World J Clin Cases 2022 April 6; 10(10): 2976-3320





#### **Contents**

Thrice Monthly Volume 10 Number 10 April 6, 2022

#### **REVIEW**

2976 Gut microbiota in gastrointestinal diseases during pregnancy

Liu ZZ, Sun JH, Wang WJ

2990 Targeting metabolism: A potential strategy for hematological cancer therapy

Tang X, Chen F, Xie LC, Liu SX, Mai HR

#### **MINIREVIEWS**

3005 Elevated intra-abdominal pressure: A review of current knowledge

Łagosz P, Sokolski M, Biegus J, Tycinska A, Zymlinski R

#### **ORIGINAL ARTICLE**

#### **Case Control Study**

3014 Changes in corneal nerve morphology and function in patients with dry eyes having type 2 diabetes

Fang W, Lin ZX, Yang HQ, Zhao L, Liu DC, Pan ZQ

3027 Combined sevoflurane-dexmedetomidine and nerve blockade on post-surgical serum oxidative stress

biomarker levels in thyroid cancer patients

# **Retrospective Cohort Study**

Du D, Qiao Q, Guan Z, Gao YF, Wang Q

Early warning prevention and control strategies to reduce perioperative venous thromboembolism in 3035 patients with gastrointestinal cancer

Lu Y, Chen FY, Cai L, Huang CX, Shen XF, Cai LQ, Li XT, Fu YY, Wei J

3047 Dose-response relationship between risk factors and incidence of COVID-19 in 325 hospitalized patients: A multicenter retrospective cohort study

Zhao SC, Yu XQ, Lai XF, Duan R, Guo DL, Zhu Q

#### **Retrospective Study**

3060 Preventive online and offline health management intervention in polycystic ovary syndrome

Liu R, Li M, Wang P, Yu M, Wang Z, Zhang GZ

3069 Evidence-based intervention on postoperative fear, compliance, and self-efficacy in elderly patients with

hip fracture

Fu Y, Zhu LJ, Li DC, Yan JL, Zhang HT, Xuan YH, Meng CL, Sun YH

Significance of dysplasia in bile duct resection margin in patients with extrahepatic cholangiocarcinoma: A 3078

retrospective analysis

Choe JW, Kim HJ, Kim JS

#### Contents

#### Thrice Monthly Volume 10 Number 10 April 6, 2022

3088 Diagnostic value and safety of medical thoracoscopy for pleural effusion of different causes

Liu XT, Dong XL, Zhang Y, Fang P, Shi HY, Ming ZJ

#### **Observational Study**

3101 Oxaliplatin-induced neuropathy and colo-rectal cancer patient's quality of life: Practical lessons from a prospective cross-sectional, real-world study

Prutianu I, Alexa-Stratulat T, Cristea EO, Nicolau A, Moisuc DC, Covrig AA, Ivanov K, Croitoru AE, Miron MI, Dinu MI, Ivanov AV, Marinca MV, Radu I, Gafton B

3113 Breast-conserving surgery and sentinel lymph node biopsy for breast cancer and their correlation with the expression of polyligand proteoglycan-1

Li FM, Xu DY, Xu Q, Yuan Y

#### **SYSTEMATIC REVIEWS**

3121 Clinical significance of aberrant left hepatic artery during gastrectomy: A systematic review

Tao W, Peng D, Cheng YX, Zhang W

#### **META-ANALYSIS**

3131 Betel quid chewing and oral potential malignant disorders and the impact of smoking and drinking: A meta-analysis

Lin HJ, Wang XL, Tian MY, Li XL, Tan HZ

3143 Effects of physical exercise on the quality-of-life of patients with haematological malignancies and thrombocytopenia: A systematic review and meta-analysis

Yang YP, Pan SJ, Qiu SL, Tung TH

#### **CASE REPORT**

3156 Primary malignant peritoneal mesothelioma mimicking tuberculous peritonitis: A case report

Lin LC, Kuan WY, Shiu BH, Wang YT, Chao WR, Wang CC

3164 Endoscopic submucosal dissection combined with adjuvant chemotherapy for early-stage neuroendocrine carcinoma of the esophagus: A case report

Tang N, Feng Z

3170 Lymph-node-first presentation of Kawasaki disease in a 12-year-old girl with cervical lymphadenitis caused by Mycoplasma pneumoniae: A case report

Kim N. Choi YJ. Na JY. Oh JW

3178 Tuberculosis-associated hemophagocytic lymphohistiocytosis misdiagnosed as systemic lupus erythematosus: A case report

Chen WT, Liu ZC, Li MS, Zhou Y, Liang SJ, Yang Y

3188 Migration of a Hem-o-Lok clip to the renal pelvis after laparoscopic partial nephrectomy: A case report

Π

Sun J, Zhao LW, Wang XL, Huang JG, Fan Y

#### Contents

# Thrice Monthly Volume 10 Number 10 April 6, 2022

3194 Ectopic intrauterine device in the bladder causing cystolithiasis: A case report Yu HT, Chen Y, Xie YP, Gan TB, Gou X 3200 Giant tumor resection under ultrasound-guided nerve block in a patient with severe asthma: A case report Liu Q, Zhong Q, Zhou NN, Ye L 3206 Myomatous erythrocytosis syndrome: A case report Shu XY, Chen N, Chen BY, Yang HX, Bi H 3213 Middle thyroid vein tumor thrombus in metastatic papillary thyroid microcarcinoma: A case report and review of literature Gui Y, Wang JY, Wei XD 3222 Severe pneumonia and acute myocardial infarction complicated with pericarditis after percutaneous coronary intervention: A case report Liu WC, Li SB, Zhang CF, Cui XH 3232 IgA nephropathy treatment with traditional Chinese medicine: A case report Zhang YY, Chen YL, Yi L, Gao K 3241 Appendico-vesicocolonic fistula: A case report and review of literature Yan H, Wu YC, Wang X, Liu YC, Zuo S, Wang PY 3251 Scedosporium apiospermum infection of the lumbar vertebrae: A case report Shi XW, Li ST, Lou JP, Xu B, Wang J, Wang X, Liu H, Li SK, Zhen P, Zhang T 3261 Woman diagnosed with obsessive-compulsive disorder became delusional after childbirth: A case report Lin SS, Gao JF 3268 Emphysematous pyelonephritis: Six case reports and review of literature Ma LP, Zhou N, Fu Y, Liu Y, Wang C, Zhao B 3278 Atypical infantile-onset Pompe disease with good prognosis from mainland China: A case report Zhang Y, Zhang C, Shu JB, Zhang F 3284 Mycobacterium tuberculosis bacteremia in a human immunodeficiency virus-negative patient with liver cirrhosis: A case report Lin ZZ, Chen D, Liu S, Yu JH, Liu SR, Zhu ML 3291 Cervical aortic arch with aneurysm formation and an anomalous right subclavian artery and left vertebral artery: A case report Wu YK, Mao Q, Zhou MT, Liu N, Yu X, Peng JC, Tao YY, Gong XQ, Yang L, Zhang XM 3297 Dedifferentiated chondrosarcoma of the middle finger arising from a solitary enchondroma: A case report Yonezawa H, Yamamoto N, Hayashi K, Takeuchi A, Miwa S, Igarashi K, Morinaga S, Asano Y, Saito S, Tome Y, Ikeda H,

Ш

Nojima T, Tsuchiya H

# **Contents**

# Thrice Monthly Volume 10 Number 10 April 6, 2022

Endoscopic-catheter-directed infusion of diluted (-)-noradrenaline for atypical hemobilia caused by liver 3306 abscess: A case report

Zou H, Wen Y, Pang Y, Zhang H, Zhang L, Tang LJ, Wu H

Pneumocystis jiroveci pneumonia after total hip arthroplasty in a dermatomyositis patient: A case report 3313 Hong M, Zhang ZY, Sun XW, Wang WG, Zhang QD, Guo WS

ΙX

#### Contents

# Thrice Monthly Volume 10 Number 10 April 6, 2022

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CASE REPORT

# Cervical aortic arch with aneurysm formation and an anomalous right subclavian artery and left vertebral artery: A case report

Yao-Kun Wu, Qi Mao, Mao-Ting Zhou, Ning Liu, Xi Yu, Jin-Cheng Peng, Yun-Yun Tao, Xue-Qin Gong, Lin Yang, Xiao-Ming Zhang

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#### **Abstract**

#### **BACKGROUND**

A cervical aortic arch (CAA) refers to a high-riding aortic arch (AA) that often extends above the level of the clavicle. This condition is very rare, with an incidence of less than 1/10000.

#### CASE SUMMARY

A 29-year-old woman was admitted to the otolaryngology department of our hospital for repeated bilateral purulent nasal discharge for the prior 3 mo. The patient was diagnosed with chronic sinusitis and chronic rhinitis at admission. A preoperative noncontrast chest computed tomography scan showed a high-riding, tortuous AA extending to the mid-upper level of the first thoracic vertebra with local cystic dilatation. A further computed tomography angiography examination showed that the brachiocephalic trunk, left common carotid artery, left vertebral artery (LVA) (slender), and left subclavian artery sequentially branched off of the aorta from the proximal end to the distal end of the AA. The proximal end of the right subclavian artery (RSCA) was tortuous and dilated. The AA showed tumorlike local expansion, with a maximum diameter of approximately 4 cm. After consultation with the department of cardiac macrovascular surgery, the patient was diagnosed with left CAA with aneurysm formation and an anomalous RSCA and LVA and was transferred to that department. The patient underwent AA aneurysm resection and artificial blood vessel replacement under general anesthesia and cardiopulmonary bypass. No abnormality was found during the 2mo follow-up after discharge.

April 6, 2022 | Volume 10 | Issue 10

#### **CONCLUSION**

A CAA is a rare congenital anomaly of vascular development. The present unique case of CAA with aneurysm formation and an anomalous RSCA and LVA enriches existing CAA data.

Key Words: Cervical aortic arch; Aortic aneurysm; Aortic anomaly; Computed tomography angiography; Haughton classification; Case report

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Core Tip: A cervical aortic arch (CAA) is a rare congenital anomaly of vascular development and refers to a high-riding aortic arch that often extends above the level of the clavicle. This paper reports a case of a unique CAA with aneurysm formation and an anomalous right subclavian artery and left vertebral artery. The present unique case enriches existing CAA data.

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#### INTRODUCTION

A cervical aortic arch (CAA) is a rare congenital anomaly of vascular development and refers to a highriding aortic arch (AA) that often extends above the level of the clavicle [1-4]. This paper reports a case of a unique CAA with aneurysm formation and an anomalous right subclavian artery (RSCA) and left vertebral artery (LVA).

#### CASE PRESENTATION

#### Chief complaints

A 29-year-old woman presented repeated bilateral purulent nasal discharge for 3 mo.

#### History of present illness

The symptoms started 3 mo prior to presentation, with dizziness and headache.

#### History of past illness

The patient underwent a cesarean birth at a local hospital 4 years prior.

#### Personal and family history

The patient had no family history that was related to the present illness.

### Physical examination

Physical examination showed a slightly congested mucosa of the bilateral nasal cavity, a slightly enlarged inferior turbinate, a hypertrophic middle turbinate, and a small amount of purulent nasal discharge in both middle nasal passages and posterior nostrils.

#### Laboratory examinations

Laboratory tests showed that the serum uric acid (428.9 µmol/L), fibrin (4.9 g/L), erythrocyte sedimentation rate (30 mm/h) and ultrasensitive thyroid stimulating hormone (5.1 µIU/mL) values were slightly elevated, the myoglobin (< 21 ng/mL) level was slightly decreased, and the complete blood count, electrolyte profiles and liver function were normal.

#### Imaging examinations

A computed tomography scan at another hospital showed bilateral maxillary sinusitis and ethmoiditis and bilateral inferior turbinate hypertrophy. The patient was diagnosed with chronic sinusitis and chronic rhinitis at admission. A preoperative noncontrast chest computed tomography scan showed a high-riding, tortuous AA extending to the mid-upper level of the first thoracic vertebra with local cystic dilatation, and a few calcified plaques were identified in the descending aorta (DA). A further computed tomography angiography examination showed that the ascending aorta was located to the right of the spine. The brachiocephalic trunk (BCT), left common carotid artery (LCCA), LVA (slender), and left subclavian artery (LSCA) sequentially branched off of the aorta from the proximal end to the distal end of the AA. The BCT bifurcated into the right common carotid artery and the RSCA. The proximal end of the RSCA was tortuous and dilated (approximately 1.5 cm in diameter). The AA showed tumor-like local expansion, with a maximum diameter of approximately 4.0 cm. The long and tortuous AA distal to the aneurysm was located to the left of the spine, extended upward, surpassed the level of the clavicle, and reached the mid-upper level of the first thoracic vertebra. Then, it extended downward tortuously along the left side of the spine to become the DA (Figure 1).

#### **FINAL DIAGNOSIS**

The final diagnosis of the present case was left CAA with aneurysm formation and an anomalous RSCA and LVA.

# **TREATMENT**

After consultation with the department of cardiac macrovascular surgery, the patient was diagnosed with left CAA plus aneurysm formation and an anomalous RSCA and LVA and was transferred to that department. The patient underwent AA aneurysm resection and artificial blood vessel replacement under general anesthesia and cardiopulmonary bypass (CPB) on July 10, 2021. The left CAA and the aneurysm (with a maximum diameter of approximately 4.0 cm) that had formed between the LCCA and the LSCA were observed during the operation. During the intraoperative period, a total of 2 units of type O suspended red blood cells, 10 units of cryoprecipitated antihemophilic factors, and 500 mL of fresh frozen plasma were transfused, and vasopressor drugs, including 8  $\mu$ g deoxyepinephrine, 128  $\mu$ g adrenaline, and 38478  $\mu$ g dopamine, were infused with an infusion pump control to manage blood pressure. The total CPB time was 106 min, the ascending aorta was blocked for 44 min, and circulation was stopped for 15 min. After successful surgery, the patient received intensive care, anti-infection treatment, respiratory and circulatory function maintenance, and microcirculation improvement therapy for 44 h and 50 min.

# **OUTCOME AND FOLLOW-UP**

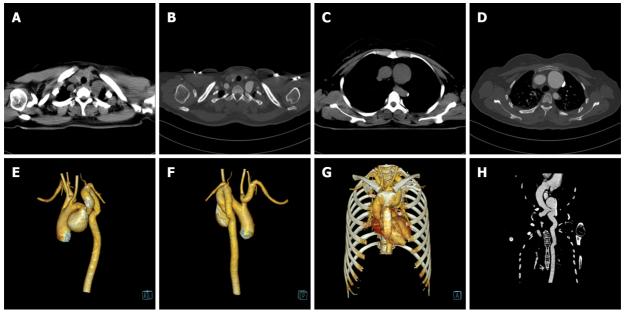
The total hospital stay was 30 d. The patient's recovery was smooth, and no abnormality was found at the 2-mo follow-up after discharge.

#### DISCUSSION

CAA was first reported by Reid[5] in 1914. CAA is very rare, with an incidence of less than 1/10000[1,3,6], although it is relatively more common in young women[6]. In the 35 cases of CAA reported by Zhong *et al*[6], the average patient age was 34.2 years, and females accounted for 65.7% of the sample.

The etiology of CAA is unclear but is possibly related to abnormal embryonic development of the AA (persistence of the 2<sup>nd</sup> or 3<sup>rd</sup> arch or incomplete descension of the normally developed fourth arch, resulting in incomplete entry of the AA into the thoracic cavity)[7-10]. Chromosome 22q11 deletion is evident in some CAA cases[3,4,11].

Most CAA cases are asymptomatic and are usually detected incidentally[4,6,12]. A few cases manifest as a pulsatile neck mass[4,6] or dyspnea and dysphagia due to compression of the trachea and esophagus[6,12,13]. Some CAA cases are combined with congenital cardiovascular anomalies, such as patent ductus arteriosus, tetralogy of Fallot, ventricular septal defect, pulmonary atresia[1,14], and various anatomical variations of the AA branches[2,3,13]. Approximately 20% of CAA cases are complicated with aneurysm formation[1,9,11], which is more common in women[8,9]. The aneurysm may be due to abnormal hemodynamics and arterial wall pressure changes caused by embryonic development, connective tissue anomalies, and a long and tortuous aorta[11,14]. Haughton *et al*[15] classified CAA into five types. Type A: The CAA and DA are contralateral, one side of the common carotid artery is missing, and the independent internal and external carotid arteries directly branch from the AA. Type B: The CAA and DA are contralateral, and two common carotid arteries branch from the AA. Type C: The CAA and DA are contralateral, and a bicarotid trunk appears. Type D: The CAA and DA are ipsilateral, and the sequence of brachiocephalic branching is normal. Type E: This type refers to



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Figure 1 Cervical aortic arch in a 29-year-old female patient. A: Non-contrast-enhanced chest computed tomography (CT) images (axial view) showing that the top of the aortic arch (AA) was at the mid-superior level of the first thoracic vertebra; B: Contrast-enhanced CT images (axial view) showing the same sign as A; C: Non-contrast-enhanced chest CT images (axial view) showing that the largest plane of the aortic aneurysm was roughly the upper plane of the fifth thoracic vertebrae; D: Contrast-enhanced CT images (axial view) showing the same sign as C; E: Volume reconstruction (VR) (left anterior oblique view) showing that the brachiocephalic trunk, left common carotid artery, and left subclavian artery were sequential branches from the proximal end to the distal end of the AA, that the AA showed tumor-like local expansion, and that the AA distal to the aneurysm and the upper descending aorta (DA) were long and tortuous; F: VR (left posterior oblique view) showing that the right vertebral artery (RVA) originated from the right subclavian artery (RSCA), the left vertebral artery (LVA) originated from the top of the AA, the LVA was thinner than the RVA, and the proximal segment of the RSCA was dilated and tortuous; G: VR (anteroposterior view) showing that the top of the AA was located above the clavicle; H: Curved planar reconstruction showing that the AA straddled the left side of the spine and that the AA distal to the aneurysm and the upper segment of the DA were long and tortuous and were located to the left of the spine.

a right CAA with an ipsilateral DA. Haughton et al[15] type D is the most common type of CAA that is combined with aneurysms[3,16], most aneurysms are located in the AA between the LCCA and the LSCA[6,8,16], but a few are located in the DA or simultaneously involve the AA and DA[16]. Shayan et al[11] reported eight cases of CAA with aneurysms (type D aneurysm in seven cases and type A aneurysm in one case), including six cases in which the aneurysm was located between the LCCA and the LSCA and two cases in which the aneurysm was located between the LSCA and the DA.

The patient described in this study had a left CAA with an ipsilateral DA. Her long and tortuous AA straddled the left side of the spine, extended upward above the clavicle, and then extended downward tortuously along the left side of the spine. The concurrent AA aneurysm was located between the LCCA and the ostium of the LSCA, and the origins of the two blood vessels were far apart. At the same time, the CAA was combined with the proximal dilatation and tortuosity of the RSCA and the anomalous origin of the LVA, reflecting the uniqueness of her manifestations.

#### CONCLUSION

A CAA is a rare congenital anomaly of vascular development. Some CAA cases are combined with congenital cardiovascular anomalies, and approximately 20% of CAA cases are complicated with aneurysm formation. The present unique case of CAA with aneurysm formation and an anomalous RSCA and LVA enriches existing CAA data.

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3294

### **FOOTNOTES**

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3295



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3296



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