A case of abnormally dilated and tortuous arc of Buhler and pancreaticoduodenal arteries in the absence of celiac trunk stenosis

Sonali Rathod1 · Riley Kolus1 · Byungchan Kim1 · Sarika Gurnani1 · Andy Kim1 · Erin Kim1 · Faisal Tan1 · Isabelle Van Roy1 · Elizabeth Whitney1 · Maryann MacNeil1 · Jonathan J. Wisco1

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Abstract

Introduction The Arc of Buhler is a rare vascular variant describing a persistent remnant of the embryologic ventral anastomosis between the celiac trunk (CT) and superior mesenteric artery (SMA), invariably reported in the context of CT stenosis. Purpose To report a case of (1) a large and tortuous pancreaticoduodenal arcade and (2) a large and tortuous Arc of Buhler in the absence of celiac axis stenosis. Methods The variant was discovered during routine cadaver dissection. We acquired transverse biopsies of variant vessels and evaluated their wall thickness. Results The donor’s anterior PDA, posterior PDA, and Arc of Buhler had larger diameters, and the common hepatic artery had a smaller diameter than the literature-reported values of a standard human body. The posterior PDA had significantly increased wall thickness compared to the other investigated vessels. Conclusions The Arc of Buhler is a rare remnant of the embryologic ventral anastomosis that is estimated to be hemodynamically active in only half of cases. Previous reports have documented hemodynamically active Arcs of Buhler only in cases of CT stenosis. To the best of the authors’ knowledge, this is a unique case of a persistent and hemodynamically active Arc of Buhler in the absence of CT stenosis. Clinicians should be aware of this variant as its abnormal position may increase risk of herniation and surgical complications, and its tortuosity may increase risk of clot formation.

Keywords Pancreaticoduodenal artery · Arc of Buhler · Embryonic ventral anastomosis · Celiac trunk · Superior mesenteric artery · Anatomical variant

Introduction

Variants in foregut and midgut vasculature have been reported widely. The variants form during the extensive development and lengthening of the digestive tube. Some of the original embryological vessels will remain intact, while others regress and are obliterated. More vessels will develop during these processes as the definitive organs assume their final anatomical position [6]. Reports show variations ranging from more than three branches off the celiac trunk (CT) to anastomoses connecting the CT, superior mesenteric artery (SMA), and inferior mesenteric artery (IMA) per Manoharan and Aland [6]. Often many of these variations can be explained with variations in regression of Tandler longitudinal anterior anastomosis into separate arteries [2, 9]. One such variation is the Arc of Buhler, first described in 1904, and is typically an anastomosis between the CT and the SMA that forms the fourth branch of the CT [1, 3, 7, 9]. The Arc of Buhler was the first of many anastomoses found between the CT and the SMA, including one between the gastroduodenal artery and the inferior pancreaticoduodenal artery, and between the dorsal pancreatic artery and the pancreaticoduodenal arcades [4]. These anastomoses are thought to be caused by an incomplete regression of a portion of the Tandler longitudinal anterior anastomosis.

Sonali Rathod, Riley Kolus and Byungchan Kim contributed equally to the project.

* Sonali Rathod
srathod@bu.edu

1 Department of Anatomy and Neurobiology, Boston University School of Medicine, 72 E Concord St, L-1004, Boston, MA 02118, USA
corresponding to the 10th and 13th vitelline artery branches [2]. The development of the Arc of Buhler is related to its interaction with the dorsal pancreaticoduodenal artery (PDA). The Arc of Buhler, when present as definitive vasculature, can provide circulation to some organs supplied by the PDA. This is especially true when, although rare, branches are present on the Arc of Buhler [8]. We looked into the possible interactions that led to abnormality of both the Arc of Buhler and the PDA. Although no branches were observed on the Arc of Buhler of our donor, there may be a functional reason that supported the persistence of this vessel beyond the embryonic period.

Variations in the PDA have not been well-documented. In a typical phenotype, the superior PDA branches off the gastroduodenal artery which originates at the CT. The inferior PDA commonly comes off the SMA directly or from the first common branch of the jejunal arteries. The superior and inferior PDAs split into anterior and posterior branches and anastomose via arcades and smaller branches at the duodenum. We report herein a case of a persistent Arc of Buhler anastomosis and PDA with unusually dilated and tortuous vessels in the absence of CT stenosis.

**Methods**

The body donor, a 6’0”, 200 lb, 79-year-old white male, was obtained through a generous donation to the Anatomical Gift Program at the Boston University School of Medicine (BUSM). There was a past medical history of Alzheimer’s disease, though the official cause of death was a cardiopulmonary failure. We requested imaging results from the family, but none existed.

A routine abdominal dissection from the lateral omentum to the midline revealed an enlarged, tortuous vessel within the mesentery near the head of the pancreas. Further dissection of the mesentery led to the discovery of two additional large, tortuous vessels embedded in the omentum, the three vessels were found to be part of a series of variants between the SMA and the CT. Visual inspection of the CT revealed no stenosis or abnormality compared with its primary branches. We acquired transverse biopsies of the proximal, middle, and distal segments, relative to the common hepatic artery, of the Arc of Buhler, anterior PDA, and posterior PDA. We also acquired a biopsy of the common hepatic artery between the Arc of Buhler and PDA, and of the proximal splenic artery. Each biopsy was embedded in paraffin, sectioned at a thickness of 6 μm, and stained with hematoxylin and eosin. Sections were scanned using a Panoramic MIDI II (3DHISTECH Ltd., Budapest, Hungary) and visualized using QuPath [1]. We defined and measured the area of the tunica intima + tunica media as the “area of the vessel wall” and the perimeter of the tunica media. We then normalized the wall area to its perimeter for each biopsy. Student’s t tests were performed using SPSS (IBM, Inc., v27) to compare each biopsy area/perimeter measurement against the mean of the same measurements across all biopsies. Statistical significance was reached at an alpha level of 0.05. From the scanned images of the arterial sections, we obtained the areas corresponding to the lumen and the tunica. We calculated the internal diameter of each artery by assuming each artery to have cyclic geometry and calculating the radius of the circle of the same area. A similar calculation was performed to obtain the external diameter of each artery from the cumulative of the lumen and the tunica.

**Results**

The gastroduodenal artery gave rise to the anterior and posterior PDAs. However, these two arteries were extremely large in diameter and formed a long, tortuous pancreaticoduodenal loop that communicated with the SMA near its origin from the abdominal aorta (Fig. 1). We identified them as PDAs, because their origin of branching fit the typical PDA origins and other candidate arteries, such as the right colic artery and splenic artery, were found in expected locations with normal phenotypes [3]. In addition, the common hepatic artery had a large variant branch that descended toward the SMA, where it was anastomosed with the common trunk of the ileocolic and right colic artery. This matched the description of the Arc of Buhler, which is a connection between the CT and the SMA, separate from the gastroduodenal artery [5, 10]. The Arc of Buhler vessel was approximately 19 cm long as measured with string. Other variations were observed in the vasculature of the foregut, neck, and axilla. In the foregut, the right gastric artery originated from the left hepatic artery rather than the proper hepatic artery. In the neck, the superior thyroid artery emerged from the common carotid artery instead of the common trunk of the common carotid artery. In the axilla, the left posterior circumflex humeral artery branched off of the subscapular artery rather than the left axillary artery. The right posterior circumflex humeral artery also abnormally shared a large common trunk with the subscapular artery.

We investigated the enlargement of the PDAs and the Arc of Buhler by conducting a literature review and then compared the arterial diameters obtained from this donor with the diameters reported in cases without a documented variant (Fig. 2). Some of the peer-reviewed articles were limited in providing information for either external or internal diameter. We used the data available and compared it to our calculation of the artery diameters. The splenic artery diameter was measured and compared to the literature, as we believe the abnormalities in the common hepatic artery would not affect the splenic artery.
significantly. Indeed, both the internal and external diameters of the donor’s splenic artery were within the normal range [5].

In comparison, the donor’s anterior PDA, posterior PDA, and the Arc of Buhler had larger diameters than normal (Table 1). Multiple measurements of the donor’s anterior PDA and posterior PDA yielded a larger external diameter than expected in the literature (1.5–3 mm) and a larger internal diameter than the mean (1.5–2.5 mm) [5]. Notably, the diameter of the common hepatic artery in this donor was smaller than that reported in the literature.

**Discussion**

Our donor’s abdominal vasculature demonstrates two abnormal phenotypes: the long and tortuous PDAs, significantly thick posterior PDA walls relative to the other vessels, and...
an equally long and tortuous Arc of Buhler. In comparison with those reported in the literature, our observed PDAs had a greater diameter and an unexpectedly tortuous phenotype (Fig. 1) [5]. In normal physiology, arteries may be tortuous, such that stretching and changing of position of connected structures do not damage the vessels. Examples include the splenic artery that is often stretched with the abdominal body wall. Tortuous vessels may also form due to mechanical instability in blood flow followed by remodeling of the vessel wall [4]. It is not clear whether the tortuosity of the vessels described in this report is embryonic in origin. There was also a difference in the wall thickness and tortuosity of the anterior and posterior PDAs: the posterior PDA was both more tortuous and thicker than the anterior PDA (Table 1). There are many possible reasons why the thickness differs. One possibility is that the patent Arc of Buhler shunted blood away from the anterior PDA, thus reducing the pressure on the vessel wall. However, the numerous and sometimes contradictory factors known to contribute to vessel thickness and tortuosity make it difficult to speculate on a likely cause for this increased thickness [7].

Arcs of Buhler have a 3% prevalence in the general population, with the vessel only hemodynamically active in 50% of cases [9]. A specimen as tortuous as the one in our donor has not been previously documented. Previous reports have demonstrated Arcs of Buhler and PDA’s of similar diameter to those in our donor; however, these have only been described in cases of CT stenosis [11]. Visual inspection of the dissection revealed that the CT was approximately the size in the caliber of the common hepatic and splenic arteries. Absent any evidence of CT stenosis in our donor, this finding represents a previously unreported case of dilated and tortuous anastomotic connections between the CT and the SMA without any obvious causative factors.

Due to the abnormal position and tortuous characteristics of this aberrant vessel, it is important for clinicians to be aware that this type of variant may be present in the general population. The abnormal position may increase the risks of herniation and surgical complications, and the tortuous nature may cause an increased risk of clot formation. The literature already includes examples of hemodynamically relevant Arc’s of Buhler, with one case report describing a fatal retroperitoneal hemorrhage thought to be caused by a persistent Arc of Buhler with associated pseudoaneurysm (BISWAS et al.). Thus, it is essential that surgeons consider abnormal anatomy both at the time of surgery and for future clinical identification of pathologies.

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### Author contributions

RS: project development, data collection, data analysis, manuscript writing/editing. KR: project development, data collection, data analysis, manuscript writing/editing. KB: project development, data collection, data analysis, manuscript writing/editing. GS: project development, data collection, data analysis, manuscript writing/editing. KA: project development, data collection, data analysis, manuscript writing/editing. KE: project development, data collection, data analysis, manuscript writing/editing, figure creation. TF: project development, data collection, data analysis, manuscript writing/editing. VRI: project development, data collection, data analysis, manuscript writing/editing. WE: manuscript writing/editing. MM: manuscript writing/editing. WJJ: project development, data collection, data analysis, manuscript wdraft and notes.

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### Data availability

Additional supporting data can be obtained through the corresponding author, including access to the histology slides. Given the nature of the specimen, the original cadaver used for study is no longer accessible.

### Declarations

#### Competing interests

The authors declare no competing interests.

| Table 1 Comparison of vessel diameter between literature reports and donor, and measured wall thickness of abnormal vessels |
|---|---|---|
| | Normal (mm) | Donor (mm) | Donor (µm) |
| | External | Internal | External | Internal | Vessel wall thickness |
| Anterior PDA | 1.5–3 [5] | – | 3.19 | 3.02 | 167 |
| | | | 3.03 | 2.86 | 166 |
| Posterior PDA | 1.5–3 [5] | – | 4.07 | 3.78 | 296* |
| | | | 4.50 | 4.21 | 291* |
| | | | 4.58 | 4.29 | 291* |
| Arc of Buhler | – | 1.5–2.5 [9] | 4.13 | 3.88 | 248 |
| | | | 3.93 | 3.70 | 230 |
| Splenic artery | 5.0–5.6 [10] | 4.32–5.32 [12] | 5.10 | 4.97 | Not measured |

Empty cells denote inconsistent results per literature search. The significantly thicker arteries (p < 0.05) are indicated with an asterisk.

PDA pancreaticoduodenal artery
Consent to participate The tissue sample used for this study was obtained through the Boston University Anatomical Gifts program. In consenting to donate to this program, subjects consent for their bodies to be used posthumously to further medical education including appropriate research projects. No other subject data was accessed, other than the records that were made available through the program.

Consent for publication We understand that the corresponding author is the sole contact for the editorial process (including editorial manager and direct communications with the office). She is responsible for communicating with the other authors about progress, submissions of revisions and final approval of proofs. We confirm that we have provided a current, correct email address which is accessible by the corresponding author.

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