

Sonographic Diagnosis of Abdominal Cutaneous Nerve Entrapment Syndrome: A Report of Two Cases

Hata J* and Imamura H

Department of Clinical Pathology and Laboratory Medicine, Kawasaki Medical School 577, Matsushima, Kurashiki-city, Okayama, 701-0192, Japan

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*Corresponding author:

Jiro Hata, Department of Clinical Pathology and Laboratory Medicine, Kawasaki Medical School 577, Matsushima, Kurashiki-city, Okayama, 701-0192, Japan, Tel: 086-462-1111; Fax: 086-462-1199, E-mail: jhata@med.kawasaki-m.ac.jp; ultrajiro@nifty.com

1. Abstract

We report two cases of Abdominal Cutaneous Nerve Entrapment Syndrome (ACNES) successfully diagnosed using Doppler ultrasound. In both cases, patients complained of chronic abdominal pain, and physical examination showed a positive Carnett's sign. Although the results of several examinations including endoscopy appeared normal, ultrasound examination revealed a perforator artery with its characteristic low peak systolic velocity at the site of the pain. These ultra sonographic findings may facilitate an accurate and objective diagnosis of ACNES.

2. Keywords

Abdominal cutaneous nerve entrapment syndrome; Doppler ultrasound; Perforator artery

3. Introduction

Anterior Cutaneous Nerve Entrapment Syndrome (ACNES) is a condition in which chronic or intermittent pain in the abdominal wall is caused by irritation of cutaneous nerve roots passing through the abdominal fascia [1]. Although ACNES is one of the important causes of abdominal pain, it is not well known among gastroenterologists in Japan. As a consequence, patients with ACNES, after a series of unnecessary examinations, could be misdiagnosed with one of several other diseases, including functional gastrointestinal disorders. These misdirected diagnoses can lead to unnecessary consultation and testing that would have been avoided if the initial diagnosis had been correct. In 2001, [2] noted that an average of \$6727 per patient was spent on diagnostic testing and hospital charges for patients with ACNES prior to achieving an accurate diagnosis. ACNES is currently diagnosed primarily by physical examination; however, there is no standard objective diagnostic method [3]. Although several imaging technologies are available, such as computed tomography and magnetic resonance imaging, there have been no reports on the diagnosis of ACNES applying these methods. Conversely, ultrasound with a high-frequency probe is known to be a non-invasive imaging modality with high spatial and temporal resolution, which can identify the minute vessels accompanying cutaneous nerves. Here, we report two cases of ACNES successfully diagnosed using Doppler ultrasound.

4. Case Report

4.1. Case 1

A 20-year-old woman complaining of left lower quadrant abdominal pain for two months

was admitted to our hospital. The cause of her pain had not been diagnosed despite consultations with several physicians, including a gynecologist, at a previous hospital. Standard physical examination revealed no abnormalities, including her vital signs. Although there was no tenderness of the abdominal region during palpation, the pinch test and Carnett's sign [4, 5] were positive. Laboratory tests showed values within the normal range. She had undergone computed tomography and magnetic resonance imaging at the previous hospital, which showed no abnormality. Ultrasound examination us-

ing an Aplio i800 (Canon Medical Systems Co., Ltd, Nasu, Japan) with a 7 MHz linear probe demonstrated a perforator artery beneath the point of her pain. The waveform analysis of the artery showed a peak systolic velocity (PSV) of 11.8 cm/s, a pulsatility index (PI) of 3.08, and a resistance index (RI) of 1.11 (Figure 1). She was diagnosed with ACNES; however, she did not agree to the recommended treatment because she felt relieved knowing that her pain was not caused by visceral organ disease.

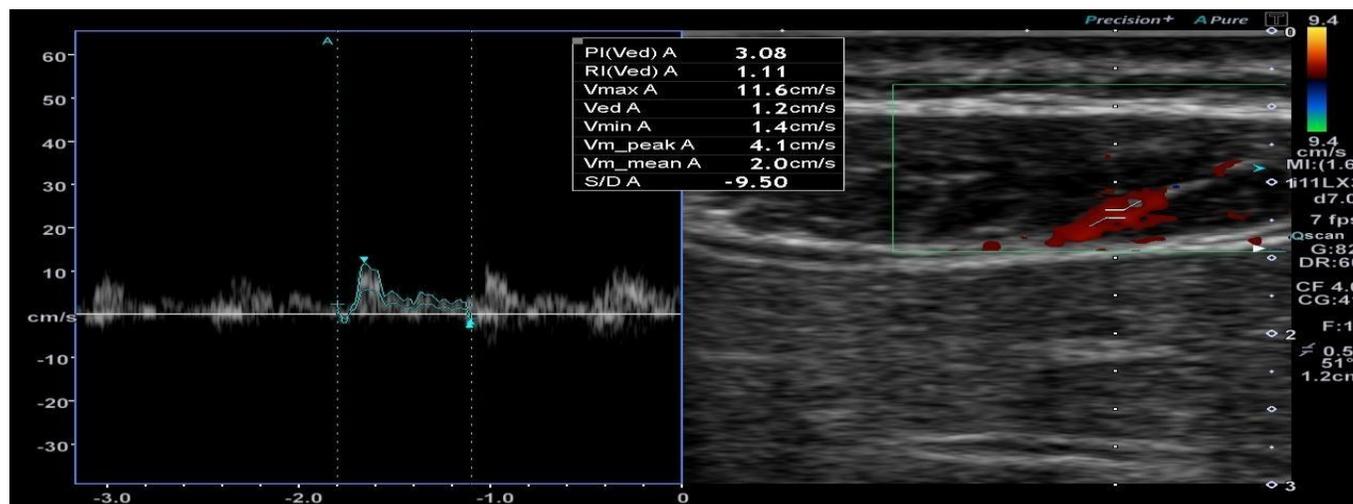


Figure 1: Duplex ultrasound of a perforator artery of Case 1. The waveform analysis of the artery showed a peak systolic velocity of 11.8 cm/sec, a pulsatility index of 3.08, and a resistance index of 1.11

4.2. Case 2

A 66-year-old man presented with chronic mild right upper quadrant abdominal pain for the past six months. The results of upper gastrointestinal endoscopy and abdominal computed tomography were normal. Both physical examination and laboratory tests showed no abnormal findings. He was treated for non-ulcer dyspepsia; however, proton-pump inhibitors and prokinetics did not improve his symp-

toms. Prior to the ultrasound examination, he could finger-point to the region of pain, and Carnett's sign was positive. Similar to Case 1, ultrasound revealed a perforator artery beneath the point of his pain, and the waveform analysis showed a PSV of 7.5 cm/s, a PI of 2.28, and an RI of 0.75 (Figure 2). After injecting 5 mL of 1% lidocaine into the sub fascial space at the site of the pain, it completely resolved and there has been no recurrence of symptoms.

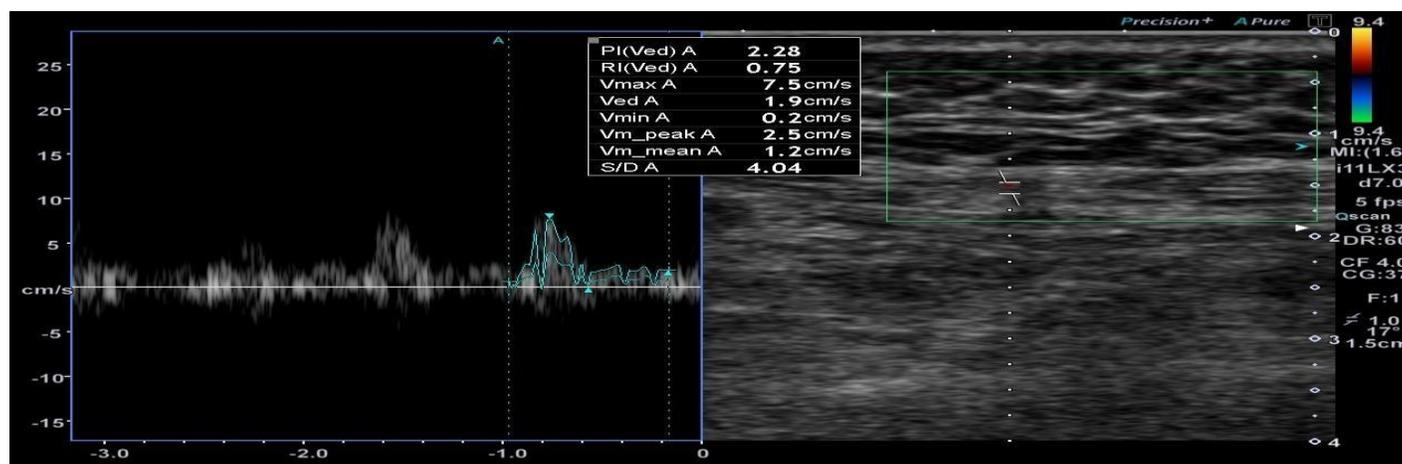


Figure 2: Duplex ultrasound of a perforator artery of Case 2. The waveform analysis showed a peak systolic velocity of 7.5 cm/s, a pulsatility index of 2.28, and a resistance index 0.75.

We also examined perforator arteries of 10 patients (9 men, 1 woman, mean age 58.8 ± 21.0 years old) with acute/chronic abdominal pain associated with other diseases (acute appendicitis, 2; colon cancer, 2; gastric cancer, 2; Crohn's disease, 1; colonic diverticulitis, 2; acute gastritis, 1) to rule out ACNES. The waveform analysis showed the peak velocity of 27.6 ± 10.7 m/s, PI 3.31 ± 1.0 , RI 0.9 ± 0.1 .

(Figure 3) shows the blood flow analysis of a perforator artery in a patient with acute appendicitis. Therefore, PSV of the perforator artery of the patients with ACNES (95% confidence interval [CI]: 16.50–36.60) was significantly lower ($p=0.034$) than that of non-ACNES patients (95%CI: 18.95–36.38), while PI or RI showed no statistically significant differences between each group.

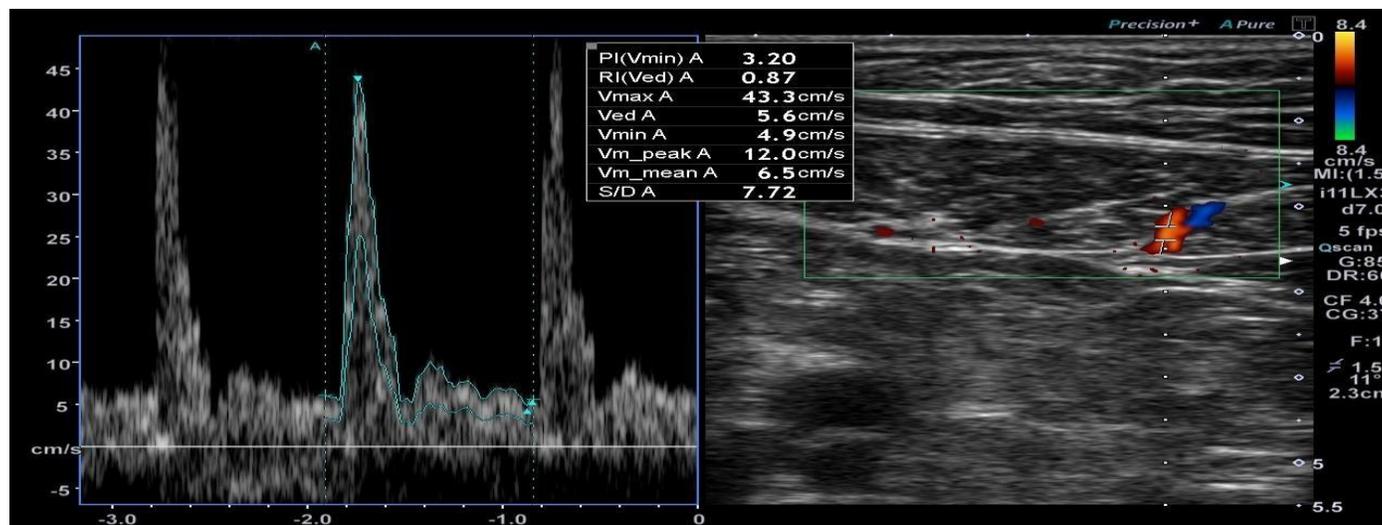


Figure 3: Duplex ultrasound of a perforator artery of a patient with acute appendicitis. The waveform analysis showed a peak systolic velocity of 43.3 cm/s, a pulsatility index of 3.20, and a resistance index 0.87.

5. Discussion

In Japan, as most patients with undiagnosed ACNES complain of abdominal pain, they frequently consult gastroenterologists. Because of the poor recognition of this syndrome among physicians and the difficulty in its diagnosis, patients with ACNES often undergo unnecessary blood tests and imaging studies, which leads to excessive physical and economic complications for patients as well as to the national health insurance system [6].

[2, 7] stated that peripheral nerve entrapment occurs at anatomic sites where the nerve enters a fibrous or osseofibrous tunnel or where the nerve passes over a fibrous or muscular band. In both the aforementioned cases, ultrasound revealed the presence of a perforator artery, which accompanies the nerve. Therefore, detection of perforator arteries at the site of the pain with color Doppler ultrasound is a characteristic finding of ACNES. In addition, the waveform analysis of the perforator arteries showed relatively lower PSVs than of non-ACNES patients, which we believe is another characteristic of ACNES. Since the waveform profiles seen in ACNES patients might resemble post-stenotic arterial flow, we also expected lower PI and RI in ACNES patients, which however did not show statistically significant difference. Based on our experience with these two cases, and the supporting pathophysiology of ACNES, we propose sonographic diagnostic criteria for ACNES as follows: 1) detection of a perforator artery with color Doppler ultrasound beneath the site

of the pain, and 2) a low PSV in waveform analysis, which indicates entrapment of the perforator artery. More experience and studies with larger numbers of patients and controls are needed to establish more precise criteria, including cutoff values for each parameter. As a control, the comparison of the waveform data of the affected perforator artery with that of a nearby non-entrapped artery would be more confirmative.

There are several advantages to establishing sonographic criteria. First, the sonographic criteria could help physicians confirm a diagnosis suspected from physical examinations, since there is no current objective diagnostic method for this syndrome. In addition, it is very difficult to diagnose the change in the arterial flow observed in patients with ACNES using other methods, such as computed tomography, because of limiting spatial resolution. Second, by distributing the concept and diagnostic criteria of ACNES through ultrasound among medical sonographers in Japan, patients will have a better chance of obtaining an accurate diagnosis, thereby avoiding unnecessary examinations, regardless of the attending physicians' knowledge of this syndrome. Third, ultrasound is an excellent guide for the focal injection of lidocaine, which makes therapy more effective and could also be used to rule out this syndrome when no relief of pain is obtained by the injection. Fourth, since ultrasound is a good imaging technique for the diagnosis of visceral diseases causing abdominal pain, performing ultrasound on patients suspected of having

ACNES is useful for either diagnosing or ruling-out this syndrome.

6. Conclusions

We described two cases of ACNES for whom Doppler ultrasound examination showed the characteristic presence of a perforator artery with its characteristic low peak systolic velocity, pulsatility index, and resistance index. These ultra sonographic findings may facilitate the accurate and objective diagnosis of ACNES.

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