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## ● Original Contribution

# CELIAC ARTERY COMPRESSION SYNDROME EVALUATED WITH 3-D CONTRAST-ENHANCED ULTRASONOGRAPHY: A NEW APPROACH

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**Abstract**—This study was performed to estimate the value of 3-D contrast-enhanced ultrasonography (3-D-CEUS) in the diagnosis of celiac artery compression syndrome (CACS). Patients suspected of having CACS were assessed with 3-D-CEUS and contrasted with computed tomography angiography. Diagnostic accuracy was evaluated with a receiver operating characteristic curve. Three-dimensional CEUS revealed 19 positive and 9 negative cases. In the negative group, the contrast agent did not change with respiration. In the positive group, the contrast agent exhibited a hook-shaped stenosis on expiration and returned to normal on inspiration. Computed tomography angiography indicated 1 false-positive case and 1 false-negative case. The sensitivity and specificity of 3-D-CEUS were 95% and 89%, respectively. The area under the receiver operating characteristic curve was 0.982 ( $p < 0.01$ ). In conclusion, 3-D-CEUS can accurately reveal the characteristic hooked appearance and dynamic nature of CACS with respiration, and thus, it represents a new, non-invasive approach to CACS diagnosis. (E-mail: [hxphuaxia@163.com](mailto:hxphuaxia@163.com)) © 2017 World Federation for Ultrasound in Medicine & Biology. All rights reserved.

**Key Words:** 3-D Contrast-enhanced ultrasonography, Celiac artery compression syndrome, Median arcuate ligament syndrome, Computed tomography angiography.

## INTRODUCTION

Celiac artery compression syndrome (CACS), also known as median arcuate ligament (MAL) syndrome and Dunbar's syndrome, is a rare clinical entity. Patients with CACS may exhibit variable symptoms, including postprandial epigastric pain, diarrhea, vomiting and weight loss (Kim et al. 2016), making CACS difficult to distinguish from other diseases that cause chronic abdominal pain. It has been reported that the clinical symptoms of patients with CACS are relieved after open surgical MAL transection and celiac artery revascularization (Sharafuddin et al. 2003). In addition, liver transplantation patients may suffer post-operative hepatic artery thrombosis or even severe liver perfusion failure if patients with CACS are not recognized pre-operatively (Douard et al. 2002). Consequently, accurate differentiation of CACS is of clinical significance. However, the diagnosis of CACS is still controversial (Kim et al. 2016).

Although contrast-enhanced ultrasonography (CEUS) was developed recently, it has already been applied extensively in clinical settings. Molinari et al. (2010) reported that 3-D CEUS (3-D-CEUS) revealed the vascular branches and tortuosity in thyroid masses. Dong et al. (2016) found that dynamic 3-D-CEUS illustrated the arterial continuity and spatial relationships of liver tumors. However, few 3-D-CEUS studies have focused on the mesenteric vasculature. Therefore, we postulated that 3-D-CEUS could be applied in the diagnosis of celiac artery abnormalities.

The aim of this study was to evaluate the diagnostic feasibility and reliability of 3-D-CEUS in patients confirmed as having CACS by computed tomography angiography (CTA), as well to reveal the characteristics of 3-D-CEUS retrospectively. To our knowledge, this is the first assessment of 3-D-CEUS as a technique for diagnosing CACS.

## METHODS

### Patients

Patients who came to our department at the Affiliated Xiaolan Hospital of Southern Medical University for chronic abdominal pain, epigastric fullness or conventional

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health care examination, from March 2011 to July 2016, were involved in the study. All patients accepted abdominal aortic ultrasonography. The inclusion criteria were as follows: (i) Systolic peak velocity when keeping calm (*i.e.*, at relaxation) exceeded 200 cm/s in the proximal celiac artery according to abdominal vascular ultrasonography, velocity decreased on inspiration and increased on expiration or celiac artery occluded (Fig. 1) (ii) clinical laboratory tests, endoscopy, abdominal CT and/or abdominal ultrasonography ruled out obvious liver, gallbladder, spleen, pancreas, urinary tract and gastrointestinal disease; (iii) 3-D-CEUS and CTA examinations were performed and yielded moderate or good image quality. The exclusion criteria were as follows: (i) affliction with an air-filled gut or polypnea and failure to undergo both 3-D-CEUS and CTA; (ii) missing results in a 3-D-CEUS or CTA examination. The study included 23 patients after the criteria described above were applied.

Five patients with suspected renal artery stenosis constituted the control group. The peak systolic velocity of the celiac artery was normal (<180 cm/s) in this group (determined by color Doppler ultrasound) (Fig. 2). The control group was subjected to 3-D-CEUS and CTA. Therefore, 28 patients were enrolled in this study. The experimental protocol was approved by a local institutional review board of Xiaolan Hospital of Southern Medical University. Informed consent for the study was obtained from all human patients in accordance with the World Medical Association Declaration of Helsinki: Ethical Principles for Medical Research Involving Human Patients, 2008.

#### Equipment and methods

A Philips iE33 ultrasound instrument (Philips Medical Systems, Best, Netherlands) with an X3-1 3-D probe was used in this study. All cases were examined by the same ultrasound physician. Patients were in a supine position with calm breathing. The physician initiated the contrast software and infused 1.0 mL of SonoVue rapidly via the basilic vein while the timer was started. Patients were asked to inhale and exhale deeply and to hold their breath when the abdominal aorta was filled with contrast agent. The physician stored the dynamic imaging data of the celiac artery for different respiratory phases (approximately 5 s). Images were transformed into DiCOM format and processed with Philips QLab 9.0 software. The percentage stenosis was measured. All patients underwent CTA examination within 1–3 d after 3-D-CEUS.

**CTA examination.** All patients were scanned during full respiration using a 256-slice CT scanner (Brilliance iCT, Philips Healthcare, Best, Netherlands). The CT technician had more than 5 y of experience. Two different radiologists with more than 10 y of experience assessing abdominal CT reviewed the images from the worksta-

tion together. Both reached a consensus after discussion when their initial diagnosis was controversial.

**CTA diagnostic criteria.** No evident lumen stenosis of the celiac artery during the inspiratory phase and a characteristic hook-shaped appearance during the expiratory phase (Baskan et al. 2015; Ilica et al. 2007), stenosis  $\geq 70\%$ , as well as no obvious atherosclerotic or calcification plaque.

#### Statistical analysis

SPSS 22.0 software was used for statistical analysis (IBM, Armonk, NY, USA). A receiver operating characteristic (ROC) curve was constructed to assess the accuracy and reliability of the diagnosis on the basis of 3-D-CEUS. The threshold of statistical significance was  $p < 0.05$ .

## RESULTS

#### 3-D-CEUS characteristics and clinical data

A total of 28 patients were successfully examined by 3-D-CEUS. No allergic reaction to the contrast microbubbles was observed in any patient. In the control group, the diameter of the contrast agent did not change with respiration, and no frank stenosis was revealed at end-expiration. In the patient group, the contrast agent revealed evident stenosis (Fig. 3a) or was even interrupted at end-expiration and became normal at end-inspiration (Fig. 3b). The percentage of celiac artery stenosis in the patient group ranged from 40% to 100%. Stenosis  $\geq 70\%$  was considered to be a positive 3-D-CEUS result. Three-dimensional CEUS revealed 19 positive cases and 9 negative cases. All 28 patients were confirmed by CTA (Fig. 4a, b). In the control group, all patients were normal and no stenosis was observed at end-expiration. In the patient group, there was 30%–100% stenosis. According to the diagnostic criteria, one false-positive case and one false-negative case were determined by CTA (Table 1). The sensitivity, specificity and accuracy of 3-D-CEUS were 95%, 89% and 93%, respectively. The  $\kappa$  value of the two diagnostic modalities was 0.84.

The 19 positive cases confirmed by CTA had the following characteristics: age range, 19–75 y; mean age,  $37 \pm 10$  y; 10 females and 9 males. Eleven patients complained of epigastric pain, upper abdominal distention, chronic diarrhea or weight loss to variable degrees. Two patients experienced typical exercise-induced abdominal pain that was alleviated after rest. One patient exhibited postprandial fever and insomnia. Five patients were asymptomatic. Physical examination revealed that only 2 of 19 (11%) patients had a vascular bruit in the epigastric abdomen. Among the 14 symptomatic patients, the stenosis of the celiac artery was 70%–95% for 11 patients and 100% for 3 patients. Among the 5 asymptomatic cases, the stenosis was 70%–90% for 3 patients and 100% for 2 patients. Post-stenotic dilation was detected in 7 of 19 (37%) patients, and reverse contrast agent flow in the

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