American Journal of Case Reports

e-ISSN 1941-5923
© Am J Case Rep, 2017; 18: 1334-1342
DOI: 10.12659/AJCR.906283

Received: 2017.07.19
Accepted: 2017.08.16
Published: 2017.12.15

Incidental Anatomic Finding of Celiacomesenteric Trunk Associated with 'Nutcracker Phenomenon,' or Compression of the Left Renal Vein

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Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Female, 91
Final Diagnosis: Nutcracker syndrome • celiacomesenteric trunk
Symptoms: Dyspepsia • dysphagia
Medication: —
Clinical Procedure: —
Specialty: Surgery

Objective: Congenital defects/diseases

Background: Celiacomesenteric trunk (CMT) is a very rare anatomic finding in which the celiac artery and the superior mesenteric artery (SMA) originate from the abdominal aorta through a common trunk. Clinical associations with CMT include arterial aneurysm, thrombosis, and celiac artery compression. However, an association between CMT and abdominal venous congestion caused by left renal vein compression, or 'nutcracker phenomenon,' has not been previously reported.

Case Report: A 91-year-old woman, who died from a cerebrovascular accident (CVA), underwent a cadaveric examination at our medical school. On examination of the abdomen, there was an incidental finding of CMT. The arterial and venous diameters were measured, and vascular histopathology was undertaken. The vascular anatomy was consistent with CMT type 1-b. Nutcracker phenomenon (NCP) (left renal vein compression) was seen anatomically as dilatation and engorgement of the left renal vein, relative to the right renal vein (10.77±0.13 mm *vs.* 4.49±0.56 mm, respectively), and dilatation and engorgement of the left ovarian vein, relative to the right ovarian vein (4.37±0.15 mm *vs.* 1.06±0.09 mm, respectively) with left ovarian varicocele. The aortoceliac angle (ACA) and the aortomesenteric angle (AMA) approached zero degrees.

Conclusions: We have described a rare anatomic finding of CMT that created an acute AMA and NCP. Awareness of this rare association between CMT and NCP by clinicians, vascular surgeons, and radiologists may be of value in the future evaluation and surgical management of patients who present clinically with 'nutcracker syndrome.'

MeSH Keywords: Celiac Artery • Mesenteric Artery, Superior • Renal Nutcracker Syndrome • Renal Veins

Abbreviations: **NCP** – nutcracker phenomenon; **NCS** – nutcracker syndrome; **CMT** – celiacomesenteric trunk; **AMA** – aortomesenteric angle; **ACA** – aortoceliac angle; **TCM I-b** – *truncus celiacomesentericus* 1-b

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/906283

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Background

Traditional anatomy texts describe the abdominal arterial supply arising from a trifurcated celiac trunk (CT), which contains the common hepatic, splenic, and left gastric branches that supply the foregut. A separate superior mesenteric artery (SMA), distal to the CT arising from the aorta, delivers the arterial supply to the midgut and distally, to the inferior mesenteric artery (IMA), which branches off to perfuse the hindgut. Variation in this arterial pattern is uncommon. In 2013, Panagouli et al. reported that almost 90% of individuals possess the standard trifurcated CT, while only 0.76% demonstrate a common origin for both the CT and the SMA, forming a single celiacomesenteric trunk (CMT) [1]. Even though the anatomy of the rare anatomic variation of CMT has been previously described in detail, its clinical significance remains incompletely characterized [2–5]. Clinically, a diagnosis of CMT is important to make as patients with this anatomic variation are at increased risk of arterial aneurysm, thrombosis, occlusive arterial disease, celiac compression syndrome, and the effects of compression from co-existent abdominal aortic aneurysm [2–5].

Nutcracker phenomenon (NCP), or left renal vein entrapment, most commonly occurs when the left renal vein is compressed between the abdominal aorta and the SMA. This anatomic phenomenon may manifest clinically as 'nutcracker syndrome,' which is characterized clinically by hematuria, left-sided flank pain, left varicocele in male patients, abnormal menstruation or pelvic congestion syndrome in female patients, orthostatic intolerance, and chronic fatigue [6–8]. Given these nonspecific findings, NCP may be underdiagnosed. If symptoms do not resolve, clinicians may suggest surgical interventions including intravascular stenting and open surgical correction [9–11]. For patients with both CMT and NCP, an appreciation of the anatomy of CMT anatomy would be of importance in planning safe and effective approaches to endovascular or open surgical correction.

In this report, we present a case of an incidental finding of coincident CMT and NCP, occurring in the body of a deceased 91-year-old woman who was anatomically examined at our medical school. We describe the anatomic relationships of CMT and NCP, discuss their embryological origin, the reasons that CMT can result in NCP, and highlight the importance of awareness CMT as a cause of NCP for clinicians who may diagnose and manage patients who present with 'nutcracker syndrome.'

Case Report

Cadaveric anatomic dissection

A celiacomesenteric trunk (CMT) was found in the body of a deceased 91-year-old woman during routine dissection by medical students at the University of Michigan, USA. The body had been embalmed with a mixture of long term preservation fluid (Trinity Products, LLC, MI, USA) and phenol, prior to dissection. Arterial, venous, and visceral relationships to the CMT, including the superior mesenteric artery (SMA), left renal vein, and the aortomesenteric angle (AMA) were carefully examined and documented during macroscopic dissection of the abdominal and pelvic cavities.

Vascular measurements performed *in situ*

We measured the external arterial and venous diameters using a Westward model 1AAU4 digital caliper (Grainger Inc., USA). At approximately 1 cm distal to the nearest vessel branch point, five measurements were performed at the same location, by a single observer to prevent inter-observer variability. In the case of venous measurements, veins were compressed to measure their external half-circumference. The equation: $d=(2/\pi)(0.5c)$ was used to calculated vein external diameters, where d is the external diameter, and $0.5c$ is the measured external half-circumference.

Vascular histology

Vascular tissue samples were fixed in neutral buffered formalin, processed, sectioned and stained histochemically with hematoxylin and eosin (H&E) and Movat's pentachrome stain by the University of Michigan Department of Pathology histology service. Photomicrograph images were acquired using an Eclipse E400 microscope (Nikon, Tokyo) fitted with a DS-Ri1 camera head (Nikon, Tokyo) and a Digital Sight DS-U3 camera controller (Nikon, Tokyo). Using NIS-Elements BR 4.13.00 imaging software (Nikon, Tokyo), vessel wall thickness (defined as the combined tunica intima and tunica media thickness) was measured and reported as the mean ± standard deviation (SD) of ten individual measurements.

Clinical past medical history

Available medical records for the deceased woman were limited to those created during her hospice stay near the end of her life and were incomplete. However, she had no diagnosis of renal disease and no symptoms of left-sided flank or back pain, orthostatic intolerance, or chronic fatigue was recorded. Renal function tests were unremarkable. No prior imaging studies of the patient could be obtained. The available past medical history was significant for hypertension, chronic gastroesophageal reflux disease, Barrett's esophagus, dysphagia requiring multiple esophageal dilations, and colon cancer. Past surgical history included surgical resection of the colon, but the specific method and extent of the colonic resection were not included. The patient's cause of death was a cerebrovascular accident (CVA). Post mortem height and weight were 64 inches and 89 pounds, respectively.

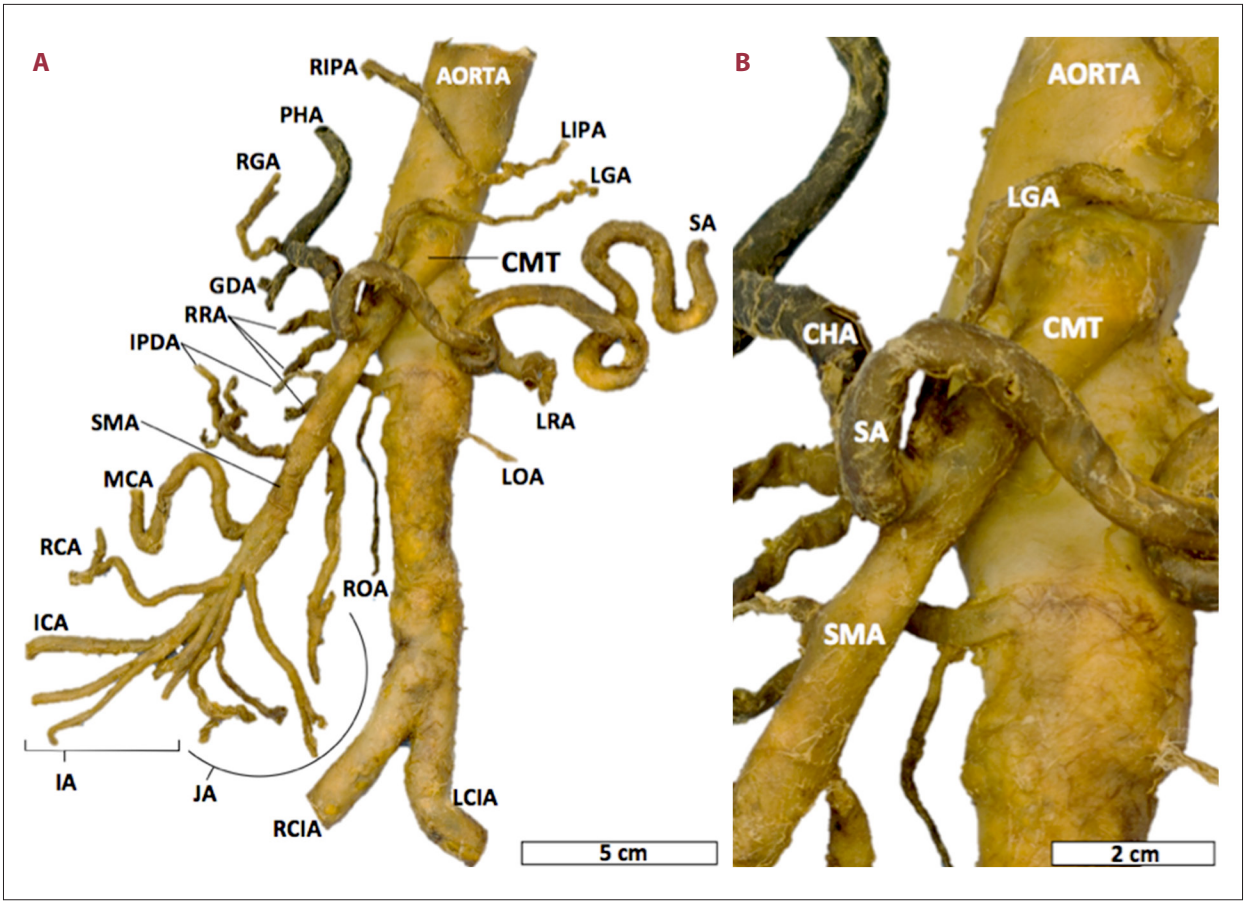

Figure 1. Celiacomesenteric trunk (CMT) in the body of a 91-year-old female discovered during an anatomy demonstration at the University of Michigan Medical School. (**A**) The abdominal aorta, removed from the body. RIPA/LIPA – right and left inferior phrenic artery; PHA – proper hepatic artery; RGA/LGA – right and left gastric artery; SA – splenic artery; GDA – gastroduodenal artery; RRA/LRA – right and left renal arteries; IPDA – inferior pancreaticoduodenal arteries; ROA/LOA – right and left ovarian artery; SMA – superior mesenteric artery; MCA – middle colic artery; RCA – right colic artery; ICA – ileocolic arteries; JA – jejunal arteries; RCIA/LCIA – right and left common iliac artery. (**B**) A close view of the celiacomesenteric trunk (CMT) branches. LGA – left gastric artery; CHA – common hepatic artery; SA – splenic artery; SMA – superior mesenteric artery.

Examination of the arteries

The right and left inferior phrenic arteries arose from the aorta, slightly left of the midline, via a common trunk (Figure 1), which is not an infrequent finding, previously reported in 7% of individuals at post mortem examination [12]. Inferior to the origin of the phrenic arteries, the common CMT arose from the aorta, and was composed of the celiac, left gastric, common hepatic, splenic, and superior mesenteric arteries. The CMT adventitia abutted the aorta directly on branching (Figure 2), eliminating an aortomesenteric angle (AMA) or aortoceliac angle (ACA), which were near-zero degrees on measurement. The AMA could not be measured as the SMA did not branch directly from the aorta, but joined the celiac trunk immediately distal to the origins of the common hepatic artery and splenic artery as if it were a continuation of the celiac artery.

The CMT measured 6.78±0.16 mm in outer diameter and 28.16±1.08 mm in length before giving rise to its first branch: the left gastric artery (Table 1). The left gastric artery was the most superior branch, whereas the common hepatic and splenic arteries were equally inferior on the right side and the left side of the CMT, respectively. The left gastric artery measured 1.98±0.95 mm in outer diameter before branching to supply the posterior body and lesser curvature of the stomach. The splenic artery outer diameter was 5.33±0.31 mm, and gave rise to the dorsal pancreatic artery, greater pancreatic artery, and left gastro-omental artery. The hepatic artery measured 4.90±0.19 mm in outer diameter before branching first into the gastroduodenal artery, and then the supraduodenal artery and hepatic arteries. The SMA measured 6.38±0.37 mm in outer diameter before branching into the following arteries, which arose in the following order: the posterior inferior pancreaticoduodenal

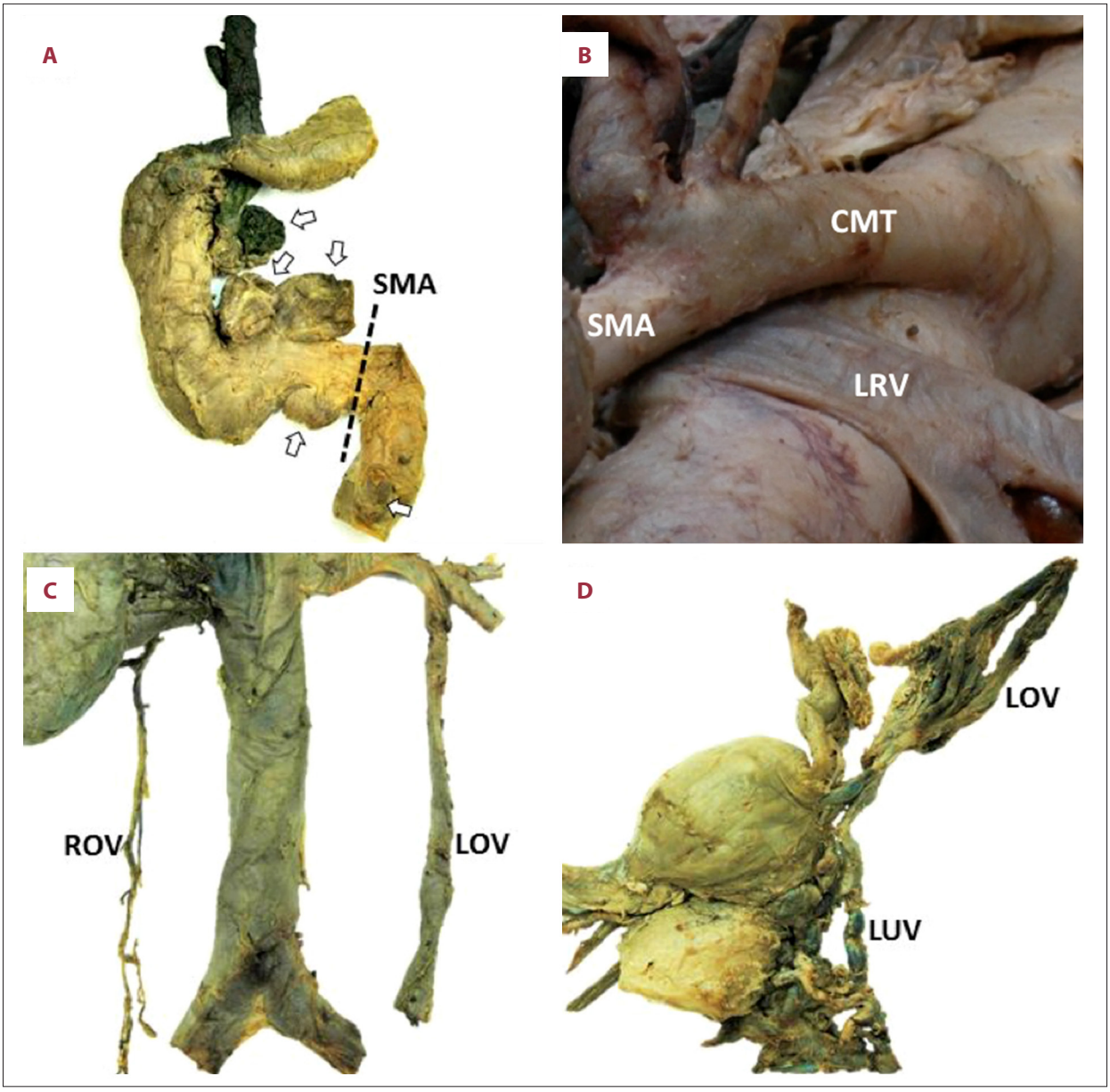

Figure 2. Postmortem anatomic features of nutcracker phenomenon (NCP) found incidentally in a 91-year-old female during an anatomy demonstration at the University of Michigan Medical School. Removed organs are shown in panels **A**, **C**, and **D** to demonstrate the signs of NCP. (**A**) The duodenum with diverticula (arrows) near the site of compression (dashed line) by the superior mesenteric artery (SMA). (**B**) The acute aortoceliac angle (ACA) and the compression of the left renal vein (LRV). The duodenum has been excised to provide a clearer view of the anatomy. (**C**) Comparison of the left and right ovarian veins (LOV, ROV). (**D**) The uterus with the left ovarian vein (LOV), the left uterine vein (LUV), and varicosity of the salpingo-ovarian venous plexus.

artery, the jejunal-ileal arterial trunk, the anterior inferior pancreaticoduodenal artery, a second jejunal-ileal arterial trunk, the middle colic artery, and the jejunal and ileal arteries.

No inferior mesenteric artery was found. Therefore, the entirety of the foregut, midgut, and hindgut was supplied by the CMT alone. The lack of inferior mesenteric artery is was explained by the previous surgical resection of the colon, as recorded in the past medical history. Although it cannot be definitively determined, the inferior mesenteric artery most likely underwent involution and scarring down into the wall of the aorta following a high tie ligation at its root, a common ligation method performed in colon resection [13,14].

Table 1. Measurements of the abdominal vessels.

Vessel	Mean diameter (mm) standard deviation (SD)	
Aorta, superior to the celiac trunk	21.15	(0.45)
Aorta, inferior to the celiac trunk	20.63	(0.78)
Left gastric artery	1.98	(0.95)
Common hepatic artery	4.90	(0.19)
Splenic artery	5.33	(0.31)
Left ovarian artery	3.11	(0.13)
Right ovarian artery	1.99	(0.19)
Left renal vein	10.77	(0.13)
Right renal vein	4.49	(0.56)
Left ovarian vein	4.37	(0.15)
Right ovarian vein	1.06	(0.09)
Superior mesenteric artery (SMA)	6.38	(0.37)
Celiacomesenteric trunk (CMT)	6.78	(0.16)
Marginal artery	2.71	(0.86)
Length of the celiacomesenteric trunk (CMT) (aorta to SMA)	28.2	(1.1)

As aneurysms and occlusive disease have been previously demonstrated to be associated with CMT, sagittal histologic sections of the anastomosis of the celiac trunk and the SMA were stained with Movat's pentachrome to evaluate vascular morphology and to quantify vessel wall thickness (Figure 3) [5]. The combined intimal and medial thickness was measured at the junction site of the SMA and the celiac trunk, as well as immediately proximal and distal to this junction. Due to atherosclerotic plaque deposits in the anterior vessel wall, measurements were taken only from the posterior wall where no deposits were observed. Combined medial and intimal thickness was 578.87±2.63 µm at the proximal celiac artery site, 387.08±2.46 µm at the junction site, and 562.93±3.44 µm at the distal SMA site. These measurements demonstrate a decrease in vessel wall thickness by 33% at the junction site, with a subsequent restoration of 97% of the celiac artery wall thickness at the distal SMA site (Figure 3).

Examination of the veins

Both the portal and caval venous anatomy showed only slight variation from normal. The distal one-third of the functional colo-rectal anatomy contributed to the portal system via the union of the inferior mesenteric vein with the superior mesenteric vein, instead of the splenic vein, which are further likely sequelae of previous colonic resection in this case [13,14]. The proximal one-third of the colon and the small intestine communicated with the portal system via the superior mesenteric vein, and the splenic and superior mesenteric vein joined to form the portal vein.

The right kidney connected to the inferior vena cava via a single right renal vein, which measured 4.49±0.56 mm in outer diameter. A right ovarian vein measuring 1.06±0.09 mm in outer diameter formed a union with the inferior aspect of the right renal vein, a not uncommon variant associated with CMT [15,16]. The outer diameter of the left renal vein as it crossed the aorta was 10.77±0.13 mm, with an adjoining left ovarian vein that measured 4.37±0.15 mm in outer diameter. The left renal vein also appeared significantly flattened as it crossed the aorta (Figure 2). Both the left renal vein and the left ovarian vein outer diameter were much larger than their right-sided counterparts: the left renal vein was increased by 240% compared with the size of the right renal vein, and the left ovarian vein was increased by 412% compared with the right ovarian vein. Furthermore, a left ovarian varicocele was found at the distal end of the left ovarian vein within the left aspect of the broad ligament of the uterus (Figure 2). These findings were consistent with venous congestion [17].

Examination of the viscera

The second portion of the duodenum contained one large diverticulum protruding near the ampulla of Vater and two large diverticula protruding from of the superior aspect of the third portion of the duodenum, and one small diverticulum protruding from the third portion of the duodenum, just proximal to the path of the SMA (Figure 2). Other than one small diverticulum protruding superiorly from the duodenojejunal flexure, no other diverticula of notable size were found in the small intestine. H&E staining showed these diverticula to be false diverticula. The inferior duodenum was flattened at the site of the SMA overlay. These findings support a functionally acute AMA.

Discussion

This report has described a case of celiacomesenteric trunk (CMT) that created an acute aortoceliac angle (ACA) and aortomesenteric angle (AMA), resulting in the development of nutcracker phenomenon (NCP), or compression of the left renal vein between the superior mesenteric artery (SMA) and the aorta [6–8]. Clinically, compression of the left renal vein results in the clinical condition of 'nutcracker syndrome,' which can be asymptomatic or can present clinically with hematuria, left-sided flank pain, abnormal menstruation or pelvic congestion syndrome in women, left varicocele in men, orthostatic intolerance, and chronic fatigue [6–8].

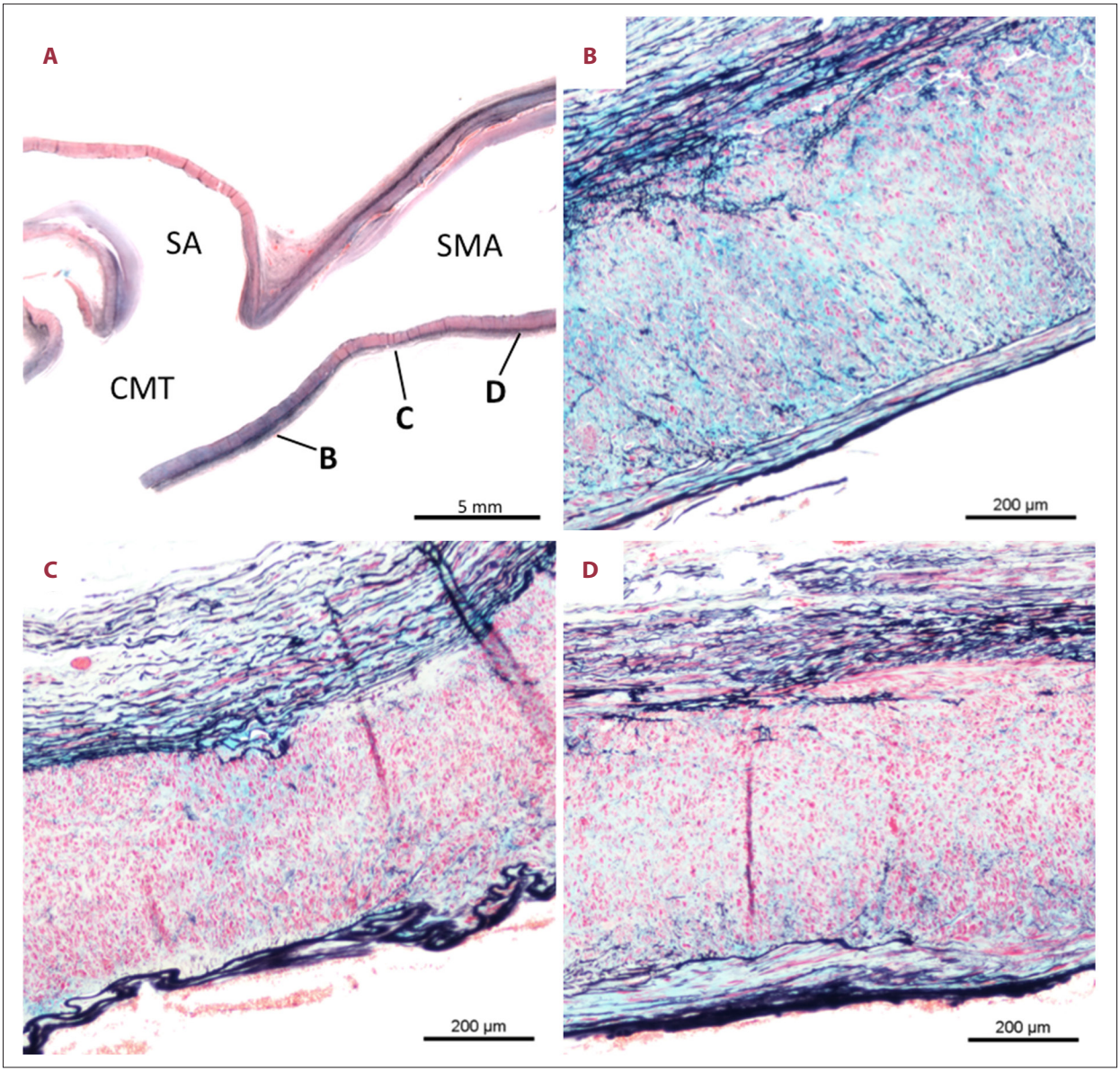

Figure 3. Photomicrographs of the histology of sections of the celiacomesenteric trunk (CMT) and superior mesenteric artery (SMA). Movat's histochemical staining method has been used, which on light microscopy shows elastic fibers (black), collagen fibers (yellow), vascular smooth muscle (red), mucin and ground substance (blue). (**A**) Photomicrograph of a sagittal section of the CMT showing the SMA and splenic artery (SA) branches. Proximal (**B**), medial (**C**), and distal (**D**) measurement points along the junction between the CMT and the SMA are indicated. (**B**) Photomicrograph of the proximal arterial wall from the CMT-SMA junction. Combined medial and intimal thickness: 578.87±2.63 µm. (**C**) Photomicrograph of an arterial section of the CMT-SMA junction. Combined medial and intimal thickness: 387.08±2.46 µm. (**D**) Photomicrograph of the distal arterial wall of the CMT-SMA junction. Combined medial and intimal thickness: 562.93±3.44 µm.

In human embryological development, vitelline (ventral segmental) arteries develop during the fourth week of gestation and a longitudinal anastomosis forms between the vitelline arteries which run parallel to the abdominal aorta [18]. For a typical embryo with distinct blood supply to foregut and midgut, the tenth and thirteenth vitelline arteries persist and develop into the celiac artery and the SMA, respectively. The eleventh and twelfth vitelline arteries and the longitudinal anastomosis bridging the four vitelline arteries each regress, and remnants of the distal tenth, eleventh, and twelfth vitelline arteries join the tenth vitelline artery root to form the left gastric artery, splenic artery, and common hepatic artery, respectively [19]. Embryological explanations for variations in the celiac trunk were proposed by Tandler (1904) who suggested that

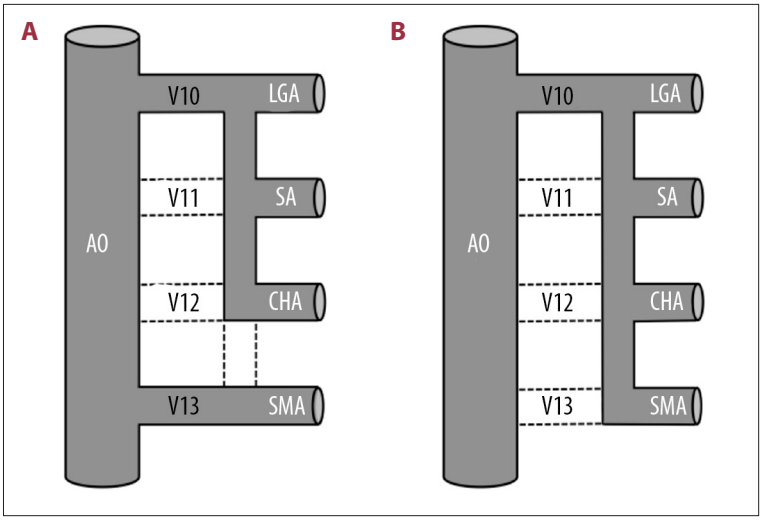

Figure 4. Schematic diagrams illustrating the anatomic variations in the celiac trunk, superior mesenteric artery (SMA), and the celiacomesenteric trunk (CMT) (**A**) The diagram shows the typical celiac trunk and superior mesenteric artery (SMA). The dashed lines indicate regions where the vitelline arteries were obliterated during embryonic development. (**B**) The diagram shows the celiacomesenteric trunk (CMT), classified as *truncus celiacomesentericus* subtype 1-b (TCM 1-b). AO – aorta; LGA – left gastric artery; SA – splenic artery; CHA – common hepatic artery; SMA – superior mesenteric artery.

persistence of the longitudinal anastomoses of the vitelline arteries could result in celiacomesenteric variants [20]. Since then, many variations of CMT have been reported and classified [1,3,21–23].

Previously established classification systems that have organized CMT variants into embryologically defined subtypes suggest a specific etiology for the CMT of this case. In the *truncus coeliacomesentericus* 1-b (TCM 1-b) anatomic variant described by Yi et al. in 2007, the longitudinal anastomosis between the twelfth with the thirteenth vitelline artery persists, while the trunk anchoring the thirteenth vitelline artery to the aorta regresses [23]. This TCM 1-b variant is consistent with the observed CMT, as the left gastric artery, hepatic artery, and splenic artery branch directly from a common trunk composed of anastomosed celiac and superior mesenteric arteries instead of branching from a celiac axis stem abutting this common trunk, as is the case in other subtypes (Figure 4). Furthermore, Wang et al. (2014) noted that CMT subtypes varied significantly in the length of the common trunk, an observation they utilized in their proposed classification parameters as they attributed long subtypes to the persistence of the longitudinal anastomosis between the 12th and 13th vitelline arteries [22]. The length of the CMT from the aorta to its union with the SMA was consistent with the average length of the long subtype reported by Wang et al. (2014) (28.16±1.08 mm *vs.* 27.4±5.0 mm, respectively) in contrast to the reported average length of the short subtype (10.1±1.7 mm) [22]. These observations indicate that the CMT arose because of regression of the thirteenth vitelline artery and persistence of the longitudinal anastomosis between the twelfth and thirteenth vitelline arteries.

Twenty-one cases of CMT and vascular aneurysm have been reported in the past 32 years, and it has been proposed that CMT is predisposed to aneurysm formation [5,18,24]. A predisposition to aneurysm formation is a known trait of other anatomic vascular anomalies, such as persistent sciatic artery, and weakened elastic and smooth muscle layers have been identified in the persistent embryonic artery [25]. Aneurysm formation is also observed in the anomalous origins of the ophthalmic artery, which are known to increase the risk of internal carotid anterior wall aneurysm formation [26]. In this case report, the observed 33% decrease in vessel wall thickness at the anastomosis of the celiac artery and the SMA may support an embryologic origin of vessel wall weakness in this region (Figure 3). CMT may also be associated with an increased risk for aneurysm formation because the vascular trunk experiences excessive blood flow due to the absence of the other splanchnic vessels [5].

Because the CMT is responsible for the blood supply of nearly all the abdominal viscera and lacks collateral blood supply, compromised CMT flow can result in severe splanchnic ischemia and serious clinical complications [27]. There are several pathological associations with CMT that have been reported in the literature, including thrombosis [27], occlusive vascular disease [18,28], celiac compression syndrome [29], and compression from an aortic aneurysm or dissection [18]. However, prior to this case report, and to our knowledge, there has not been a previously reported case of CMT associated with NCP.

The 'nutcracker phenomenon' (NCP), was named for its anatomic etiology, because the left renal vein, much like a nut in a nutcracker, becomes compressed between the superior mesenteric trunk and the aorta [9]. The resulting stenosis increases distal venous pressure from less than 1 mmHg to greater than 3 mmHg, often achieving pressures in the range of 10–20 mmHg [6,30,31]. This increase in venous pressure leads to the pathological conditions associated with 'nutcracker syndrome,' and rupture of thin veins in the renal calyceal fornix of the left kidney produces hematuria; engorgement and reflux of pelvic and gonadal veins produce painful pelvic congestion

or varicocele [6,31–33]. While the diagnosis of 'nutcracker syndrome' relies on clinical manifestations, NCP is established by imaging studies that evaluate for left renal vein stenosis, left renal vein flow velocity, reflux of the left gonadal vein, and engorgement of pelvic veins; these criteria were used to establish NCP in this case, post mortem [34,35].

Consistent with NCP imaging criteria, anatomic dissection of the patient showed a compressed left renal vein, pelvic congestion in the form of a prominent left ovarian varicocele, and a left ovarian vein engorged to 4.37±0.15 mm in outer diameter, more than four times larger than the diameter of its right-sided counterpart (Figure 2). Also, the left renal vein was dilated to an outer diameter of 10.77±0.13 mm compared with the right renal vein outer diameter of 4.49±0.56 mm. Although knowledge of the past medical history was limited, there were no ante-mortem signs or symptoms of 'nutcracker syndrome,' which is not an uncommon association with NCP when identified incidentally on patient abdominal imaging [8,36]. Also, previous reports have shown that only 59% of women with pelvic congestion experience flank pain [32]. Symptoms of 'nutcracker syndrome' are also known to be intermittent, and the resolution over time after the formation of collateral vasculature has been observed [8,37]. Therefore, in this case, the anatomic findings alone sufficiently demonstrate NCP in the absence of reported symptoms.

Narrowing of the AMA, the angle between the SMA and the aorta, is a known cause of NCP. While the reported average AMA varies widely between studies from 38 to 90 degrees, smaller AMA measures are consistently associated with NCP [8,36,38,39]. While a direct measurement of the AMA could not be obtained from this case, because of the embryologic absence of the SMA and aortic root, macroscopically, an acute aortoceliac angle was identified, which is functionally analogous to AMA in the setting of variant CMT anatomy (Figure 2). Notably, a study of varicocele in 75 male patients by Graif et al. (2000) reported a statistically significant association between engorged testicular veins measuring greater than 3 mm in diameter and diminished AMA (mean of 24 degrees), and therefore the engorged left ovarian vein (4.37±0.15 mm in outer diameter) further supports an association between a reduced acute aortoceliac angle and the observed left ovarian varicocele, a known sequela of NCP due to venous hypertension from left renal vein compression [33].

In this case report, the CMT found and described in detail raises further consideration of the relationship between AMA and NCP. In this TCM I-b variant, the left renal vein was no longer juxtaposed to the SMA, and the nearest arterial root, the celiac artery, was displaced superiorly. Therefore, the left renal vein does not benefit from the cavity created by the elastic bending of the SMA as it courses inferiorly to the colon. Given that the TCM I-b SMA followed the normal course, but did not benefit from the normal elastic resistance against compression by overlying structures that is usually provided by a generous AMA, the left renal vein is likely more susceptible to compression in this variant. However, in this case, left renal vein compression may have been exaggerated by the patient's low body mass index (BMI), resulting in the minimal visceral fat being present between the SMA and left renal vein. Also, it is possible that the connection of the SMA to the celiac trunk during development diminishes the exit angle of the celiac artery from the aorta by exerting a greater downward tension, causing a reduced acute ACA. However, the study of more cases of CMT variants, including TCM 1-b variants, may be necessary before specific developmental mechanisms can be proposed. However, the anatomic evidence reported in this case has demonstrated evidence of NCP secondary to a markedly acute ACA. Therefore, CMT and similar anatomic variations should be considered when determining the etiology of NCP.

Conclusions

We have described a case of an incidental and rare finding of celiacomesenteric trunk (CMT) combined with nutcracker phenomenon (NCP) in the body of a deceased 91-year-old woman, donated for anatomy teaching. The CMT created an acute aortomesenteric angle (AMA) and acute aortoceliac angle (ACA), resulting in the development of NCP in this case. Awareness of this rare association between CMT and NCP by clinicians, vascular surgeons, and abdominal radiologists, may be of value in the future evaluation and surgical management of patients who present clinically with 'nutcracker syndrome.'

Acknowledgements

The authors acknowledge the expertise and input of Dr. John Zeller, MD, of the University of Michigan Department of Anatomy. We also acknowledge and thank Dr. Scott Owens, MD, of the University of Michigan Department of Pathology for his expertise regarding the histological analysis, for assisting with tissue processing, histochemical staining, and light microscopy. We especially thank all of the anatomic donors at the University of Michigan and acknowledge that without their generous gift, anatomy research and teaching would not be possible.

Conflicts of interest

None.

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