

# Endoscopy-induced anterior cutaneous nerve entrapment syndrome: a case series



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## ABSTRACT

**Background and study aims** Anterior cutaneous nerve entrapment syndrome (ACNES) is a common but frequently overlooked disorder. Here we report on a series of patients with ACNES following endoscopy.

**Patients and methods** This case series included consecutive patients with localized abdominal pain following an endoscopic procedure that was consistent with ACNES who presented to the author's Gastroenterology Outpatient Clinic from February 2019 to February 2021.

**Results** Six patients presented with complaints compatible with ACNES. All of them were successfully managed with local injection therapy (n=5) or pulsed radiofrequency (PRF) (n=1).

**Conclusions** It appears that ACNES can be induced by endoscopy. Early recognition is important to avoid unnecessary diagnostics and delayed pain relief in patients. Most patients can be managed with local injection therapy.

## Introduction

Anterior cutaneous nerve entrapment syndrome (ACNES) is a common, but often overlooked syndrome first described by Carnett in 1926 [1]. It is characterized by a more or less continuous, sometimes waxing and waning, abdominal pain that is limited to a small localized area (<2 cm<sup>2</sup>), mostly at the lateral margins of the rectus abdominis muscle.

It is postulated that a small, anterior, cutaneous branch of an intercostal nerve while passing through the abdominal wall via a neurovascular channel may undergo pushing or pulling forces due to a triggering factor. This may then lead to a vicious cycle of muscle spasms, altered dimensions of the neurovascular channel, nerve kinking, swelling, and progressive entrapment [2]. The site of maximum pain (SMP) is found in the dermatome of the corresponding intercostal nerve (Th7–12) [2]. Triggers of the syndrome are presumed to be previous abdominal (bariatric) surgery, pregnancy, abdominal sports- or work-related ac-

tivities, trauma and possibly infections [2,3]. In most cases, though, there is no clear-cut culprit [2].

ACNES is more common in women (1:3); it can occur at all ages, with a mean age of 47 years and a peak in the 20s [4]. It has been reported in Western as well as in Eastern populations [5–7].

In the Netherlands 1 in 50 patients presenting to the Emergency Room with acute abdominal pain were found to have ACNES [8]; the authors also calculated an incidence rate of 57/100,000 in the general population [8].

Several findings are helpful in establishing the diagnosis: a positive Carnett's test (i.e. non-relenting or increasing pain during palpation of the trigger point with one finger, while tightening the abdominal muscles); the pinch-test (i.e. a different sensation – most often hypersensitivity – while pinching the skin at the painful site compared to the contralateral site); and finally, the demonstration of hyperesthesia or hypoesthesia at the SMP.

A local injection with anesthetics (with or without corticosteroids) just subfascial to the ventral anterior rectus sheath may ameliorate the pain and help confirm the diagnosis. Other treatment options are pulsed radiofrequency (PRF) at the trigger point, and ultimately, a neurectomy of the affected cutaneous nerve.

Here a series of cases with ACNES triggered by endoscopy is presented.

## Patients and methods

After encountering two patients with ACNES following endoscopy (Patients A and B), all consecutive patients referred to the author's outpatient clinic with a similar problem were registered in a database from February 2019 to February 2021. There was no predefined study protocol. Patients were treated according to local protocols. All patients gave their consent to publication of their anonymized data.

The Albert Schweitzer Hospital is a regional teaching hospital (565 beds) providing care for a catchment area with 300,000 inhabitants. In this period, 8946 colonoscopies, of which 1493 were in context of the Dutch Colorectal Carcinoma Screening Program (DCRCSP), and 7828 upper endoscopic procedures were performed, by nine gastroenterologists, one internist, three nurse endoscopists, and three fellows.

The author, as part of this group of coworkers, specializes in abdominal wall pain such as ACNES. All patients visiting our department who are suspected of having this disorder, therefore, are referred to him.

At our department, all patients are interviewed before endoscopy by a gastroenterologist (or trainee) or a specialized nurse, who inquires about any preexisting complaints, including abdominal pain.

Specific maneuvers (e.g. change of patient position or manual pressure on the abdomen) during endoscopy are not routinely recorded. All procedures were performed using CO<sub>2</sub> insufflation.

## Results

Eleven patients were registered with newly developed localized abdominal pain compatible with ACNES after endoscopy. Patients with a history of abdominal pain before endoscopy (n = 4) or a likely other cause (n = 1, severe constipation after endoscopy) were excluded from this report to avoid confusion with other causes of pain. Here we describe the six patients with a clear diagnosis fitting ACNES. For all but Patient F, it concerned their first endoscopic examination. No hemostatic powder was used in any of the cases.

Data regarding patient and endoscopy characteristics, treatment, and outcome are summarized in ► **Table 1**.

### Patient A

A 62-year-old man underwent a colonoscopy in the context of the DCRCSP. A 10-mm polyp was removed from the rectosigmoid by hot snare polypectomy. The Gloucester Comfort Score (GCS) was 2 on a scale of 5.

Immediately after the endoscopy, the patient developed continuous pain in his right lower abdomen. The pain sometimes radiated to his scrotum or upper abdomen. The patient had no fever or other discomfort.

At presentation 6 days later, abdominal examination showed a localized SMP at the level of dermatome Th12. The Carnett's test was positive with a negative pinch test. A single injection with 10 mL lidocaine 1% resolved the pain. Two years later, the patient is still free of symptoms.

► **Table 1** Patient and endoscopy characteristics, treatments, and outcomes.

Patient	Procedure	Sex	Age (yr)	Premedication	Gloucester Comfort Scale	Procedure-time (minutes)	Location dermatome/side	Time of onset of pain after endoscopy	Treatment	Result
A	Colonoscopy + hot snare polypectomy	Male	62	M/F	2	35	Th 12/right	immediately	L 1 ×	Remission
B	Duodenoscopy	Female	68	None	NA	NA	Th 9/midline	immediately	L 3 ×	Remission
C	Colonoscopy + EMR	Male	70	None	NA	105	Th10/left	immediately	L 2 ×	Remission
D	Colonoscopy	Male	64	M/F	1	25	Th12/right	3 days	L 1 ×	Remission
E	Colonoscopy	Female	59	M/F	2	20	Th11/left	immediately	L 2 × L/M 1 ×	Remission
F	Duodenoscopy + PEG-J	Female	20	P	2	44	Th7/left	immediately	L 2 × L/M 1 × PRF 1 ×	Remission after PRF

M, midazolam; F, fentanyl; EMR, endoscopic mucosal resection; NA, not available; L, lidocaine; L/M, lidocaine with methylprednisolone; P, propofol; PEG-J, percutaneous endoscopic gastrostomy with jejunal extension; PRF, pulsed radiofrequency.

## Patient B

A 68-year-old woman underwent an upper endoscopy for assessment of a contained gastric perforation that had occurred 2 months earlier and which had been treated conservatively. No abnormalities were found, but immediately after the endoscopy, she experienced continuous pain in the epigastric region, which was worse when she was seated. Proton pump inhibitor therapy was started by the general practitioner without relief. For this reason, the patient was sent to our outpatient clinic 3 months later. We found a localized epigastric SMP at the level of Th9, slightly lateral to the midline. There was no amelioration of pain during the Carnett's test and a positive pinch test. The first injection with 10 mL lidocaine 1% significantly diminished the pain, and after a total of three injections over a 2-month period, the patient was free of symptoms. She patient was instructed to let us know when the pain recurred. One and a half years later, she has not returned.

## Patient C

A 70-year-old man underwent a colonoscopy because of rectal blood loss. During the procedure, a 4 × 2-cm villous adenoma with low-grade dysplasia in the descending colon was removed by endoscopic mucosal resection (EMR). Immediately after this procedure, the patient developed pain in his left abdomen. A surgeon was consulted and could not find the cause, and the patient was referred to our outpatient clinic 2 months later. The man complained of continuous pain and the feeling of a bulge at the level of Th10. He could not lie on his left side. Cycling especially worsened the pain. The Carnett's test was indifferent and the pinch test negative. Ultrasound examination and abdominal computed tomography showed no abnormalities. Four weeks later, the pain was found to be more localized, with a positive Carnett's test. A pinch test showed hyposensitivity on the affected side. An injection was given with 10 mL lidocaine 1%, and over a period of 3 weeks, the patient's pain ameliorated and finally disappeared.

One year later, the patient was referred again for the same pain. It turned out the pain had recurred soon after his last visit but had not been intense enough for a new consultation. Physical examination showed the same SMP, and a positive Carnett's test. Another injection with lidocaine 1% was given, after which the pain disappeared. Six months later, the patient was still free of pain.

## Patient D

A healthy 64-year-old man underwent a colonoscopy in the context of the DCRCSP. An ulcer was found in his cecum and biopsies showed ischemia. The terminal ileum was normal. The GCS was 1.

Ten weeks later, the patient was referred to our outpatient clinic because of continuous pain in his right lower abdomen, which he said had started 3 days after the colonoscopy. Physical examination showed a right-sided, localized SMP at the level of dermatome Th12. The Carnett's test was positive, as was the pinch test. A local injection with 10 mL lidocaine 1% resolved

the pain. Six months later, he reported that he was still free of pain, although he sometimes felt a slight itch.

## Patient E

A 59-year-old woman had a colonoscopy in the context of the DCRCSP, which had shown no abnormalities. The GCS was 2. Immediately afterward, she developed progressive, continuous pain in the left lower abdomen, for which she was referred to our outpatient clinic 3 months after the endoscopy.

On physical examination, a localized SMP was found at the level of dermatome Th11. Carnett's test was positive and a pinch test negative; there was hypoesthesia at the SMP. An abdominal ultrasound was unremarkable. An injection with 10 mL lidocaine 1% was given, without any effect. An injection with lidocaine and corticosteroids was refused by the patient.

3 years later she returned to our outpatient clinic because the pain, which had persisted all the time, had worsened during the last 3 months. She had been referred to a gynecologist, who could not explain the pain.

The patient stated that the pain got worse when she was sitting forward and when she had abdominal bloating. She was limited in her sports activities and could not sleep on her left side. The Carnett's test and the pinch test were both positive. An injection with 10 mL lidocaine 1% again had no effect. An injection 3 months later with 5 mL methylprednisolone (40 mg/mL)/lidocaine 1% significantly reduced the patient's pain and no further therapy was needed. One year, later she reported that she was free of symptoms.

## Patient F

A 20-year-old woman with refractory gastroparesis underwent a percutaneous endoscopic gastrostomy with jejunal extension (PEG-J). Four days later, she presented to the Emergency Room because of severe pain in her upper abdomen. She had no fever. The pain was found to be localized about 2 cm above and left to the introduction site, which looked completely normal. The pain worsened on movements and breathing. Ultrasound examination showed no abnormalities. The correct position of the feeding tube was confirmed with an abdominal X-ray. The patient was referred to our outpatient clinic 2 days later. A SMP was found just below the left costal margin, at the level of Th7, which showed a positive Carnett's test and pinch test. Local injection of 10 mL lidocaine 1% led to remission of the patient's pain for 5 days; a second lidocaine injection worked for 1 week. An injection of 5 mL methylprednisolone (40 mg/mL)/lidocaine 1% was given, which resulted in pain relief for 1.5 weeks. Three weeks later, a PRF was performed, which led to complete remission of pain. One year after PRF, the patient reported that she was still free of pain.

## Discussion

This case series indicates that endoscopic procedures on the upper as well as the lower gastrointestinal tract can induce ACNES. To the best of our knowledge, only one case of ACNES after an upper endoscopy has been reported [5]. This case was well-described, but hampered by the presence of gastric ulcer

as a potential cause of pain. Moreover, the initial reason for the examination had been abdominal pain. The absence of prior pain or significant abnormalities at the index endoscopy makes the current report more convincing.

Hypothetically, in line with the theory of pushing and pulling forces at the intercostal nerve branches referred to in the introduction [2], anything that (temporarily) changes the features of the abdominal wall may induce ACNES. Hence, in the case of endoscopy, abdominal distension with air insufflation may be the trigger by stretching the small nerve branches that pass through the abdominal wall. Apparently even a very time-limited offense, such as in routine upper endoscopy, can induce ACNES, as suggested by case B as well as by the one in Okamoto's report [5].

Other possible diagnoses, such as a perforation or post-coagulation syndrome, are highly unlikely in view of the long duration of pain in most cases and the absence of other discomfort, fever or alarming findings at physical examination and additional diagnostics. Moreover, the long-lasting response to locally applied therapy supports the conclusion that the pain actually concerned ACNES.

Takeda et al. reported that a positive Carnett's test is often found in patients with psychogenic pain [7]. However, this is unlikely in our cases, as none of the patients had a past history of psychiatric issues, and in general, there were no characteristics indicating psychogenic pain, as described by Takeda et al, such as long symptom duration, an extensive painful region with a poorly defined border, the absence of relieving factors, and no change in the severity of pain regardless of the site of palpation. Whether anxiety regarding endoscopy might have contributed to the development of ACNES is unclear, but one would not expect patients to be cured so easily. Besides, presumably symptoms due to anxiety would be more likely to arise before endoscopy.

With regard to Patient F, one might presume the pain to be parietal due to the PEG-J insertion. However, data argue against this, such as the very circumscribed area of pain, its location distant from the insertion point, the positive tests on physical examination, and the favorable response to therapy.

Two of the four patients undergoing a colonoscopy developed pain in the right lower abdomen. This may seem odd as the examination is mostly performed in the left lateral position. However, this is also the most common location in non-endoscopy-related ACNES for unknown reasons [2]. Furthermore, one might speculate that during endoscopy, the air rises up to the cecal region, leading to the highest pressure in the right lower abdomen. Manual pressure exerted on the patient's abdomen during colonoscopy could be considered to play a role in development of ACNES. However, this seems unlikely, as manual pressure is rarely applied to the right lower abdomen.

Half the patients in this series were middle-aged men, in contrast to the known predominance of ACNES in women and those who are younger. Apart from being a merely coincidental finding, this may be caused by selection bias: Endoscopic procedures are more often indicated in older patients. Moreover, the DCRCS starts at age 55 and the majority of patients referred for colonoscopies in this program are men.

All but one patient was free of pain after only injection therapy. The remaining patient was cured with PRF. In the literature, varying data about how many patients can be cured with injections alone have been reported, but a range of 33% to 50% seems safe to say, based on the most recent studies [4, 9]. In our series five of six patients (83%) were cured with injections alone. Injection therapy may be more effective due to the rather short existence of the nerve entrapment in this series.

The addition of corticosteroids was beneficial in Patient E. Although older data indicate that adding corticosteroids could lead to higher success rates [9], in a randomized trial comparing first-line therapy with anesthetics with or without corticosteroids, no difference was found [10]. Whether the addition of corticosteroids to the anesthetic after failure of an anesthetic alone is of value remains to be studied.

PRF, as performed in patient F, is a valuable second-line therapy. In a randomized controlled study comparing PRF vs. neurectomy in 66 patients already scheduled for neurectomy, up to 40% of patients did not need surgery after PRF [11].

## Conclusions

In conclusion, in case of persistent pain following endoscopy, one should be aware of the possibility of ACNES. This can prevent unnecessary consultations, diagnostics, and treatment delay. Local injection therapy is an easily applicable and effective first-line treatment.

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## Competing interests

The authors declare that they have no conflict of interest.

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