

[ CASE REPORT ]

## Anterior Cutaneous Nerve Entrapment Syndrome Possibly Triggered by Oral Contraceptives

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### Abstract:

We herein report a teenage girl who had been taking oral contraceptive pills for three months and complained of left lower abdominal pain that had continued for two months. A physical examination indicated anterior cutaneous nerve entrapment syndrome (ACNES), although no abnormality was found in various biochemical and imaging examinations. The pain was only transiently ameliorated by trigger-point injection, and neurectomy surgery was eventually effective. Sex steroids can be involved in the progress of local tissue edema causing ACNES. ACNES should be considered in cases of abdominal pain in patients taking oral contraceptives.

**Key words:** abdominal pain, ACNES, cutaneous nerve entrapment syndrome, oral contraceptives

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### Introduction

Anterior cutaneous nerve entrapment syndrome (ACNES) is characterized by chronic abdominal wall pain caused by strangulation of sensory nerve branches leading to the abdominal wall due to excessive abdominal pressure, ischemia, or compression of adipose tissue (1). Trigger-point injection of local anesthetics is useful not only for treatment but also for the diagnosis (2). ACNES patients may visit various clinical units or departments for consultation, such as primary care, general medicine, pediatrics, gastrointestinal surgery departments, and/or pain clinics (3). We experienced a case of ACNES triggered by oral contraceptives that was successfully treated by repeated neurectomy surgeries.

### Case Report

A 16-year-old girl was referred to our hospital because of left lower abdominal pain that had continued for 2 months.

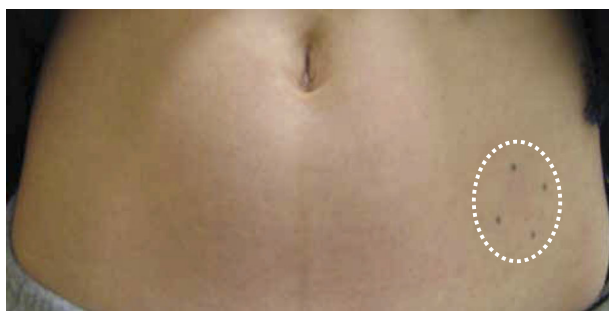
From one month before the occurrence of pain, the patient had been taking small-dose oral contraceptive pills containing ethinylestradiol and desogestrel because of severe dysmenorrhea. The patient felt a tumor-like sensation where the pain was localized. The patient had taken loxoprofen, acetaminophen and celecoxib; however, these agents were not effective for the abdominal pain. The results of blood cell counts, biochemical, immune- and endocrine-related tests, and abdominal echography were all normal, and computed tomography (CT) and magnetic resonance imaging (MRI) showed no significant abnormality. The patient was admitted to our hospital for a further examination.

Her height was 157.2 cm and her weight was 44.8 kg (BMI: 18.1 kg/cm<sup>2</sup>). She had specific tenderness in her left lower abdomen but had no rebound pain around that region (Figure). Pinching the tender part triggered severe pain, and Carnett's sign was positive, suggesting pain due to the abdominal wall. The patient also had a low-grade fever with a maximum temperature of 37.6°C and tachycardia around 120 bpm, while no change in the abdominal pain was seen.

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**Figure.** Tender region of the abdomen. The part marked in black indicates the region with tenderness (circled with white dotted line). The region was less than 2 cm<sup>2</sup>, and Carnett's sign was positive.

Considering the possibility of ACNES, 1% lidocaine was injected into the tender part of the abdominal wall, and the effect was only transiently observed. Although various analgesics, including pregabalin, tramadol-acetaminophen combination, and codeine phosphate, were administered on trial, the agents were not effective for the pain.

We eventually decided to perform percutaneous selective nerve dissection to alleviate the pain. After the surgery, her abdominal pain was remarkably improved, and the patient was able to return to daily life without taking any painkillers. However, one month after neurectomy, her pain occurred again when she attempted to use her abdominal muscles. The severity of the pain was almost the same as before, suggesting a relapse of ACNES.

During initial surgery for percutaneous nerve resection, the distal part had already been removed from the pain region penetrating the abdominal muscles, so additional laparoscopic nerve resection was therefore performed to excise the proximal part. Since then, the pain and related symptoms have completely disappeared without the need to take any drug for two years.

## Discussion

ACNES is reportedly associated with surgery, pregnancy, and exercise, and 77% of such patients are women, while 23% are men, with a wide age range (13-87 years) (4). In children 10-18 years of age, an analysis of chronic abdominal pain led to a diagnosis of ACNES in 12 (12.6%) of 95 children complaining of chronic abdominal pain at outpatient visits (5). The high prevalence of ACNES in teenagers suggests that nerve entrapment might be associated with acute weight gain and/or obesity caused by secondary sexual character changes related to increased endogenous sex steroids (1, 6, 7).

Estrogen and progesterone affect not only the reproductive function but also various physiological functions. For instance, estrogen and progesterone have a regulatory effect on the body fluid volume. Estrogen also promotes the accumulation of subcutaneous fat in contrast to visceral fat (8, 9). In this regard, it has been reported that the ad-

ministration of estradiol to ovariectomized rats actually increased the amount of subcutaneous fat (10). It is also speculated that pain may occur as a result of physical nerve compression due to fat deposition in the abdominal wall (1).

There has been only one well-documented case report suggesting that ACNES may be caused by taking oral contraceptive pills (11). In that case, the region of pain corresponded to the scar after an operation for acute appendicitis. Although the detailed mechanism is still unknown, it has been reported that nerve entrapment in abdominal wall tissues may occur as an adverse effect of oral contraceptive pills, as has been shown in cases of carpal tunnel syndrome (12, 13). It is thought that localized tissue edema caused by estrogen and progesterone may be a trigger for abdominal wall pain (11). A detailed history taking regarding the steroid ingredients is informative when considering the causal relationship of pills with the occurrence of ACNES. The percentage of ACNES patients who have taken oral contraceptive pills is not clear; however, some cases of ACNES during pregnancy, which can induce increased levels of sex steroids, have been reported. Mechanical changes in the abdominal skin and/or subcutaneous edema due to increased estrogen and progesterone levels during pregnancy might cause the ACNES condition in such patients (14).

In conclusion, we encountered a case of ACNES possibly triggered by small-dose oral contraceptives. It is important to consider the possibility of ACNES when patients who are taking pills complain of chronic abdominal wall pain. If surgeons and primary-care physicians are closely involved in patients' care, identifying those who need surgical intervention is easier and may result in the early alleviation of pain. Further studies and the accumulation of cases are necessary to elucidate the mechanism by which oral contraceptives affect the adipose tissue in a specific area of subcutaneous tissue.

**The authors state that they have no Conflict of Interest (COI).**

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