Hernia (2023) 27:15–20
https://doi.org/10.1007/s10029-022-02721-8

REVIEW

The role of a simple questionnaire predicting treatment success in children with ACNES

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Received: 14 August 2022 / Accepted: 13 November 2022 / Published online: 8 December 2022

Abstract

Background Some children with chronic abdominal wall pain or groin pain do not have an inguinal hernia but suffer from anterior cutaneous nerve entrapment syndrome (ACNES). Diagnosing ACNES is challenging, especially in children as a diagnostic gold standard is lacking. A paediatric questionnaire containing 17 simple items was earlier found to discriminate between abdominal pain due or ACNES or IBS. Scores range from 0 points (ACNES very unlikely) to 17 points (ACNES very likely). The present study investigates whether this 17-item questionnaire predicted treatment success in children receiving therapy for ACNES.

Methods Children < 18 years who presented in a single institute between February 2016 and October 2021 with symptoms and signs suggestive of ACNES completed the questionnaire before intake and treatment. Treatment success after 6–8 weeks was defined as self-reported 'pain-free' (group 1), '> 50% less pain' (group 2) and '< 50% less pain' (group 3). Group differences regarding sex, age, BMI, symptoms duration and questionnaire scores were analysed.

Results Data of 145 children (female 78%, mean age 14.7 ± 2.3 years, mean BMI 21.1 ± 3.9) were analysed. All children received a diagnostic trigger point injection using an anaesthetic agent, and 75.5% underwent subsequent surgery for untractable pain. The three groups were comparable regarding sex distribution, age, BMI and symptoms duration. In addition, questionnaire scores were not different (group 1: $n = 89$, mean score 13.4 ± 2.7, group 2: $n = 24$, 13.4 ± 2.3 and group 3: $n = 32$, 13.0 ± 2.7, $p > 0.05$).

Conclusions Treatment success was attained in 78% of children undergoing surgery for ACNES. A simple questionnaire scoring items associated with abdominal pain did not predict treatment success.

Keywords Paediatrics · Surgery · Childhood ACNES · Abdominal wall pain

Introduction

Chronic abdominal pain or groin pain is common in adult and paediatric populations. Groin discomfort is often related to the presence of an abdominal wall hernia or inguinal hernia. However, sometimes hernias are not visible during inspection or ultrasound, and alternative diagnoses must be considered. A portion of these adults and children may suffer from anterior cutaneous nerve entrapment syndrome (ACNES).

ACNES is characterised by severe abdominal pain due to entrapment of cutaneous terminal branches of intercostal nerves (7th–12th) penetrating the rectus abdominis muscle [1, 2]. Children (and adults) with ACNES present with pain that is localised within the lateral boundaries of the rectus abdominis muscle. Children are often not able to go to school due to the severity of the pain. When asked where the pain is located, they often point towards a small area in the abdominal region. A swab test may identify altered skin sensation at the point of pain reflecting its neuropathic character. Pinching the skin overlying the painful spot is often disproportionally painful. A positive Carnett's test supports the diagnosis. This test is executed as follows. The investigator localizes the point of maximal pain with his index finger.

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The patient is then asked to lift the head or upper torso or the legs, while the palpating index finger remains on the painful spot. If the pain aggravates with this lifting or is at least equal, its origin is probably located in the abdominal wall (Carnett positive). In contrast, pain of visceral origin is attenuated during this test.

The incidence of ACNES in children is unknown. ACNES is considered as a diagnosis 'per exclusionem' as the absence of abnormalities in blood analysis or ultrasonography/ computed tomography is essential for the diagnosis [3]. Childhood ACNES appears to be gender related. A recent study shows that 77% of children suffering from ACNES are females [1]. Any factors predisposing to ACNES are unknown. In addition, the vast majority of children were never operated before [3].

If pain levels due to ACNES are not acceptable, treatment starts with local abdominal wall infiltrations using anaesthetic agents. An approach of one or multiple trigger point injections is long-term effective in one-third of a childhood population. For those with refractory symptoms, surgery can be considered as a neurectomy of the affected terminal branches may provide extended pain relief [4–6]. A 78% success rate of this procedure was reported in children [7].

The diagnosis ACNES may be illusive. Interestingly, a simple 18-item questionnaire aided in distinguishing adult ACNES patients from a population diagnosed with irritable bowel syndrome [8]. An adapted 17-item questionnaire was validated in children [9]. It was found that an 11-points cut-off score together with a positive physical examination and a positive rectus sheath block fitted the diagnosis ACNES. Nevertheless, 20–30% of the neurectomies are still unsuccessful. Therefore, it is interesting to study whether scores of this questionnaire may predict outcome. The objective of the present study was to determine whether questionnaire scores differed between successfully and unsuccessfully treated children. If so, these findings may contribute to the counselling process prior to initiating surgery for ACNES.

Materials and methods

General information

This childhood ACNES study is a case series of prospectively monitored data analysed between August 2021 and December 2021 at Solvimáx in Maxima Medical Centre (MMC location Eindhoven). MMC is distinguished by offering a number of specialties comparable to an academic hospital profile. Solvimáx is a 'centre of expertise' within MMC, and this centre specializes in abdominal wall pain and groin pain.

In line with the European General Data Protection Regulation, a previously approved Medical Ethical Review Commission (METC) application within the MMC was used for this study. Also, informed consent was obtained in all patients.

Outpatient evaluation and follow up

Each outpatient visit at Solvimáx is proceeded by completing an intake form containing a set of questionnaires related to abdominal wall pain. Under the tuition of the parents, the child fills in these questionnaires at home. Based on an evaluation of this intake form by one of Solvimáx' specialists, children with suspected ACNES are invited for an outpatient visit. History and physical examination are conducted during this intake determining whether the suspected ACNES can be confirmed [2, 3]. Once data obtained during history taking and physical examination suggests the presence of ACNES, children received a diagnostic trigger point injection (TPI) using 5 ml of 2% lidocaine. In some children, this diagnostic intervention is therapeutic as pain relief is sufficient in the long term. However, when pain relief following 1–3 injections is repeatedly of short duration, surgery is considered as a second step in the treatment programme [4].

It is standard procedure in Solvimáx to have a telephone contact approximately 6–8 weeks after injection therapy or surgery to evaluate short term outcome. Primary outcome measure following treatment determined during this telephone contact was categorized in 3 levels as 'pain-free', '0–50% residual pain', or '50–100% residual pain'.

Population and design

Data of all children under the age of 19 at time of the Solvimáx intake between February 2016 and October 2021 who were diagnosed with ACNES and who received therapy in our institution were included in the study database. Excluded were children who did not complete the questionnaire, whose questionnaires were missing two or more answers, or children with missing follow up. Electronic files were checked in November 2021, and data on sex, age, BMI, previous surgery or injection therapy, duration of pain and other pertaining details were stored in the database allowing statistical analysis.

Questionnaire

In 2012, an 18-item questionnaire was found to distinguish adult patients who had chronic abdominal wall pain (in particular ACNES) from a population diagnosed with IBS [8]. This questionnaire was also introduced for children but was later on adapted to a 17-item list as one question ('my pain is the biggest problem') appeared irrelevant for the paediatric population (Table 1). Ten questions are ACNES related and seven questions are IBS related. The

Table 1 Childhood ACNES questionnaire containing 17 items related to the abdominal pain

1. My stomach feels bloated or I fart
2. I feel the pain at different places all over my belly
3. Lying on the painful side makes the pain worse
4. My poo has a strange texture (hard, soft, hard little pieces, watery, sloppy)
5. I feel like the pain is just underneath my skin
6. The pain feels sharp (like being stabbed with a knife)
7. The pain feels like it is deep inside my belly
8. I feel like I need to poo, but then I do not actually poo
9. Coughing, sneezing or pushing makes the pain worse
10. My pain is always in the same place or places
11. If you would draw a line from the top to the bottom of your belly through your tummy button, the pain is just to the left or right of that line
12. If my poo changes it can change my pain
13. My pain gets worse if I am doing things like walking, sitting, playing, cycling or bending over
14. The skin around the painful place feels different, numb or strange
15. Stress makes my pain worse
16. I can point with my finger to the most painful place
17. If I press on the painful place, it makes the pain worse

answer to each question yields either 1 point or no point. The score is obtained by adding up all points of affirmative ACNES questions (= 1) and negative IBS questions (= 1). Total score ranges from 0 points (IBS very likely) – 17 points (very likely) With a cut-off point of 11 in this adapted 17 item questionnaire, a 0.86 sensitivity combined with a 0.89 specificity was attained [9].

Statistics

Statistical analyses were performed by using SPSS. Children were categorized in three groups (i.e. pain-free, 0–50% residual pain and 50–100% residual pain), based on the primary outcome. Differences between groups regarding patient characteristics were statistically analysed using independent sample t-tests and Chi-square tests. The mean questionnaire scores as well as the individual questions of the questionnaire, were also compared between groups using Chi square tests.

Multivariate multinominal regression analyses were performed to test a potential association between the questionnaire score as well as scores of individual questions and the primary outcome allowing for correction for possible confounders. Patients' characteristics with p values ≤ 0.20 related to the outcome in univariate analyses, were included as possible confounders. One multiple regression analysis included the mean questionnaire score as independent variable and a second regression analysis was done with individual questions as independent variables. For this latter analysis, backward selection was performed to construct the final model. p values ≤ 0.05 were considered significant.

Results

Population characteristics

Between February 2016 and October 2021, 166 children were evaluated for possible ACNES at the Solvimáx department. As some questionnaires were missing ($n = 15$) or incomplete ($n = 4$) whereas 2 children did not undergo treatment, a total of 145 children qualified for the present analysis.

The majority of the children were female (78%) with a mean age of 14.7 ± 2.3 years and a mean BMI of 21.1 ± 3.9. On average, they had suffered from pain for 22.5 ± 33.1 months before they had an operation (Table 2). A total of 9.7% had already undergone a neurectomy in the referring hospital prior to treatment at Solvimáx.

Outcome after injection therapy for ACNES

All children received one ($n = 15$) or multiple abdominal wall infiltrations ($n = 130$, 2–6 infiltrations). One in four children ($n = 36$, 25.5%) considered themselves as 'pain free' after this injection regimen. The majority of children with persistent pain after injections chose to undergo a surgical neurectomy although a small group ($n = 21$, 13.5%) declined surgery. This group opted for an ongoing conservative treatment regimen such as TENS, psychotherapy or physical therapy.

Outcome after operative therapy for ACNES

The follow-up index was 100%. At the telephone or outpatient control, 61.4% of the children ($n = 89$) reported total

Table 2 Characteristics of 3 groups of children with different outcome after ACNES treatment

Variable	Total sample ($n=145$)		Group 1 pain Free ($n=89$)		Group 2 0–50% pain ($n=24$)		Group 3 50–100% pain ($n=32$)		p-value
Gender (fem, %)	113	77.9	66	74.2	20	83.3	27	84.4	0.38
Age	14.7	2.3	14.5	2.5	14.4	2.7	15.6	1.5	0.07
BMI	21.1	2.8	20.7	3.5	20.9	4.4	22.2	4.6	0.08
Duration	22.5	33.2	18.6	28.5	32.0	41.8	26.1	37.3	0.17
Questionnaire score	13.3	2.6	13.4	2.7	13.4	2.3	13.0	2.7	0.81

Age, BMI, Duration (months) and Questionnaire score: mean ± SD.

pain absence and return to daily routines. Another 16.6% ($n=24$) had substantial pain relief (> 50%) as they mostly experienced pain during physical exercise. In contrast, 22.1% ($n=32$) reported no or just a minor improvement (< 50% pain relief). Gender and previous surgery elsewhere were not different between groups. Age and BMI were almost significant ($p=0.07$-0.08) with the oldest children and the highest BMI in the unsuccessful group (Table 2).

Questionnaire scores

Mean score was 13.3 ± 2.6 and not different among groups (Group 1, $n=89$: 13.4 ± 2.7; Group 2, $n=24$: 13.4 ± 2.3; Group 3, $n=32$: 13.0 ± 2.7, NS) (Table 2). A separate analysis per question revealed that Question 1, 7, 12, 14 and 16 were distinctive in relation to outcome ($p<0.20$). They were subsequently used for the multivariate regression analysis (Table 3).

Multivariate regression analysis

Multivariate multinominal regression analysis showed no significant relationship between questionnaire scores and treatment outcome (group 2 vs group 1: OR 1.04, 95%CI 0.86–1.26; group 3 vs group 1: OR 0.97, 95%CI 0.83–1.14). Interestingly, the second regression analysis showed that scores of questions 12 and 16 differed significantly between outcome groups 3 and 1 after correction for confounders (Question 12: OR 11.20, 95%CI 1.23–102.43; Question 16: OR 0.25, 95%CI 0.07–0.88). For both regression analyses, patient characteristics (gender, BMI and duration of the pain) demonstrated no significant relation with outcome except for a minor difference in duration of pain between group 2 and 1 (OR 1.01, 95%CI 1.00–1.03, $p=0.44$) in the first analysis.

Discussion

The vast majority of research on ACNES hitherto focused on optimizing the diagnostic process and reporting treatment efficacy. To our knowledge, this is the first study aimed at determining whether patient characteristics and properties of a simple intake questionnaire can predict treatment outcome. The database covered a period between 2016 and 2021 and considered data in ACNES children that were obtained from a prospectively completed electronical patient file and a validated questionnaire. Treatment success was categorized in three groups as patient reported absence of pain, a more than 50% pain reduction, and less than 50% pain reduction two months after treatment. The results demonstrate that 4 in 5 children who were diagnosed with ACNES benefitted from a regimen of abdominal wall infiltrations or surgery. Failures tended to be older and heavier ($p=0.07$–0.08). Overall questionnaire scores were high (mean 13.3 on a 1–17-point scale) but not different in the 3 groups of children. However, answers to two of the 17 individual questions (If my poo changes it can change my pain; I can point the pain with

Table 3 Scores of individual questions in relation to success following ACNES treatment

Question	Group 1	Group 2	Group 3	p value
Q1	76	71	59	0.19
Q2	81	79	78	0.94
Q3	56	63	50	0.65
Q4	79	79	75	0.90
Q5	46	54	50	0.76
Q6	72	71	78	0.76
Q7	75	83	63	0.19
Q8	84	96	91	0.26
Q9	63	58	63	0.92
Q10	97	92	97	0.53
Q11	72	67	78	0.63
Q12	83	88	97	0.14
Q13	83	71	88	0.25
Q14	48	63	31	0.06
Q15	73	75	69	0.86
Q16	91	83	75	0.07
Q17	92	92	94	0.95

Numbers represent percentage of patients with affirmative answer to the question

one finger') were different among groups. These findings may potentially be used in the counselling process prior to deciding on invasive treatments such as surgery.

The diagnosis ACNES is notoriously difficult to establish, in adults but even more so in children. The role of imaging such as ultrasound is not diagnostic but to exclude alternative diagnoses causing pain such as inguinal hernia. As imaging is of no value in ACNES, doctors heavily rely on physical examination for the diagnosis. A simple 17-item symptom questionnaire was earlier validated in children [9]. It was found that an 11-points cut-off score together with a positive physical examination and a positive rectus sheath block fitted the diagnosis ACNES. Nevertheless, one in 4 operations for ACNES is still unsuccessful. Considering these failure rates, there is an ongoing need for exploring the merits of alternative diagnostic approaches such as a questionnaire that may support the diagnosis ACNES in children.

The current study found that the mean questionnaire score in 145 children who were referred for analysis of chronic abdominal pain possibly due to ACNES was 13.3 on a 1–17 scale. An earlier study in children who were diagnosed with ACNES found an 11-point threshold value for the diagnosis ACNES [3]. Such an overall high score suggests that the questionnaire plays an important role in the diagnostic process of ACNES. It should be realised, however, that the population of the present study is biased as referring paediatricians were already considering the diagnosis ACNES, and some children had even undergone a neurectomy for presumed ACNES. However, a large study aimed at determining screening properties of this 17-item questionnaire study in a select population with abdominal children is not performed yet.

One of the aims of the current study was to determine if a 17-item questionnaire predicted outcome after an intervention for childhood ACNES. However, the analysis indicated that scores were not different among the three groups ($P < 0.83$). By focussing on questions separately there are some interesting findings. For instance, responses to Q12 and Q16 significantly differ among groups. Q12 ('If my poo changes it can change my pain ') is an IBS-related question. A greater proportion of children whose treatment later on failed responded with the answer yes compared to the children reporting treatment success (failure: 96.9% vs 83.1%). This finding may seem to suggest that altered defecation pattern is not very characteristic of ACNES. Conversely, Q16 is an ACNES related question ('I can point the pain with one finger'). A total of 91% children reporting success responded with a yes to this question compared to 75% in the group with failures. This finding may possibly indicate that the localized pain is highly characteristic of childhood ACNES.

A strength of this research is the time of completion of the questionnaire. These were filled in at home, even before children were evaluated at our institution. As a consequence, answers are not influenced by medical specialists and scores were not affected by the outcome of the intervention. Processing of the data was blinded. Data accrual was prospective. The relative restricted number of children is a limitation although the volume is the largest to date.

In conclusion, children who are referred for possible ACNES have an overall high score on a validated ACNES symptom questionnaire. Four in five children who were treated for ACNES report a successful outcome after 2 months. Individual questionnaire scores do not predict treatment success.

Declarations

Conflict of interest All authors declare that they have no conflict of interest.

Ethical approval In line with the European General Data Protection Regulation, a previously approved Medical Ethical Review Commission (METC) application within the MMC was used for this study.

Human and animal rights All procedures performed in this studie involving human participants were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standartds.

Informed consent Informed consent was obtained in all patients.

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