

Questionnaire-based diagnosis of hidradenitis suppurativa: specificity, sensitivity and positive predictive value of specific diagnostic questions

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Summary

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Background Estimates of the prevalence of hidradenitis suppurativa (HS) range from 0.33% to 4%. Further epidemiological data are therefore needed. Because of the hidden nature of the disease, physical screening may be cumbersome and questionnaire-based screening may be more appropriate.

Objectives To establish the sensitivity (SE), specificity (SP) and positive predictive value (PPV) of simple diagnostic questions used in HS.

Methods Potential diagnostic questions regarding HS were identified and sent to 85 patients with HS and to an age- and sex-matched control group randomly selected among outpatients not being treated for HS. All respondents were recruited in the Department of Dermatology in Roskilde Hospital.

Results In total, 74 of 85 patients with HS (87%) returned the questionnaire (61 women and 13 men). Of these, 72 reported repeated outbreaks of painful nodules or boils in locations typical for HS compared with 13 patients in the control group. The SE ranged from 0.92 to 0.97, the SP from 0.82 to 0.86 and the PPV from 0.85 to 0.89. Boils appeared significantly more often in patients with HS, who also reported significantly greater suffering from their lesions.

Conclusions The high diagnostic power suggests that all the questions are potentially useful. The clear symptomatology of HS may be a key factor. It is suggested that further improvement may be achieved by adding definitions of pimples, nodules or boils to future questionnaires. Similarly, adding the possibility to indicate uncommon locations, duration and quality-of-life impairment may benefit the diagnostic power.

The descriptive epidemiology of diseases is of interest not only in order to identify risk factors but also to quantify the burden of the disease on society. Hidradenitis suppurativa (HS) is a chronic, inflammatory, recurrent, debilitating, follicular skin disease which causes considerable morbidity in patients.¹ It is painful, malodorous and it leaves spots on clothes.^{2,3} HS can involve major parts of the armpits, groins, genitals and buttocks and is hence most often hidden.^{4,5}

In earlier studies estimates of HS prevalence have shown a considerable range from 0.33% to 4%,^{6,7} with an estimate of 1% of the general population in France.⁸ It is therefore necessary to make further prevalence studies in order to obtain a better estimate of general validity. For screening purposes this is most easily done through questionnaires, but because of the easily recognizable symptomatology and restricted location of HS it may be speculated that simple questionnaires may even provide the diagnosis of this disease.

Diagnostic questions related to HS used in other prevalence studies were identified in the literature. In Norway a major health survey, 'The Tromsø Study',⁹ was established in 1974. It was initiated in an attempt to help combat cardiovascular diseases, but has gradually been expanded and repeated since its inception. The latest, sixth study was in 2007–2008 and included questions concerning a variety of skin diseases. Among these were questions regarding HS.

A French survey was established in March 2005 and the purpose was 'to evaluate the prevalence of major dermatologic disorders in the French population and any associated factors, including environmental and socio-demographic characteristics'.⁸ In the study an estimate was made of the prevalence of HS based on HS-specific questions.

Finally, a major health survey is presently being conducted in the eastern part of Denmark (Zealand region) and includes diagnostic questions about HS.

The questions describe similar clinical features, but vary somewhat, and we have therefore investigated the sensitivity (SE), specificity (SP) and positive predictive value (PPV) of the questions used in order to validate the findings of these studies.

Materials and methods

Potentially diagnostic questions regarding HS were identified and translated into Danish. A set of questionnaires was then arranged with questions from the Norwegian Tromsø general health investigation,⁹ with a question composed by the group of French investigators⁸ and with questions from the general health investigation in Denmark (Zealand region). The primary questions were:

Question 1: Do you repeatedly have outbreaks of big sore or painful nodules or boils that heal with scars in any of these locations: groins, armpits, sexual organs, anal region, under the breasts, or in folds on the stomach/around the navel? (Norway)

Question 8: During the last 12 months did you repeatedly have big painful nodules or boils located in the armpits or in the groins, a disease called hidradenitis? (France)

Question 10: Have you had outbreaks of boils during the last 6 months? (Denmark)

In addition, a range of secondary questions relating to HS was chosen. These were intended to describe the groups in greater detail, and to provide possible secondary diagnostic clues which might improve the diagnostic accuracy of the primary questions.

The questionnaires were sent to the private address of 85 patients with HS aged 19–70 years, who were registered as having regular follow ups for their HS at the outpatient clinic of the Department of Dermatology in Roskilde, Denmark. All the patients with HS were Hurley stage II or III.¹⁰ The control group comprised randomly selected outpatients in the Department of Dermatology in Roskilde, Denmark. Controls were selected by age and sex on random days of the investigation and were approached by one of the authors (S.E.) who is not a trained physician and is therefore unable to provide independent confirmation of diagnoses. They participated anonymously. The survey was conducted during July–September 2009. Questionnaire surveys do not legally require Ethics Committee approval in Denmark, but the survey was reported to the Danish Data Protection Agency.

Statistical methods

Descriptive statistics were used and binary classification tests of SE, SP and PPV were performed.

Results

The overall response rate was 148 of 159 (93%). Among the patients with HS, 85 patients were contacted and 74 patients (87%) filled in and returned the questionnaires (61 women and 13 men). The mean age was 42 years (range 19–70); 70

patients indicated a mean duration of 17.8 years with HS symptoms (median 15 years).

A control group of 74 randomly chosen patients (45 women and 29 men) in the outpatient department answered similar questions in the outpatient clinic, giving a response rate of 100%. The mean age in the control group was 43 years (range 18–85). The distribution of skin diseases in the controls is seen in Table 1.

All patients with HS reported repeated outbreaks of boils (except for one patient who no longer had HS symptoms and a patient who later in the questionnaire indicated boils at locations not mentioned), whereas there was a significantly lower occurrence of boils in the control group ($P < 0.0001$). In total, 13 patients in the control group indicated that they had had painful nodules or boils in the groins ($n = 6$), in the armpits ($n = 7$), around the sexual organs ($n = 6$), around the anal region ($n = 3$), under the breasts ($n = 2$) and in folds on the stomach/around the navel ($n = 1$). The pattern of lesions among patients with HS is shown in Table 2.

The SE, SP and PPV of the primary questions studied are given in Table 3, and the SE, SP and PPV of the secondary questions studied are given in Table 4. These questions include the healthcare-related behaviour of the patients who self-reported boils compatible with the diagnosis of HS.

When boils were reported in the armpits or around the sexual organs, boils additionally were occasionally found at other

Table 1 The distribution of various skin diseases in the control group ($n = 73$)

Skin diseases among controls	n
Eczema	22
Skin tumours	12
Psoriasis	7
Infections	7
Autoimmune diseases	6
Acne	5
Other diseases	14 ^a

^aErythema annulare, Gougerot–Cartaud syndrome, granuloma annulare, hyperhidrosis, urticaria, melasma, porphyria cutanea tarda, pruritus, ulcers, vitiligo.

Table 2 Pattern of lesions in patients with hidradenitis suppurativa (HS; $n = 72$)

	HS question 1	HS question 10
Groins	60	53
Armpits	49	37
Sexual organs	43	31
Around the anal region	23	–
Under the breasts	15	9
In folds on the stomach/around the navel	10	–
Other locations	–	26

Table 3 The sensitivity (SE), specificity (SP) and positive predictive value (PPV) of the primary questions

Primary questions	SE	SP	PPV
Question 1 Do you repeatedly have outbreaks of big sore or painful nodules or boils that heal with scars in any of these locations:			
Indirectly yes to boils (HS = 72, Controls = 13) P < 0.0001	0.97	0.82	0.85
Groins (HS = 60, Controls = 6) P < 0.0001	0.81	0.92	0.91
Armpits (HS = 49, Controls = 7) P < 0.0001	0.66	0.91	0.88
Sexual organs (HS = 43, Controls = 6) P < 0.0001	0.58	0.92	0.88
Anal region (HS = 23, Controls = 3) P < 0.0001	0.31	0.96	0.88
Under the breasts (HS = 15, Controls = 2) P = 0.001	0.20	0.97	0.88
Folds on the stomach/around the navel (HS = 10, Controls = 1) P = 0.005	0.14	0.99	0.91
Question 8 During the last 12 months did you repeatedly have big painful nodules or boils located in the armpits or in the groins, a disease called hidradenitis?			
Yes (HS = 67, Controls = 8) P < 0.0001	0.92	0.86	0.89
Question 10 Have you had outbreaks of boils during the last 6 months?			
Yes (HS = 70, Controls = 11) P < 0.0001	0.95	0.85	0.86
Groins (HS = 53, Controls = 3) P < 0.0001	0.72	0.97	0.96
Armpits (HS = 37, Controls = 4) P < 0.0001	0.50	0.95	0.90
Sexual organs (HS = 31, Controls = 2) P < 0.0001	0.42	0.97	0.94
Under the breasts (HS = 9, Controls = 2) P = 0.028	0.12	0.97	0.82
Other locations (HS = 26, Controls = 4) P < 0.0001	0.35	0.91	0.79

HS, hidradenitis suppurativa.

Table 4 Sensitivity (SE), specificity (SP) and positive predictive value (PPV) of the secondary questions

Secondary questions	SE	SP	PPV
Ever visited the doctor because of boils (P < 0.0001)	1.00	0.33	0.90
Received treatments			
Antibiotic ointment/cream (P = 0.002)	0.89	0.50	0.93
Antibiotic tablets (P = 0.307)	0.83	0.30	0.90
Surgical opening/emptying (P = 0.40)	0.72	0.60	0.93
Surgical removal of skin (P = 0.133)	0.33	0.90	0.96
Surgical laser treatment (P = 0.594)	0.03	1.00	1.00
Patient impressions of possible provokers			
Stress/psychiatric influence (P = 0.001)	0.69	0.77	0.94
Narrow/tight clothes (P = 0.049)	0.44	0.85	0.94
Menstruation (women, HS = 60, Controls = 5) (P = 0.342)	0.32	0.77	0.88
Pregnancy (women, HS = 60, Controls = 5) (P = 0.387)	0.07	0.92	0.83
Other (P = 0.088)	0.49	0.77	0.92

HS, hidradenitis suppurativa. Only respondents who indicated that they had had boils were included (HS = 72, Controls = 13).

locations as well. Figure 1 shows the PPV for combinations of affected regions.

On a numeric rating scale (0–10) the degree of suffering from pimples and boils was indicated in relation to the present moment and to the worst case. Patients with HS reported significantly greater suffering from pimples at the present moment (2.2 vs. 0.9; P = 0.005) and in the worst case (3.4 vs. 1.6; P = 0.005), and they reported an even greater suffering from boils at the present moment (5.8 vs. 0.5; P = 1.94×10^{-19}) and in the worst case (9.2 vs. 1.0; P = 4.33×10^{-35}).

Boils appeared significantly more often in patients with HS than in controls when reporting one or more boils during the past year (P ≤ 0.0001; see Table 5). The onset of boils was indicated, with a majority at the age of 13–19 years among the patients with HS while the majority among the controls was found at the age of 26–35 years (see Table 6).

Discussion

The aim of the study was to evaluate the diagnostic accuracy of simple diagnostic questions, with a view to establishing a valid questionnaire-based diagnosis of HS. The SE, SP and PPV of the primary questions are very high compared with diagnostic questions concerning, for example, occupational dermatosis.¹¹ The SE ranges from 0.92 to 0.97, the SP from 0.82 to 0.86 and the PPV from 0.85 to 0.89. The high values may be based on both the strict topical limits of HS and the clear and easily recognizable symptoms. This suggests that all the questions are valid and potentially useful, although the subtle differences in the wording appear to play a role.

The first question (no. 1) has six questions integrated in one single question giving conditions of repeatability, pain, size, healing, type and location, but with no time limit. The next question (no. 8) is very narrow with a specific point of focus on armpits and groins and a time limit of 12 months. The last question (no. 10) is nonconditional except for a time limit of 6 months. A comparison of these three questions reveals both strong and weak sides of each.

The Norwegian question (no. 1) gave the highest SE but the lowest PPV, which was, however, still 0.85 and may be considered very good. The question correctly identified 72/74 of the patients in the sample, suggesting that it was easily understood. The question also lists several locations where HS may occur, while there is no information about nodules or

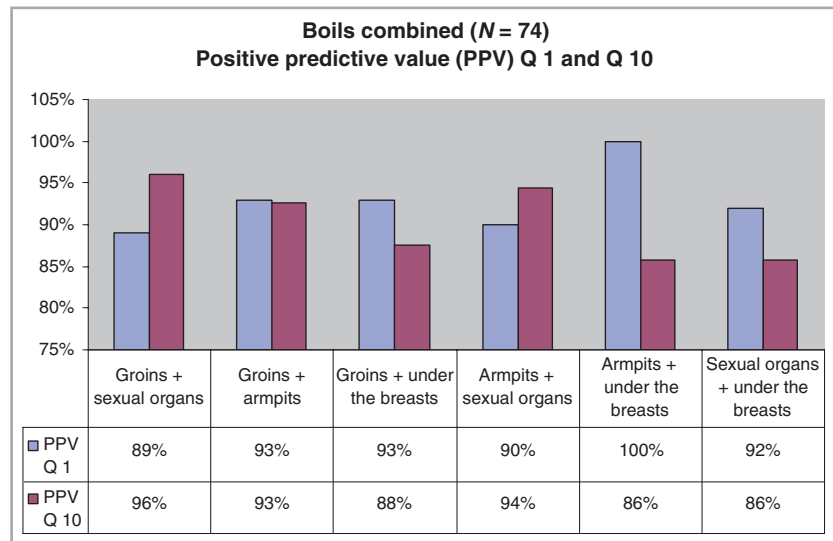


Fig 1. Diagnostic strength of combined locations of boils.

Table 5 Number of boils during the preceding year

Number of boils during the preceding year	HS	Controls	Total
0	1	62	63
1	1	6	7
2–3	9	2	11
4–6	16	1	17
> 6	47	3	50

HS, hidradenitis suppurativa.

Table 6 The age at onset of boils (years)

	0–12	13–19	20–25	26–35	36–50	Over 50
HS	10	26	18	9	8	2
Controls	0	1	4	6	2	0

HS, hidradenitis suppurativa.

boils located on other parts of the body. Nodules or boils located on other parts of the body may indicate a differential diagnosis, e.g. acne conglobata.

Another issue is the lack of time limit. One patient answered positively to having HS in the armpits, but later indicated that his HS had ceased at the age of 26–35 years. For a long period he has had no symptoms, but he is still aware of the risk of new symptoms and continues to use antiseptic soap in the hope of preventing recurrence.

The question from the French investigation (no. 8) gave the highest SP and PPV. Nevertheless, the question may be too narrow with a specific focus on armpits and groins. Only 67 of 74 patients with HS were identified correctly (six negative answers and one nonresponder). If the patients have boils on other locations they are prevented from giving a positive answer and their HS is not diagnosed, when compared with a clinical examination.

The Danish question (no. 10) had 70 of 74 positive answers, and intermediate results. It may be speculated that the shorter time limit of 6 months influenced the results. The time limit may be too narrow, leaving out milder HS symptoms. In this question it is possible to indicate boils in other areas to suggest differential diagnoses.

The PPVs of the single questions appear to be satisfactory with values being > 0.85 for all the screening questions studied. The disease is, however, multifocal and combining positive reports on lesions from different regions may therefore increase the PPV. Only two of the screening questions make this possible (questions 1 and 10). The results suggest that a further improvement of the PPV is possible, as shown in Figure 1. The degree of improvement, however, depends on which of the two questions is used.

Among the secondary questions the higher self-reported suffering associated with the lesions in HS also supports the ability of the questions to discriminate between the two groups correctly.^{2,3} Additionally, covering all questions, it may be of importance to focus on the visibility and readability of the questions. The questions in the present survey were placed in rows and columns as similar to the original as possible. An increased visibility and readability of the questionnaire might improve responses further.

In the control group 8–13 participants indicated boils depending on how the question was posed. As the control group members were chosen randomly and anonymously a possible HS diagnosis in these patients can therefore not be confirmed, but they were unlikely to have had HS as they all were under dermatological treatment. The figure of 8–13 of 74 controls is, furthermore, significantly larger than suggested in earlier studies, where HS was serendipitously found in approximately 1 of 1000 outpatients.¹²

It may be speculated that pimples and boils are mixed up by patients who are not familiar with boils. Twenty-six participants from the control group answered that they had had boils during their life. Eight had seen a doctor because of

boils, and one did not answer, but indicated having had treatment on prescription for boils. Seven participants indicated that they had no longer symptoms of boils. Based on the varying answers some of the controls may have had difficulties with the discrepancy between pimples and boils, which may affect population studies using these questions.

The chosen primary questions describe similar clinical features and they vary, but our investigation shows an SE, SP and PPV of the questions that underlines the usefulness of the questions used. Areas of possible improvement were, however, also identified. In future questionnaires a description of the difference between pimples, nodules and boils is needed to reduce false-positive answers. Next, a positive or negative response to having nodules or boils is essential. The possibility of indicating a variety of locations for the boils including uncommon locations may further benefit diagnostic power. An indication of duration, using multiple choices, may also strengthen the validity, as would an indication of the degree of suffering on a numeric rating scale.

What's already known about this topic?

- Previous estimates of the prevalence of hidradenitis suppurativa range between 0.33% and 4%.
- The disease has easily recognizable symptoms and signs.
- Questionnaires have been used to make the diagnosis without reporting on the validity of the questions.

What does this study add?

- Data on the sensitivity, specificity and positive predictive value of diagnostic questions concerning hidradenitis suppurativa.
- A point of reference for the diagnostic validity of other diagnostic questionnaires in dermatology.
- A tool for future epidemiological studies of hidradenitis suppurativa.

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