 67 operations were analyzed, as 11 patients were operated on more than once (9 patients were operated on twice, one three, and one even seven times). The following procedures were performed in

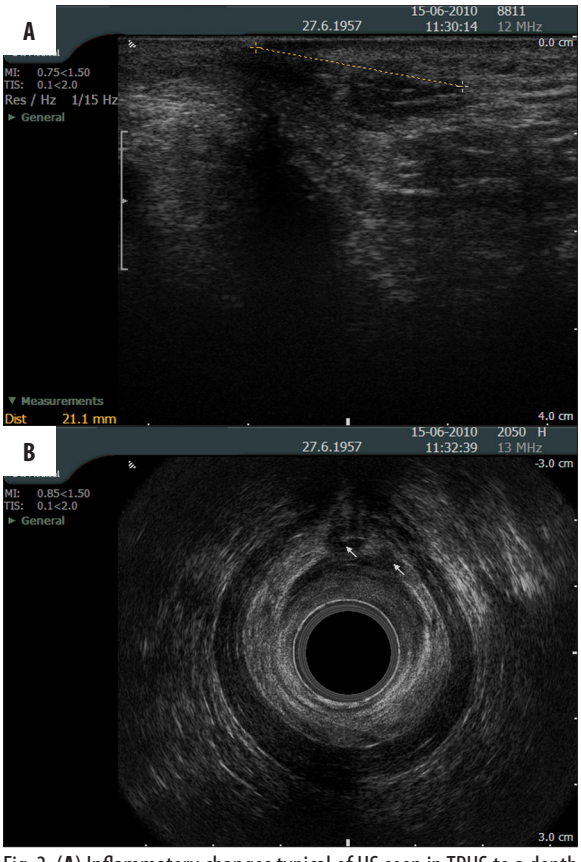

Fig. 3. (**A**) Inflammatory changes typical of HS seen in TPUS to a depth of 21 mm (between crosses); (**B**) with a low, multiple anterior transsphincteric fistula in AUS (arrows)

the above-listed patient group: deep excision of the changes to the depth of the fascia with wound closure and Redon drainage, excision of the lesions and wound closure using skin flaps, and multi-stage operations in cases of very widespread inflammatory changes. If there was co-existence of an anal fistula, excision of the fistula was done concomitantly, usually with fistulectomy, but in one patient using the Hippocratic technique. The pilonidal cyst was treated surgically through excision with wound closure and Redon drainage.

All patients gave their written informed consent with permission for publication of any of the material concerning them.

Results

In all 67 surgical cases, TPUS showed typical fluid-solid changes localized in the subcutaneous adipose tissue (Fig. 1). In 1 out of 67 changes, such an ultrasonographic picture corresponded to a pilonidal cyst diagnosed intraoperatively confirmed through surgery and histopathology (Fig. 2).

In 6 out of 67 HS-type changes (8.9%), the AUS study showed co-existence of an anal fistula, with 1 patient having multiple fistulas (Fig. 3); all of these findings were confirmed intraoperatively. Four patients (5.9%) with intraoperatively confirmed HS were initially referred for an ultrasound study with suspicion of a fistula or anal abscess,

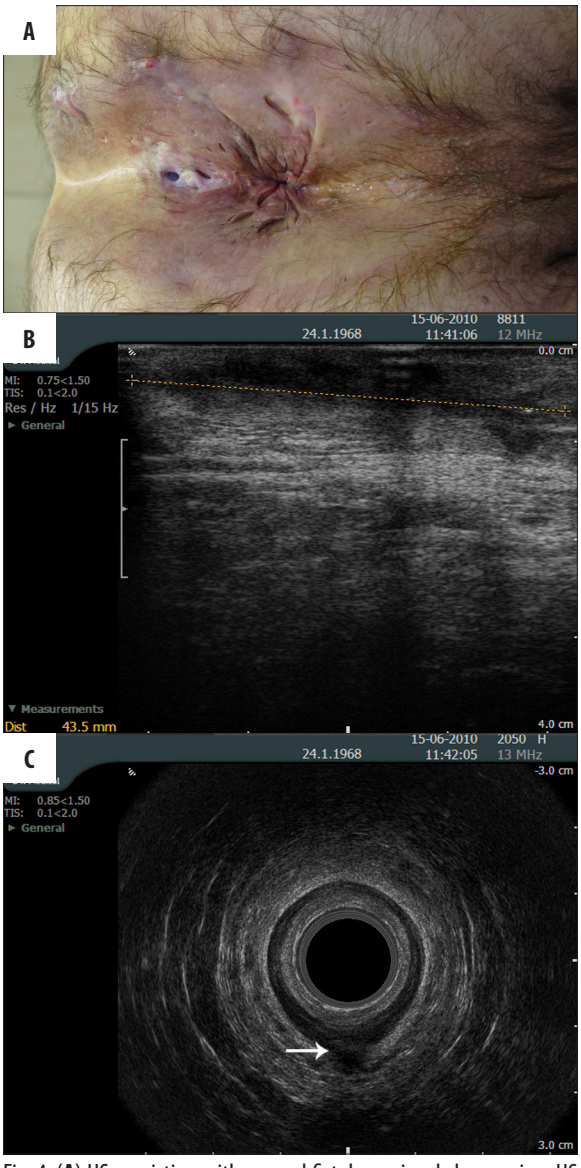

Fig. 4. (**A**) HS coexisting with an anal fistula; perianal changes in a HS patient and external outlets of a posterior anal fistula; (**B**) TPUS showing fluid-solid changes in the subcutaneous tissue measuring 43 mm (between the pluses) representing HS, (**C**) with a low posterior transsphincteric anal fistula with an internal orifice at the level of the distal end of the internal anal sphincter

from which 1 patient (1.5%) had been previously operated on for an anal fistula. In this group of 4 patients, only 1 had a fistula found in AUS imaging (Fig. 4). In 2 patients (2.9%) with HS, the postoperative histopathological examination revealed squamous cell carcinoma (SCC). On the basis of taken specimens recurrent HS was diagnosed in 11 patients (21.5%).

Discussion

Apocrine gland inflammation of the anal region requires initial differentiation from an anal fistula, particularly due to the similar clinical picture of both entities. On one hand, the internal orifice of the fistula could close, which will not allow for the exclusion of a fistula solely on the basis of a proctological examination. On the other hand, the external to the skin around the anus found in HS could imitate the external opening of a fistula (Fig. 5). TPUS is extremely helpful in this differentiation, and shows the superficial location of liquid/fluid-solid changes in HS. Meanwhile, AUS can confirm the presence of a fistula running towards the anal canal and in many cases shows its internal orifice [5,16].

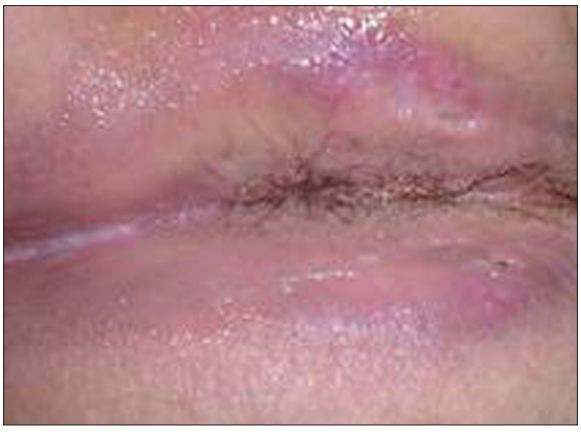

Fig. 5. Inflammatory changes characteristic for HS in the perianal area with external openings suggestive of an anal fistula

The above presented data show the preoperative ultrasonographic findings to be compatible with the intraoperative ones in 98.5% of cases. In one case, the lesion turned out to be a pilonidal cyst. It should be mentioned that the ultrasonographic examination can only evaluate the morphology and location of changes which are identical in the case of HS and a pilonidal cyst. Even the intraoperative assessment of low-grade HS changes could be ambiguous, especially in males with a hairy perianal area. In such cases, the histopathological examination serves to differentiate the two, and carries certain consequences as the patient must be moved from a lithotomy position to prone.

The co-existence of HS with an anal fistula occurs when inflammatory changes occupy the anal canal and lead to a secondary anal fistula, or the anal fistula could form independently of Verneuil's disease. In our study sample, the coexistence of HS and an anal fistula occurred in 8.9% of cases, among which the fistula was recognized in only 1 case through the proctological exam (the remaining 3 out of 4 clinical suspicions of a fistula turned out to be HS). The percentage of anal fistulas discovered in our patient group was surprisingly high, given that only individual cases of anal fistulas occurring due to HS have been published [1,13].

In 2 patients with a course of HS lasting several years (3% of patients) squamous cell carcinoma (SCC) of the skin was discovered (Fig. 6). This is in accordance with the findings of other authors, who indicate an increased risk for the development of skin cancer in long-standing HS (mean length of 20 years) and emphasize the need for greater awareness that long-standing gluteal or perineal HS is a premalignant condition which should not be managed conservatively [2,10,11,19].

HS-type changes in the perianal area are particularly difficult to treat. Patients are often misdiagnosed, reach a

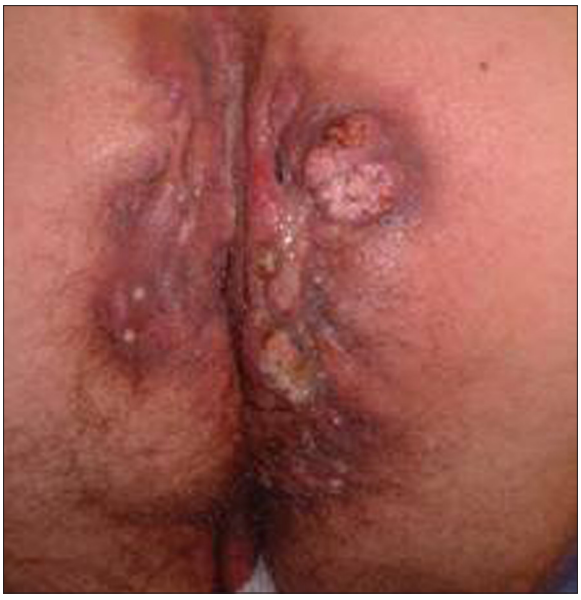

Fig. 6. Squamous cell cancer in the vicinity of the perianal inflammatory changes of HS

specialist late in the disease course, and even if they appear early on, recurrence occurs despite proper treatment [11]. In our study, the recurrence rate was 21.6%. From the data of Ritz et al. [18], the rate of recurrence of HS after surgical treatment was 45%, broken down to 100% after drainage of abscesses, 42.8% after a partial excision and 25% after a radical excision of inflammatory changes. In contrast, the study of Harrison et al. [8] had 0% recurrence after perianal surgery and a high 37% rate after an inguinoperineal approach. In our patient group, 9 patients were operated on twice, 1 patient three times and 1 patient even seven times. The latter patient had very diffuse changes occupying the groin, thighs, and buttocks. Additionally, he was initially treated with radiotherapy, to no effect.

Conclusion

In conclusion, TPUS is a simple and accessible imaging method, which confirms the typical localization of changes of HS. Together with AUS, it allows for the proper differentiation of HS from an anal fistula or abscess. Coexistence of HS with an anal fistula not evident on proctological examination was found via AUS in 8.9% of cases, which was confirmed intraoperatively. AUS did not allow for differentiation between HS and a pilonidal cyst, which was possible through the clinical, intraoperative and histopathological examinations. In 3% of cases skin malignancy had developed within the course of HS.

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