PAIN MEDICINE CASE REPORTS

ISSN 2768-5152

Volume 5, Number 7, pp. 367-371

FASCIAL TEAR IN THE MEDIAL RECTUS ABDOMINIS MUSCLE LEADING TO ANTERIOR CUTANEOUS NERVE ENTRAPMENT SYNDROME

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Background: Although anterior cutaneous nerve entrapment syndrome (ACNES) classically presents with nerve entrapment along the lateral edge of the rectus abdominis, fascial tears can result in ACNES presenting in other locations within the muscle.

Case Report: Over 4 years, a 26-year-old woman had undergone numerous tests to find and treat the cause of her debilitating abdominal pain. She presented to our clinic, in January 2021, with a history of nausea, vomiting, and shooting epigastric pain. Diagnostic anterior and posterior abdominal wall exploration was performed and a percutaneous nerve was entrapped in a fascial tear of the medial rectus abdominis muscle.

Conclusions: This case report highlights that: ACNES can present at locations other than the lateral border of the rectus abdominis; ACNES is a common and curable cause of serious chronic abdominal pain; and knowledge of this pathology has yet to permeate into most of the medical community leading to detrimental outcomes for patients.

Key words: ACNES, abdominal pain, anterior neurectomy, chronic pain, Carnett's test

BACKGROUND

Anterior cutaneous nerve entrapment syndrome (ACNES) is a common, yet overlooked, cause of chronic abdominal pain. One study suggests the average time to diagnosis is almost 4 years (1). Heightened awareness of this disease would likely reduce health care costs and improve quality of life for a substantial number of patients (2). Although ACNES classically presents with nerve entrapment along the lateral edge of the rectus abdominis, fascial tears can result in ACNES presenting in other locations within the muscle. This case report reviews one such case in which a young woman suffered from severe abdominal pain for 4 years due to nerve entrapment along the medial border of the right rectus abdominis muscle.

CASE

A 26-year-old woman presented to our clinic, in January 2021, with a long history of nausea, vomiting, and shooting epigastric pain. The patient had initially presented 4 years earlier, in February 2017, to a gastroenterologist with months of increasingly frequent dyspepsia. She had no significant past medical history, but had graduated college in 2016, and noted that she gained over 30 lbs during her first 2 years of college before subsequently losing 45 lbs during her final 2 years through diet changes and increased exercise. Her body mass index was 22 kg/m^2. Over the next 4 years, she would be prescribed trials of numerous medications. Many of them appeared to relieve symptoms initially, however, the pain would always return. She underwent

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Disclaimer: There was no external funding in the preparation of this manuscript.
Conflict of interest: Each author certifies that he or she, or a member of his or her immediate family, has no commercial association (i.e., consultancies, stock ownership, equity interest, patent/licensing arrangements, etc.) that might pose a conflict of interest in connection with the submitted manuscript.
Accepted: 2021-08-24, Published: 2021-09-30

an esophagogastroduodenoscopy, in July 2017, that demonstrated Grade A esophagitis, erythematous gastric mucosa, and modest duodenal mucosal changes. Biopsies were negative for H. pylori and celiac sprue. In November 2017, the patient reported sharp pain in her abdomen between the rib cage that worsened with exercise. By May 2018, she had changed providers and underwent a full allergy testing panel along with a thyroid and celiac disease lab test workup which was negative. She was presumed to have irritable bowel syndrome (IBS) and further pharmaceutical interventions were initiated. In May 2020, she had severe nausea, vomiting, and abdominal pain, but a contrast abdominal computed tomography and colonoscopy were unremarkable. In August 2020, there were no abnormal findings on abdominal ultrasound. By this time, she had been seen by at least 4 gastroenterologists and a neurologist.

In September 2020, she saw a new gastroenterologist who reported the patient described pain in the epigastric region that felt like a tightness. On exam, the physician noted epigastric pinpoint tenderness and referred the patient to pain management for a trigger point injection (TPI). A first injection helped for a month before severe pain returned. A second injection alleviated the pain for 6 weeks. When the pain returned after the second injection, the patient was referred to our clinic for surgical intervention. Although previously unnoted in documentation, the patient reported she had been suffering significant psychological distress over the course of her medical workup and the pain interfered with her activities of daily living. Upon examination, a defect was palpable in the epigastric area raising suspicion for ACNES or a hernia. A diagnostic laparotomy with additional open abdominal wall exploration and possible neurectomy was recommended and the patient consented.

Surgery was performed 10 days after the consult. Normal stomach, duodenum, gallbladder, appendix, colon, and small bowel were all visualized during the diagnostic laparoscopy. Inspection of the posterior aspect of the anterior abdominal wall revealed no evidence of umbilical or epigastric hernia and no defect at the point of maximal tenderness. A midline abdominal incision was made and blunt dissection was performed to expose the linea alba and medial aspect of rectus abdominis muscle bilaterally. A percutaneous nerve was observed coming through a fascial tear on the right side of the linea alba corresponding to the location of her chronic pain (Figs. 1 and 2). The nerve was excised along with a corresponding neuroma. The fascial defect was closed and the rest of the rectus muscle was inspected for other entrapped nerves. The patient's pain was resolved at one week follow-up surgery and she remained pain free 6 months later, both on subjective pain scale reporting (0/10 severity) and on physical exam with deep palpation at the prior point of maximal tenderness. This marks the longest time she has been pain free in 4 years. She also reported improved quality of life which she attributes to both the pain relief and having fewer instances of anhedonia and depressed mood.

DISCUSSION

This report reviews the case of ACNES presenting in an atypical location due to a fascial tear along the medial border of the rectus abdominis muscle. Despite the atypical location, the patient presented with typical symptoms and exam findings of ACNES and was treated with surgical intervention.

Prevalence, Risk Factors, Pathophysiology

ACNES has been reported numerous times in the literature since Carnett first described it in 1926 (5), however, it remains underrecognized leading to significant and unnecessary morbidity (3,4). The rate of occurrence of ACNES has not been thoroughly documented. McGarrity et al (7) estimated that 10-30% of patients with chronic abdominal pain have ACNES and van Assen et al (6) calculated an incidence of 1/1800 people in the general population and 2% of all patients presenting to the emergency department with abdominal pain in the Netherlands. It is suggested practice for physicians to consider ACNES a likely diagnosis in all patients with chronic abdominal pain and negative workups, although many physicians are unaware of the condition (8). Purported risk factors for ACNES include prior surgery, rectus muscle overuse, obesity, oral contraceptives, and pregnancy (9,10). More recently, it has been reported that patients with weight loss after bariatric surgery may be at higher risk for ACNES (11), although the mechanism is not clear. Moreover, it is difficult to disentangle the independent effect of weight loss from the effect of surgery in bariatric cases. Notably, however, the patient in the current study had significant weight fluctuations prior to the onset of her symptoms, though a causal link was never established. The pathophysiology of the disease is believed to be nerve compression of the cutaneous branches of the intercostal nerves. Most

previously published case reports have documented foramina near the lateral border of the rectus as the location of nerve entrapment, however, in our practice we have seen multiple cases of nerves compressed in fascial tears in various locations across the rectus abdominis muscle (10).

Typical Presentation and Physical Exam Maneuvers

Patients with ACNES typically present with a sharp or burning pain at a pinpoint spot on the abdomen worsened by physical activity or changes in posture. The patient may report altered sensation at the point of tenderness, as well as less specific symptoms, such as changes in appetite, nausea, and vomiting (10). Typically, laboratory and imaging findings are unremarkable (12). Most of these symptoms were reported by our patient early in her workup; the sharp, focal abdominal pain in a single location exacerbated by exercise being the most glaring. Physicians can often make the diagnosis with a focused physical exam. The point of tenderness is roughly the size of a fingertip, and pressing on the area with a single finger intensifies the pain. Carnett's test (14) should be performed in which the patient contracts the rectus muscle while the physician presses on the point of maximal tenderness. This test differentiates intra-abdominal pain from abdominal wall pain with a reported sensitivity of 78% and 88% specificity, respectively, and is likely an effective cost-saving mechanism (13-16). A diagnostic injection of lidocaine at the point of tenderness will often alleviate symptoms. Patients usually see multiple doctors for chronic abdominal pain prior to diagnosis of ACNES and are often misdiagnosed with functional abdominal pain due to apparent manifestations of the psychological effects of living with chronic pain (17,18). Women may be more susceptible to this misdiagnosis due to gender biases in identifying psychosomatic disorders (19,20). In addition to functional abdominal pain, ACNES is often misdiagnosed as IBS. To help alleviate such confusion, van Assen et al (4) have developed a questionnaire to differentiate chronic abdominal wall pain from IBS in patients with overlapping symptoms and negative workups. It is able to detect ACNES with 85% sensitivity and 92% sensitivity, respectively, during an initial clinical encounter.

Treatment

The most common treatment modalities for ACNES are TPIs and anterior neurectomy (8). A diagnostic dose of lidocaine can be given initially, advancing to a larger dose with corticosteroids if pain returns within a short-time frame, and finally surgical intervention for patients with refractory symptoms (1,8,21,22). Ultrasound guidance may improve TPI outcomes by ensuring proper location of injection (23). Anterior neurectomy is a surgical intervention performed under general anesthesia in which the surgeon enlarges the fascial defect of the rectus abdominis muscle through which the entrapped nerve travelled, ligates the nerve in the region of the entrapment, and then closes the defect (24). Anterior neurectomy is efficacious in many patients, with one study (24) citing success rates of 70% in the short term and 61% in long term. Still, this leaves

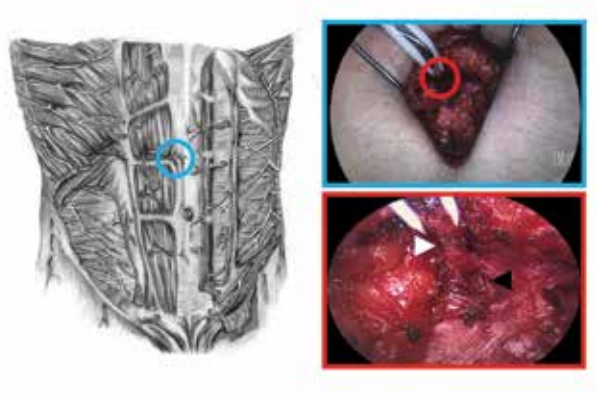

Fig. 1. Images from the surgical intervention. The blue circle on the left panel illustrates the location of the entrapped nerve in the epigastric region along the medial border of the right rectus muscle. The top right panel shows the skin incision and subsequent dissection to the rectus abdominis muscle with the entrapped nerve circled in red. The bottom right panel is a magnified view of the area within the red circle. The nerve (white arrowhead) is entrapped in a fascial tear (black arrowhead).

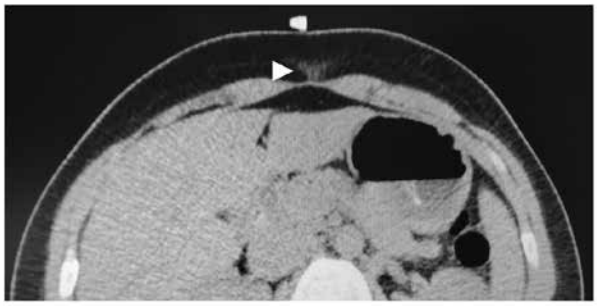

Fig. 2. Preoperative CT scan. The figure shows the preoperative CT scan at the level of the lesion. The location of the lesion is identified by the white arrowhead.

many other patients whose pain returns or never dissipates after surgery. Other diagnoses should certainly be reconsidered if the patient does not improve, however, it is important to note that exploration of the posterior wall has been found to improve outcomes for patients with pain refractory to initial anterior neurectomy (25).

Difficulties With Diagnosis and Management of ACNES

The biggest difficulty with diagnosis of ACNES is lack of awareness by physicians. One study (18) calculated that up to 400,000 patients in the US may be diagnosed with functional abdominal pain that is actually better explained by abdominal wall pain. In fact, for many years, the efficacy of lidocaine injections in treating ACNES was questioned as a possible placebo effect. However, it has now been demonstrated that the therapeutic benefit of lidocaine is due to the analgesic properties of the injection (26). There is an argument to be made that ACNES should become more prevalent in medical school curriculums and board examinations. In one study (27), 85% of physician respondents, including 82% of surgeons, claimed they were unaware of the existence of ACNES. The physical exam techniques to elicit the diagnosis are well within the capacity of every medical trainee and the disease is much more prevalent than many others that are required knowledge for examinations. Since Carnett in 1926 (5), researchers have routinely commented on the need for increased awareness of this pathology in the medical community, yet little progress has been made and patients continue to suffer, with one study (1) reporting an average of 41 months of abdominal pain prior to ACNES diagnosis.

Costs of Untreated Abdominal Pain

Untreated chronic abdominal pain has many adverse effects on patients. For example, chronic pain can lead to many psychiatric conditions and has been associated with increased risk of patients resorting to self-medication with alcohol and narcotic pain medications (28). It increases the risk of suicidal behaviors and completed suicide attempts even when controlling for comorbid psychiatric illness. In a review on the topic, Spiegel et al (29) reported that patients with chronic abdominal pain were up to 11 times as likely to commit suicide as control patients. Although these associations have not been specifically investigated in the ACNES literature, anecdotally, a patient in our clinic did commit suicide 2 days prior to her anterior neurectomy. Furthermore, estimated yearly health care costs were found to be twice as high in the year prior to diagnosis compared to the year after diagnosis in patients with chronic abdominal wall pain (2). Thirty years ago, Greenbaum et al (30) conservatively estimated that over $700 was spent ruling out visceral disease in patients with confirmed abdominal wall pain. Based on current costs of diagnostic imaging procedures, that figure is likely much higher. Together, these results suggest significant health care savings are possible if the diagnosis of ACNES is recognized earlier which is feasible with ameliorations in medical education.

CONCLUSIONS

This case report describes an atypical location of the nerve entrapment in a young woman with a typical clinical presentation of ACNES. Similar to many patients ultimately diagnosed with ACNES, this patient experienced years of chronic abdominal pain, underwent many unnecessary procedures, and suffered psychological distress due to a lack of awareness of the condition. This case report highlights that: ACNES can present at locations other than the lateral border of the rectus abdominis; ACNES is a common and curable cause of serious chronic abdominal pain; and knowledge of this pathology has yet to permeate into most of the medical community leading to detrimental outcomes for patients.

Author Contributions

TH drafted the manuscript. TH, TDPvT, and BJ revised and edited the manuscript. TDPvT provided the illustration. BJ supervised the project.

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