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The clinical and operative impact of the hexafurcation of the celiac trunk - Report of an extremely rare case

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SUMMARY

The celiac trunk is a main splanchnic artery that supplies the intra-abdominal organs of the upper gastrointestinal system. Anatomic variations of the celiac trunk are encountered in 10% of the population, but can pose challenges to many surgical and interventional procedures. We present here a case of hexafurcation of the celiac trunk discovered preoperatively after image staging for gastric cancer. The branches of the celiac trunk were: left and right inferior phrenic artery, arc of Buhler along with the common pattern of common hepatic, left gastric and splenic artery. The highlight of the case is the rarity of this specific pattern, which is seen in less than 1% of the population, and also the presence of an anastomotic path, the arc of Bühler, as a branch of the celiac trunk. It is evident that the variations of the celiac trunk play a major role in operations of the upper abdomen, radiological interventional procedures and other uncommon cases. Thus, the surgeon must be aware of these variations and their frequency to avoid complications. Also, any physician should report any variation discovered, accidentally or not, to help the literature to be updated.

Key words: Computed tomography – Arc of Bühler – Inferior phrenic arteries variations – Celiac trunk variations – Celiac trunk hexafurcation

INTRODUCTION

The celiac artery, most commonly known as celiac trunk (CT), is one of the main branches of the abdominal aorta, which originates at the level of T12. It is surgically significant as a main splanchnic artery; thus, it has been studied thoroughly (Lipshutz, 1917; Adachi, 1928; Panagouli et al., 2013; Santos et al., 2018; Whitley et al., 2020). The common branching pattern of the celiac artery is the tripus Halleri, which is comprised of the common hepatic, the left gastric and the splenic artery (Panagouli et al., 2013). Anatomical variations of the celiac trunk include variants of the tripod (total absence, bifurcation), aberrant branches (more than three branches of CT), and a common origin with the superior mesenteric artery (celiacomesenteric trunk), or together with the inferior mesenteric artery (celiaco-bimesenteric trunk) (Adachi, 1928; Panagouli et al., 2013). Herein, we present an extremely rare case of hexafurcation of

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the CT, where two parietal branches of the abdominal aorta derived from the CT and also an arch of Buhler (AOB), which is a significant anastomotic pathway, was detected.

CASE REPORT

An 82 years-old male patient was diagnosed with a distal (antrum) gastric cancer. He had a computed tomography (C/T) of the abdomen as part of the staging procedure. The C/T scan revealed a celiac trunk with six branches: common hepatic artery, left gastric artery, splenic artery, left and right inferior phrenic arteries and arc of Bühler (Figs. 1-4). The patient underwent an open subtotal gastrectomy with D2 lymph node dissection and a Roux-en-Y reconstruction. The left gastric artery was ligated for oncologic purposes during lymph node dissection. However, the aberrant branches of the CT were recognized and preserved. There was no intraoperative complication.

His postoperative period was uneventful.

COMMENTS

The variations of the CT are encountered in approximately 10% of the population according to the latest systematic reviews (Panagouli et al., 2013; Whitley et al., 2020). Many different classification systems of the configuration and branching patterns of the CT have been reported, the one by Adachi (1928) being the most frequently used. However, it seems that no existing system covers all the variations discovered, and there is an ongoing effort to create a universal classification of CT variations (Panagouli et al., 2013). In previous century, many large cadaveric studies established the common human vascular anatomy and started to mention variations unknown until then, despite the wide range of the percentages of these variations (Bühler, 1904; Tandler, 1904; Lipshutz, 1917; Adachi, 1928). Nowadays, the study method of the variations has shifted from cadaveric studies to studies via the multi-detector computed tomography (MDCT). Notwithstanding that a few studies comparing these two methods exist, it seems that MDCT has allowed for much larg-

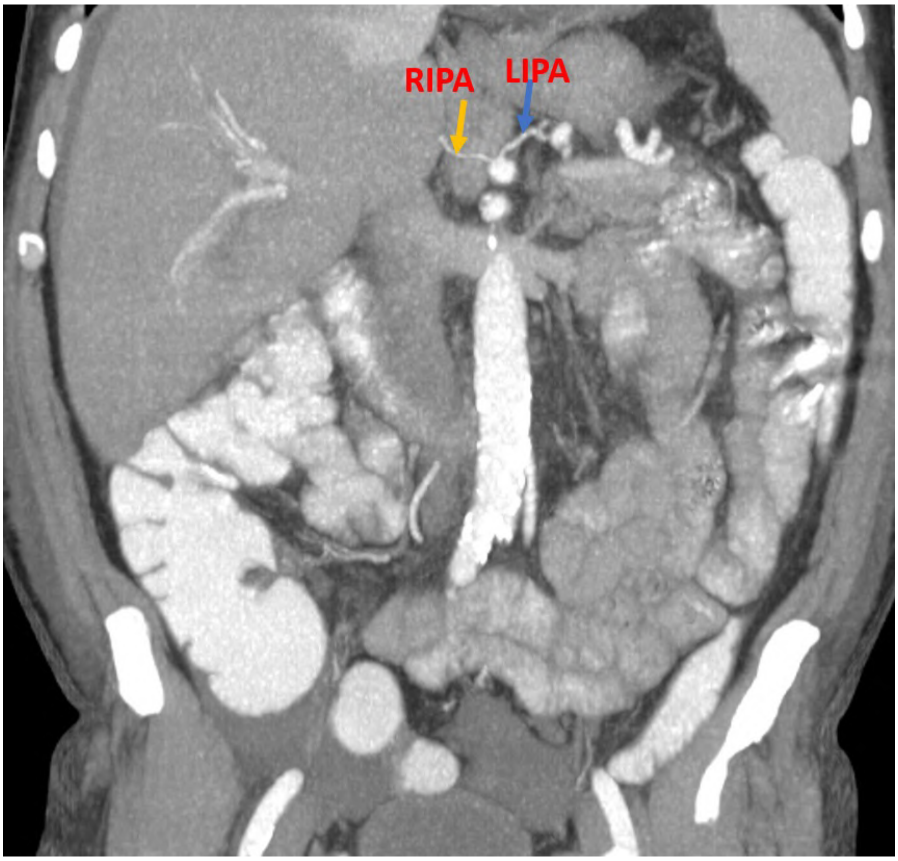

Fig. 1.- MDCT coronal view showing the right inferior phrenic artery (RIPA) (yellow arrow) and the left inferior phrenic artery (LIPA) (blue arrow) arise from the celiac trunk.

er study populations in fewer time and perhaps more accurate percentages, with the downside of the omission of variations beyond MDCT's image quality and the selection bias of the computer tomography's study (Marco-Clement et al., 2016; Whitley et al., 2020).

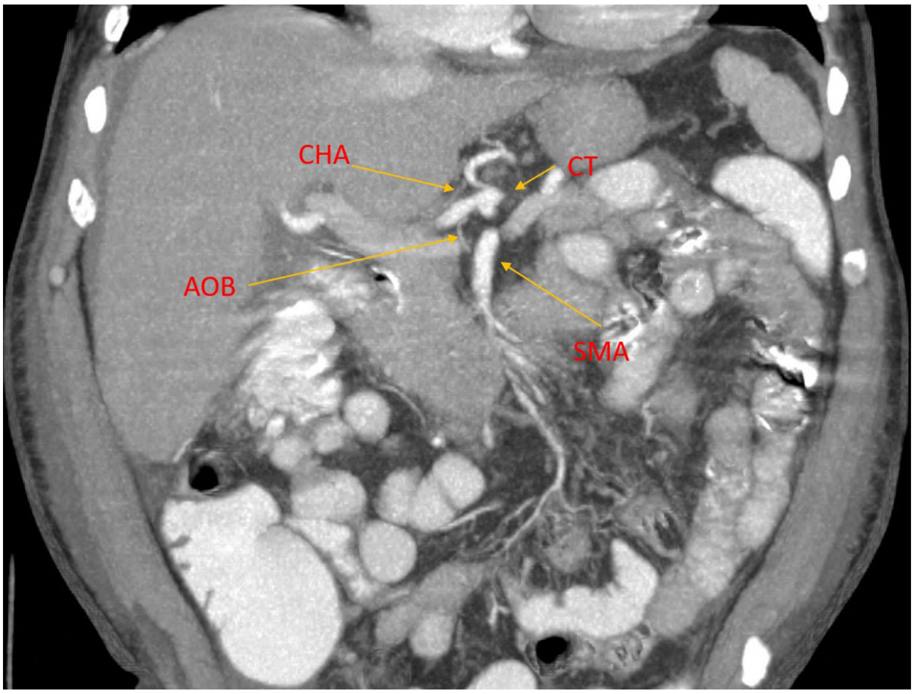

Fig. 2.- MDCT coronal view showing the arc of Bühler (AOB) derived from the celiac trunk (CT) and its anatomical correlations with the common hepatic artery (CHA) and the superior mesenteric artery (SMA).

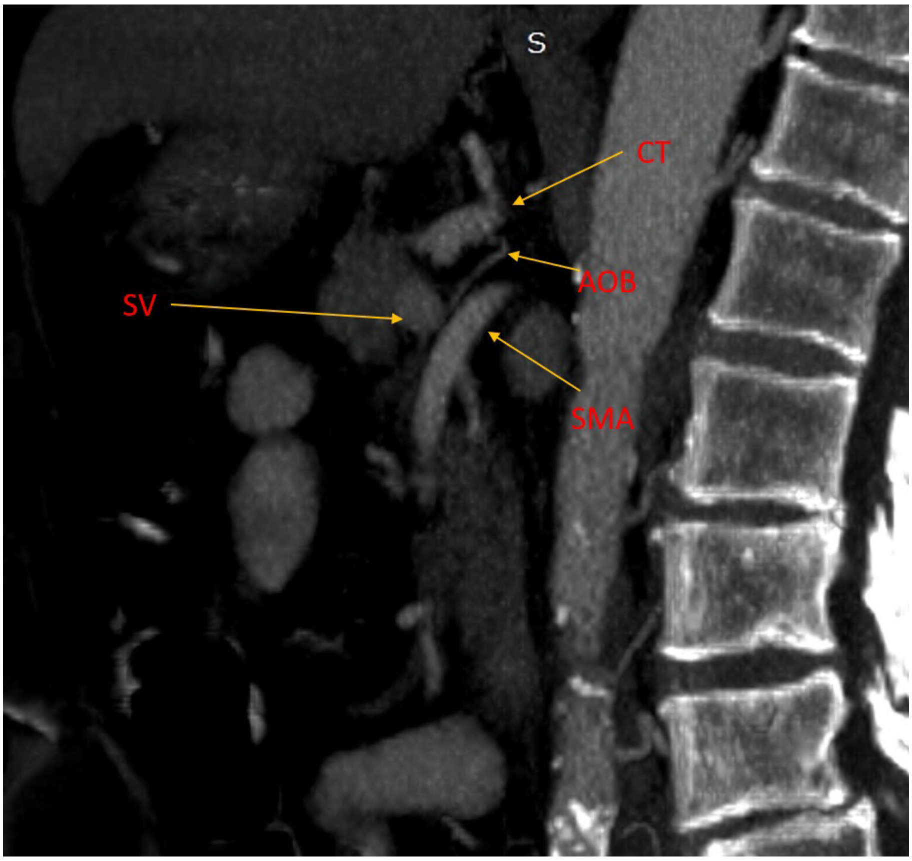

Fig. 3.- MDCT sagittal view showing the arc of Bühler (AOB) with its correlation with the splenic vein (SV). SMA: superior mesenteric artery.

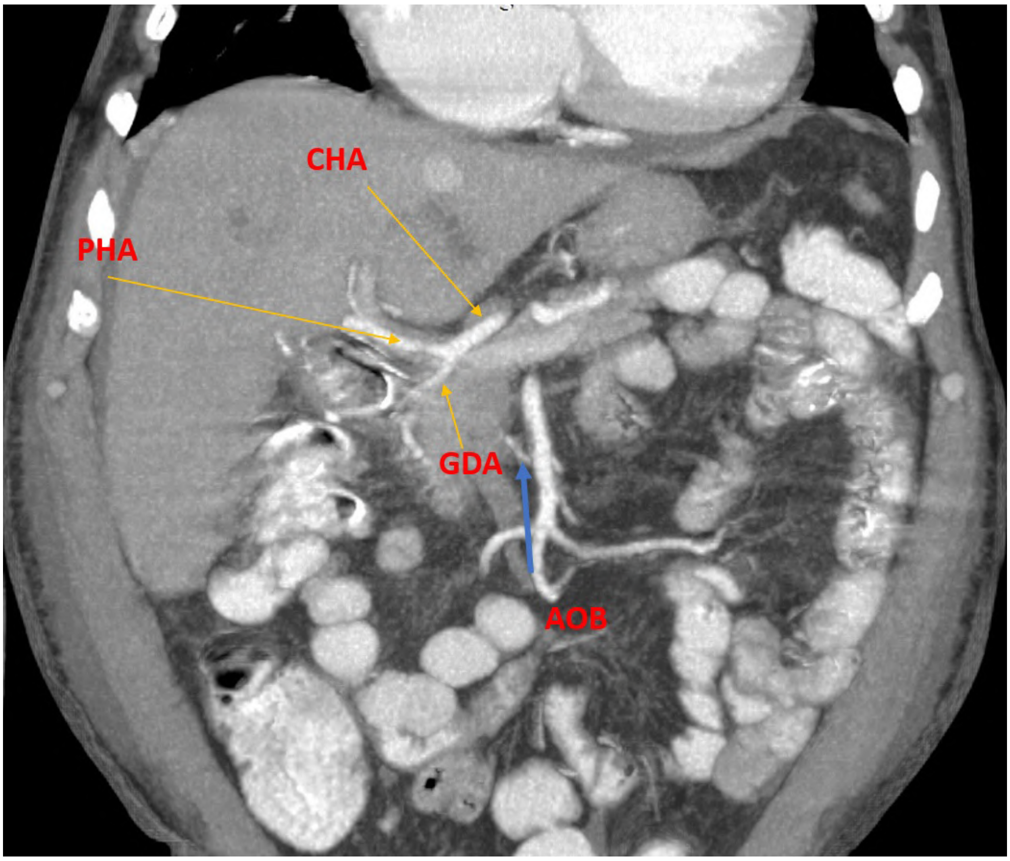

Fig. 4.- MDCT coronal view showing the arc of Bühler (AOB) (blue arrow) with its correlations with the common hepatic artery (CHA), the proper hepatic (PHA) and the gastroduodenal artery (GDA).

The anatomical variations of the CT include an incomplete celiac trunk (missing one of the branches of the tripod, following different patterns), the absence of the tripod, additional branches and a common trunk with other major arteries such as the superior mesenteric artery and the inferior mesenteric artery (Panagouli et al., 2013).

Additional branches of the celiac trunk may be parietal and visceral. The parietal branches include the inferior phrenic arteries (Lipshutz, 1917; Adachi, 1928; Whitley et al., 2020). The inferior phrenic artery may arise as a common trunk, the common inferior phrenic artery (CIPA), or as two individual arteries, the right and left inferior phrenic arteries (RIPA and IPA) (Whitley et al., 2021). They mainly supply the diaphragm, but may also contribute to the arterial supply of the esophagus, stomach, spleen, suprarenal glands, liver, inferior vena cava and adjacent structures (Whitley et al., 2020). The CIPA, according to the latest systematic reviews, is present in approximately 25-29% and most commonly originated from the aorta (57%) (Whitley et al., 2021). Two individual phrenic arteries are seen in about 70-75% of the cases. The origin of the RIPA is primarily the aorta (50%) and the CT (35%), and secondarily the right renal artery and the left gastric, whereas the origin of the LIPA is the aorta and the celiac trunk (appr. 46% each) (Marco-Clement et al., 2016). The concurrent origin of RIPA and LIPA from the celiac trunk is seen in 14%.

The variation of the inferior phrenic arteries can play a major role in some cases of an upper gastrointestinal bleeding due to its contribution to the arterial supply to the esophagogastric junction (Whitley et al., 2021). Also, these arteries could be the cause of extra-bronchial massive hemoptysis, and the lack of awareness could lead to rebleeding after bronchial embolization (Yoon et al., 2002). Also, this may be of great clinical significance in cases of transcatheter arterial chemoembolization (TACE) for inoperable hepatic malignancy (HCC), as the inferior phrenic arteries could be among the tumor-feeding arteries and thus should be embolized for palliative reasons (Whitley et al., 2021).

The arc of Bühler is a rare variation, which was first described from Buhler in 1904 (Buhler, 1904). It anastomoses the CT with the superior mesenteric artery and is encountered in less than 3% of the population, although its exact incidence is still unknown (Saad et al., 2005; Michalinos et al., 2018). The usual anastomoses between the celiac trunk (CT) and the superior mesenteric artery (SMA) are via pancreaticoduodenal arcades and dorsal pancreatic artery (Michalinos et al., 2018). The arc of Bühler (AOB) is an alternative arterial connection of these major arteries, and two major theories have been used to explain its origin. The Mac Kay arc theory suggested that in a 5 mm embryo the distribution of aorta branches is metameric, with each metamere giving rise to 3 pairs of branches, uniting in an arch: the posterior (vertebroparietal), the lateral (genitourinary) arch and the anterior (visceral) arch. During embryonic development variant processes such as fusion and obliteration of branches and development of the digestive tube result in the adult's common anatomy (celiac trunk, superior and inferior mesenteric arteries) (Saad et al., 2005; Douard et al., 2006; Michalinos et al., 2018). The Tandler's longitudinal anastomosis theory, first described by Tandler (1904), is generally used to explain the most of the celiac-mesenteric an intermesenteric anastomoses between splanchnic branches (Lipshutz, 1917; Michalinos et al., 2018). So, AOB is usually thought to be an embryonic remnant due to the failure of regression of the ventral anastomosis between the anlages of the celiac trunk and the superior mesenteric artery (Saad et al., 2005; Douard et al., 2006; Michalinos et al., 2018).

The arc of Bühler is of significant clinical importance as it forms a collateral pathway for arterial perfusion in case of mesenteric or liver ischemia and could be a life-saving variation (Michalinos et al., 2018). Especially, during pancreatic oncologic operations where the ligation of the gastroduodenal artery is mandatory, the presence of the arc acts as an alternative arterial supply to the liver especially if celiac stenosis coexists (McNulty et al., 2001; Manta et al., 2022b; Schumacher et al., 2022). Liver transplantation could also benefit from this arc in the presence of stenosis in the major splanchnic arteries (Incarbone et al., 2021). Also, even mesenteric ischemia due to an aortic dissection could be prevented due to the presence of the Arc of Bühler (Schizas et al., 2020). This arc, augmenting the anastomotic channels between the two arteries, is a positive predictor of adequate blood supply to organs (McNulty et al., 2001; Saad et al., 2005; Douard et al., 2006; Michalinos et al., 2018; Schizas et al., 2020). In interventional radiology it can provide direct access to SMA through the CT and vice versa for diagnostic and even therapeutic reasons (Michalinos et al., 2018).

The clinical significance of the hexafurcation of the CT is evident, during liver, pancreatic, esophagogastric surgery, liver transplantations, organ procurements for transplantation such as liver, pancreas, small bowel or multivisceral ones, lymph node dissection for oncological procedures and vascular surgery (abdominal aortic aneurysm, or even aortic dissection) (McNulty et al., 2001; Saad et al., 2005; Douard et al., 2006; Michalinos et al., 2018; Schizas et al., 2020; Manta et al., 2022b; Schumacher et al., 2022). These variations could complicate the aforementioned procedures with bleeding because of inadvertent injury if the surgical team is not aware of them. Moreover, the effort to avoid any injury to them, may prolong any surgical procedure. In addition, these variations could pose a challenge during the angiographic evaluation of a gastrointestinal bleeding or hemoptysis and the embolism of a tumor or a bleeding lesion (Michalinos et al., 2018).

The importance of this case report relies on the fact that the hexafurcation of the celiac trunk is a rare variant. According to the latest literature, the six-branch pattern of celiac trunk is seen in less than 1% (Panagouli et al., 2013; Manta et al., 2022a). Moreover, this case has a specific clinical impact as, additionally to the two inferior phrenic arteries, the sixth branch of the celiac trunk is not a splanchnic branch but an anastomotic path. The celiac trunk and the superior mesenteric artery have regularly anastomotic channels, but the uniqueness of this case is found in the direct connection of these large arteries with the arc of Bühler. To the best of our knowledge, this specific pattern has been reported only once before in literature with the arc of Bühler inserting into the first jejunal artery, and not into the trunk of the superior mesenteric artery as we have mentioned here (Manta et al., 2022a).

Despite the well-established importance of AOB, there is still no mention of it in the current textbooks of anatomy (McNulty et al., 2001). Furthermore, the embryological hypothesis has not been proved yet (Michalinos et al., 2018). Surgeons and radiologists around the world have not concluded a commonly agreed system of classification of vascular variations (Panagouli et al., 2013). Pre-operative staging even for the most obscure areas of upper abdomen such as pancreas does not universally include MDCT angiography for establishment of vascular pattern. So, there is still work to be done and this case underline this fact.

CONCLUSION

It is obvious that the variations of the celiac trunk have an important clinical impact especially on the operative approach of the upper abdomen, and the surgeon should be aware of them. Thus, it is crucial to establish a pre-operative investigation which may include evaluation of the celiac trunk, in order to map out a better intra-operative plan. It is important and extremely useful to report any new variants discovered, accidentally or not, because the knowledge of CT branching pattern could improve the therapeutic results for a wide range of surgical and radiological, abdominal and vascular procedures.

AUTHORS' CONTRIBUTION

A Anagnou: Manuscript elaboration, data analysis, literature search and documentation. A. Pentheroudaki: Data management and collection. E. Lolis: Original idea, manuscript editing and critical review. All the authors read and approved the final manuscript.

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