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ORIGINAL ARTICLE

ACNES syndrome, exploring the abdominal wall as source of abdominal pain

Síndrome de ACNES, explorando la pared abdominal como noxa del dolor abdominal

Clara García R.[a], Gonzalo Botija A.[a], Aránzazu Recio L.[a], Consuelo Nieto I.[b], Alfonso Barrio M.[a]

[a]Departamento de Gastroenterología, Hepatología y Nutrición Pediátrica, Hospital Universitario Fundación Alcorcón. Madrid. España.
[b]Unidad del Dolor, Hospital Universitario Fundación Alcorcón. Madrid. España.

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What do we know about the subject matter of this study?

ACNES (Anterior Cutaneous Nerve Entrapment Syndrome) is a type of abdominal pain that originates in the abdominal wall and its diagnosis is based on a compatible clinical history and physical examination.

What does this study contribute to what is already known?

We present a descriptive study in children under 17 years of age to raise awareness of ACNES in this age group. We have described a series of 20 patients detailing their demographic and clinical characteristics that allow their diagnosis, as well as the initial treatment administered.

Abstract

Abdominal wall pain, specifically ACNES syndrome (Anterior Cutaneous Nerve Entrapment Syndrome), is part of the differential diagnosis of chronic abdominal pain. This syndrome is frequently overlooked and therefore underdiagnosed. **Objectives**: To describe the clinical and evolutionary characteristics of patients diagnosed with ACNES and to draw attention to this pathology. **Patients and Method**: A retrospective descriptive study was carried out in a reference center, between October 2016 and July 2021, in patients under 17 years of age, diagnosed with ACNES, who met at least two of four of the following findings: Carnett's sign, Pinch test, dysesthesia at the point of maximum pain, improvement after infiltration of local anesthetic, having ruled out visceral or functional abdominal pathology. Epidemiological variables, symptoms, physical examination, complementary tests, treatment, and evolution data were collected. Descriptive statistics were used. **Results:** 20 patients diagnosed with ACNES, 75% women, median age 12.85 years. The abdominal examination revealed Carnett's sign in 95%, Pinch test sign in 65%, and dysesthesia in 90% of patients. 65% reported pseudovisceral symptoms. 7 patients were overweight or obese. The most frequent location (50%) was the right iliac fossa, at T10-T11 level. One patient reported spontaneous improvement; 7 improved with oral analgesia; 9 patients were referred to the pain unit, of which 5 attended, and improved with anesthetic infiltration with bupivacaine-triamcinolone. The remaining 4 were lost to follow-up.

Keywords:
Abdominal Pain;
Anterior Cutaneous Nerve Entrapment Syndrome;
Abdominal Wall Pain;
Chronic Abdominal Pain;
Carnett's Sign

Correspondence:
Clara García R.
claragr2391@gmail.com

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Luisa Schonhaut Berman

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Conclusion: ACNES should be considered in patients with chronic pain. A combination of typical findings in medical history and physical examination allows its diagnosis, therefore, avoiding unnecessary complementary tests. A step-up treatment strategy should be applied, beginning with oral analgesia, followed by anesthetic infiltration, and, finally, anterior neurectomy.

Introduction

Chronic abdominal pain (CAP) presents a high prevalence (10-19%) in Primary Care and Specialized Care[1,2]. According to its etiology, it is classified as visceral when it comes from the intra-abdominal organs; as parietal when it occurs in the abdominal wall; and functional, defined by the ROMA IV criteria and based on the gut-brain axis alteration[3-5].

One type of parietal CAP is ACNES (Anterior Cutaneous Nerve Entrapment Syndrome), first described in 1926 by J. Carnett[6]. Each anterior cutaneous nerve passes through a fibrous neurovascular channel located near the lateral border of the anterior rectus abdominis muscle to the skin. Pain is caused by compression and/or traction of the nerves or due to fibrosis[3,7-9]. A recent histological study on ACNES ruled out as a possible cause an infection or inflammation[10].

In ACNES, pain is located between the external and middle third of the anterior rectus abdominis muscle, most frequently at the level of the Th10 and Th11 dermatomes, with 86% classified as unilateral and 13-14% as bilateral. The most affected quadrant is the right lower quadrant (55-75%), which implies ruling out acute appendicitis by performing complementary explorations and even hospital admissions[5,9,11-12].

Clinically, it is characterized by selective pain, localized in a small area (the size of the physician's fingertip). It worsens with play, cough, exercise, and postural changes[7,9,13]. In most patients, its onset is gradual, but according to one study, 50% of patients have an abrupt and lancinating onset[7]. A study of 139 patients classifies it as severe on the Visual Analog Scale (VAS)[9].

Regarding the findings on the physical examination, with the patient in the supine position, the area of maximum pain will be located with the tip of the finger between the middle third and external third of the anterior rectus abdominis muscle, the area where the anterior cutaneous nerve branch originates. Once the pain is located, this point is compressed and, without releasing, the patient is asked to flex the abdominal muscles. If this maneuver significantly increases the pain, it is considered as a positive Carnett's sign (Figure 1). This maneuver differentiates it from pain of visceral origin since, in this case, the pain is relieved.

Similarly, pinching and lifting the panniculus adiposus overlying the point of maximum pain increases pain disproportionately compared with the healthy contralateral side, since the pinched nerve does not slide through the fibroadipose tunnel through which the muscle passes; known as the positive Pinch test sign (Figure 2). Finally, compared with the contralateral side, the presence of altered skin sensitivity (dysesthesias) to touch and temperature of the affected area using an alcohol-soaked cotton swab strongly supports the diagnosis[3,5,6,9,14-15].

Pain is often associated with 'pseudo-visceral' symptoms due to alteration of the autonomic nervous system including nausea, abdominal distention, loss of appetite, weight loss, or altered bowel movement[11,16-17].

As in the evaluation of any abdominal pain, alarm symptoms suggestive of organic pathology should be ruled out, such as fever, unintentional weight loss, dysphagia, persistent vomiting, sudden change in bowel movement, hematemesis, melena, hematochezia, and family history of cancer or gastrointestinal disease (such as inflammatory bowel disease or celiac disease)[13].

A validated 17-item pediatric questionnaire with a sensitivity of 85% and specificity of 89% has recently been published, which indicated irritable bowel syndrome as one of the most frequent differential diagnoses[18]. Table 1 shows other differential diagnoses[7,19-23].

Treatment should be sequential based on the severity and duration of symptoms. Initially, treatment may start with analgesics and lifestyle modifications. If there is no improvement, the treatment of choice is infiltration of local anesthetic at the point of maximum pain[7,9,24] and, if pain persists after repeated infiltrations, surgical treatment by anterior neurectomy may be considered[7,9].

Recent reviews of CAP have highlighted the lack of knowledge of abdominal pain of parietal origin, focusing exclusively on visceral and functional abdominal pain, leading in some cases to an excess of unnecessary or invasive complementary tests, as well as delayed and misdiagnosis[3,25,26].

The objectives of this study are to describe the clinical and evolutionary characteristics of patients diagnosed with ACNES and to draw attention to this pathology, which is poorly known.

Patients and Method

Retrospective descriptive study of patients aged under 17 years selected from the Pediatric Gastroenterology unit and hospitalizations between October 2016 and July 2021, of the *Hospital Universitario Fundación Alcorcón*, Community of Madrid (Spain). This is a second-level hospital of the public health system, for a population of approximately 171,098 inhabitants, out of which 29,000 are under 17 years of age. The hospital has all pediatric subspecialties, except Pediatric Surgery.

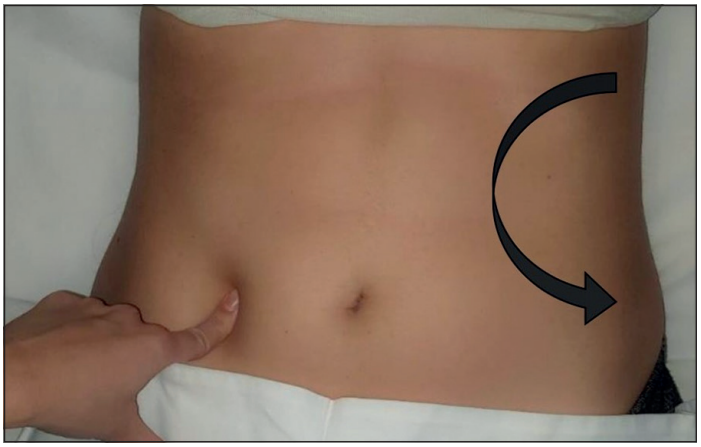

Figure 1. Carnett's sign: the compression of the point of maximum pain and simultaneously the contraction of the abdominal muscles ("look at your navel"), produces worsening of the pain.

Figure 2. Pinch test: the impingement and elevation of the overlying adipose panicle to the point of maximum pain, produces a disproportionate increase in pain compared to the healthy contralateral side.

Patients were identified through electronic medical records. All diagnoses that included the word "ACNES" in patients under 17 years of age, whether they came from the gastroenterology unit or the hospital discharge report, were used for the search. The diagnosis was made by the hospital's pediatric gastroenterologists.

Patients were selected if they presented a clinical history and physical examination compatible with a diagnosis of ACNES and had at least two of the following four findings: Carnett's sign, Pinch test, dysesthesia at the point of maximum pain, or improvement after infiltration of local anesthetic[12].

Depending on the intensity of the pain, the physician decides the initial treatment: 1) oral analgesia and modification of physical activity in mild symptoms or 2) infiltration with local anesthetic (bupivacaine-triamcinolone in our center) in cases with moderate-severe pain or pain that did not improve after oral analgesia after 2-4 weeks of treatment[12]. If the patient does not improve in the first 6 months of follow-up, the patient is referred to a tertiary hospital (with Pediatric Surgery) to evaluate anterior neurectomy.

Those patients who have a confirmed visceral or functional abdominal pathology as the final cause of the symptoms through anamnesis, serial physical examinations, and complementary tests were excluded. The complementary tests used were blood analysis (CBC, biochemical tests, renal, hepatic, and thyroid function, C-reactive protein), abdominal ultrasound, gastroscopy, and stool cultures[7,13].

Overweight was defined as those patients presenting a BMI between the 85-97 percentiles and obesity, percentile greater than 97, according to the Spanish Transversal Study of Growth 2010.

Statistical analysis was performed using "IBM SPSS Statistics", version 17, presenting absolute and relative frequencies for qualitative variables, and median and interquartile range in the case of quantitative variables.

Epidemiological, anthropometric, exploratory variables (localization, Carnett's sign, Pinch test, dysesthesias), and associated symptoms were analyzed. The complementary tests and treatment administered were described.

The study protocol was reviewed and approved by the Hospital Ethics Committee.

Results

During the study period, 21 patients were diagnosed with ACNES. One patient was excluded due to refusal to sign the informed consent. Of the remaining 20 patients, 75% were female, with a median age of 12.85 years (IQ 11.18-14.10). Five of the patients were

overweight (BMI p85-97) and 2 patients were obese (BMI greater than p97). 75% of the patients were born in Spain. 30% (6/20) of the patients (4 males), had a psychiatric history, including depression, ADHD, and adjustment disorder (Table 2).

Before the diagnosis, 12 patients (60%) had consult ed the emergency department due to CAP and 5 patients (25%) required admission for pain control and study. Most of the patients [80% (16/20)] were diagnosed in outpatient consultation. The median time from the onset of pain to consultation in Specialized Care was 2 months (IQ 1-4) and the time from the onset of pain to diagnosis was 2 months (IQ 2-6.25).

Physical examination showed positive Carnett's sign in 95% (19/20), Pinch test in 65% (13/20), and dysesthesia in 90% (18/20) of the patients. 65% (13/20) of the patients presented pseudo-visceral symptoms, and the most frequent were vomiting 20% (4/20) and diarrhea 40% (8/20). One patient reported weight loss. The most frequent location of pain was the right iliac fossa at the level of Th10-11 (50%), left iliac fossa (40%), and bilateral (10%) (Table 3).

The complementary tests requested before the diagnosis of ACNES were blood analysis 80% (16/20), ultrasound 70% (14/20), gastroscopy 20% (4/20), and stool cultures 20% (4/20).

Treatment with anti-inflammatory drugs and oral analgesics was prescribed in all patients, and symptoms subsided in 7 patients (35%). One patient reported spontaneous improvement and 3 patients were lost to follow-up. Nine patients (45%) were referred to the Pain Unit for infiltration with bupivacaine-triamcinolone. One patient refused treatment and 3 others did not come to the Pain Unit. After the first infiltration, pain disappeared in 3 of the 5 patients treated. Of the remaining two, one required a second infiltration, and the other one was referred to a tertiary hospital with pediatric surgery for evaluation of anterior neurectomy due to persistent pain after 2 infiltrations. This intervention was not performed due to the disappearance of pain after a new ultrasound-guided infiltration in the tertiary hospital.

Table 1. Differential diagnosis and clinical pain entities mimicking an ACNES-like presentation[8,20-24]

Abdominal myofascial pain syndrome
Hernia (Spigelian, umbilical, epigastric)
Hematoma
Exaggerated lumbar lordosis/leg length difference
Sportsman hernia or sports groin/pubalgia
Neurofibroma/subarachnoid cysts, Schwannoma
Radiculopathy
Abdominal wall tear
Rib abnormalities
Slipping rib syndrome
Abdominal wall tumour (desmoids)
Abdominal wall endometriosis
Abnormalities of vertebral column including joints
Herniated disk
Ilioinguinal or iliohypogastric nerve entrapment
Diabetic radiculopathy
Herpes zoster
Scar tissue (appendectomy, caesarean section, laparoscopy)

Abbreviatures: ACNES: Anterior Cutaneous Nerve Entrapment Syndrome

Discussion

Abdominal pain with origin in the abdominal wall, and specifically ACNES, should be part of the differential diagnosis of chronic abdominal pain. However, at present, its systematic exploration continues to be overlooked[3,25,26], probably due to the "visceral thinking" of clinicians when evaluating the patient with abdominal pain, as Bishop reports[26]. The objective of this study is to raise awareness of this syndrome. Therefore, we have described a series of 20 patients detailing their demographic and clinical characteristics that allow the diagnosis, as well as the initial treatment administered.

Despite this lack of knowledge, the series recently described by Siawash M et al. report 13% ACNES prevalence in the pediatric population of patients initially diagnosed as functional abdominal pain[11]. Like our sample, the female sex presented a clear predisposition (75%) and a mean age around 15 years[3,5].

Risk factors have been described in adults that predispose to this situation, especially obesity, abdominal surgical scars, pregnancy, ascites, and overuse of the abdominal muscles[3,12]. However, in up to 60% of cases no precipitating risk factors are observed[27]. The prevalence of overweight and obesity in patients with ACNES (35%) is very similar to that described in the Spanish population (30%) according to a recent study in 2020[28].

The diagnosis is eminently clinical, based on the clinical history and physical examination, presence of Carnett's signs, Pincht test, and dysesthesias in 95%, 65%, and 90% of our patients, respectively, which is in line with previously published literature[5,9,12].

Pseudo-visceral symptoms were present in 65%, and the most frequent was diarrhea. The appearance of these symptoms has been postulated due to the re-

Table 2. Demographics of patients with ACNES

Variable}	Results
Sex (male:female)	1:3 (5/15, 75% mujeres)
Age (median)	12,85 años (IQ 11.18-14.10)
Spanish nationality	75%
Psychiatric history	30% (6/20)
Anthropometry:	
Body mass index (median)	19,09 (IQ 17.31-26.23)
Overweight (BMI p85-97)	5/20 (25%)
Obesity (BMI > p97)	2/20 (10%)

Abbreviatures: BMI (body mass index), ACNES: Anterior Cutaneous Nerve Entrapment Syndrome

Table 3. Pain characteristics and findings in physical examination in patients with ACNES (n = 20)

Variable	Results
Carnett's sign	95% (19/20)
Pinch test	65% (13/20)
Altered skin sensitivity (dysesthesias)	90% (18/20)
Location (dermatomes Th10-11)	
Bilateral	10% (2/20)
Unilateral	90% (18/20)
Right	50% (10/20)
Left	40% (8/20)
Pseudovisceral symptoms	65% (13/20)

ACNES: Anterior Cutaneous Nerve Entrapment Syndrome.

lationship between the affected intercostal nerves and the internal organs by splanchnic nerve chains. These symptoms are very unspecific and may delay diagnosis[3,7,11].

Our patients present a 30% prevalence of child and adolescent mental health disorders which, although minimally higher than the general Spanish population (20%)[29], we believe that they should be considered as a risk factor.

In relation to treatment, it is initially essential to inform patients and their families of the benignity of the process. In pediatrics, it should be sequential in order to that those patients with mild symptoms can improve with oral analgesia (anti-inflammatory drugs or paracetamol) and changes in daily activities, as occurred in 35% of our patients. Activities involving vigorous exercise of the abdominal muscles should be avoided, and wearing abdominal binders, local heat, massage, and rehabilitation are useful[13].

In those patients with moderate or severe symptoms (pain interferes with daily activities) or no improvement with oral analgesia, infiltration of local anesthetic at the point of maximum pain is indicated with or without corticoids, guided or not by ultrasound, with a successful response between 38 and 87% of patients (reduction of the VAS scale greater than 50%)[7,9,24]. One study ruled out the placebo effect by demonstrating no improvement with saline infiltration[30].

Ultrasound-guided infiltration may be more expensive, however, offers greater accuracy in infiltration and less risk of injecting into the peritoneal cavity[13].

The association of local anesthetic and corticoids (mainly lidocaine-betamethasone) is probably more effective than the use of anesthetics alone, although there is no consensus in this regard[31].

In most studies performed in the adult population, diagnostic confirmation is made by clinical improvement after infiltration of local anesthetic in the affected area[7,9,12]; however, pediatric studies do not require it for definitive diagnosis[13].

45% of our patients were referred to the Pain Unit, and of these, all those who accepted treatment with infiltration showed improvement or disappearance of pain. In those patients with severe pain who do not respond to repeated infiltrations for more than 6 months, anterior neurectomy may be indicated with a short-term success rate of 78-85% and few side effects[7,9,32-33].

Recently, pulsed radiofrequency therapy has proven to be an effective and minimally invasive alternative to be considered in patients in whom conservative treatment was unsuccessful before neurectomy. Although the latter is more effective, it has a greater number of potential complications (hematoma, infection) and the need for general anesthesia[34].

Our study has limitations, such as the limited number of patients included in the study, the inter-observer variability, the lack of adherence to treatment with nerve infiltration because it is an invasive technique, and the lack of long-term follow-up of patients to determine possible relapses.

ACNES should be considered in children with chronic abdominal pain, mainly adolescent females, with compatible clinical history and characteristic physical examination. The diagnosis is clinical, through positive Carnett signs, Pinch test, and dysesthesias in the area of maximum pain, avoiding unnecessary complementary tests.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed ac-

cording to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

Financial Disclosure

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