CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 10 (2015) 32-34



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com



"Dysphagia lusoria" – Right subclavian retroesophageal artery causing intermitent esophageal compression and eventual dysphagia – A case report and literature review*



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ARTICLE INFO

Article history:
Received 21 January 2015
Received in revised form 23 February 2015
Accepted 25 February 2015
Available online 7 March 2015

Keywords:
Aberrant subclavian artery
Dysphagia lusoria
Anatomic variations
Aortic arch
Retroesophageal subclavian artery

ABSTRACT

We report a case of an uncommon anatomical anomaly in which a right subclavian retroesophageal artery (RSRA) was discovered during a routine chest CT scan in a patient with intermittent upper digestive symptoms (occasional dysphagia for solids, the so called "globus hystericus"). Subclavian arteries vary in their origin, course or length. RSRA is a relatively common embryological anomaly of the aortic arch. In this case we report a single carotid trunk. This variation is due to interruption of the fourth right aortic arch between the origins to the common carotid artery and subclavian artery, while the fourth left arch is intact. The regression of the proximal portion of the right subclavian artery occurs and the retroesophageal aortic arch persists, rarely leading to symptoms, as were present in this case.

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1. Introduction

A right subclavian retroesophageal artery (RSRA) is an unusual anomaly. In large necropsy series, the incidence of aberrant right subclavian artery ranges from 0.2% to 2.5% [1]. This is the most common anomaly of the aortic arch, representing 17% of this malformation group. This abnormality is usually silent and in most times is an incidental finding at necropsy. Other embryological development abdormalities may be present, either from the aortic arch complex or from carotid and pulmonary system [1].

The term "disfagia lusoria" was first used by Bayford in 1794 to describe a type of dysphagia secondary to a right subclavian retoesophageal artery (aberrant). He described an elderly woman who died as a consequence of esophageal obstruction and consequent weight loss. This condition, in his own words "may be called lusoria, from lusus naturae (Latin for "freak of nature") that gives rise to it" [2–4].

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2. Case report

We report a case of an incidental CT finding of a RSRA with a single carotid trunk: a 70-years-old female patient with long history (more than 10 years) of upper digestive symptoms (occasional dysphagia for solids, sensation of "globus hyistericus") without presenting weight loss during this period or any other progressive digestive complaint.

This patient had a history of gastroesophageal reflux disease (GERD) for at least 15 years, without pathological repercussion on esophageal mucosa (no esophagitis), managed with proton pump inhibitor. She was, at that moment, asymptomatic from this gastrointestinal disorder. The patient had no other findings from medical history or physical examination besides a cough secondary to the intake of angiotensin enzyme inhibitors, which have motivated the solicitation of a thoracic CT.

No other vascular anomalies were detected throughout the exam. The heart presented normal dimensions in the patient's previous tomographic study and ecocardiographies. The upper digestive endoscopy has not demonstrated signs of extrinsic compression Figs. 1 and 2.

3. Discussion

There are several reports on the literature about the retroesophageal course of the right sublavian artery. The first description was written in 1735 by Hanauld [3]. In 1794, Bayford described

[☆] In this case report we present an interesting case of a right subclavian retroesophageal artery that we have discovered in our institution. The data was collected from our records and from the literature.

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Fig. 1. Digital reconstruction of axial CT: (a) posterior view and (b) anterior view. T – carotid trunk.

SE - left subclavian artery.

ASDR - right subclavian retroesophageal artery.

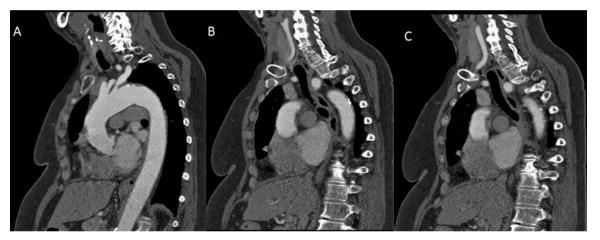


Fig. 2. Digital reconstruction of sagital CT.
(a) Common origin of both carotid arteries in an single trunk.
(b) RSRA compressing entirely the esophagus behind the trachea.

(c) Pervious esophageal lumen above and below the RSRA.

symptoms caused by an aberrant right subclavian artery [3]. Dysphagia lusoria represents a deglutition disorder caused by the positioning of the artery, which compresses the esophagus, and is the most common symptom found in a RSRA [2,3]. In 1946, Gross reported for the first time the surgical correction of this condition in a 4-monts infant, dividing the artery through a left antero-lateral thoracotomy. The first treatment in adults was described in 1963 by Lichter [5].

Literature estimates the presence of an aberrant RSRA in about 0.4% of dissection rooms and in 1.6% of autopsy procedures. It is likely to exist an increased frequency of congenital heart diseases (4.4%), mainly Fallot's tetralogy (12%) [5,6].

Throughout the embryological period, the aortic arches are non-paired arteries which appear in the fourth week of foetal development and they have the function of connecting the aortic sack (anteriorly localized from the pharynx). Normally, six pairs of aortic arches appear and, between the 6th and the 8th week they will be transformed in some of the main vascular structures of the head, neck and thorax [6–8]. The right subclavian artery has its origin in three sites: (I) the fourth aortic arch (forming the artery's proximal portion); (II) the portion from the right dorsal aorta between the

forth aortic arch and the seventh right intersegmental artery; (III) the seventh right intersegmental artery [6].

In this RSRA anomaly the fourth right aortic arch and/or the dorsal right aorta between the fourth arch and the seventh right intersegmental artery disappear [2]. Hence, the proximal portion of the retroesophageal artery is formed by the most caudal portion of the right dorsal aorta (in this case, abnormally persistent, because normally it is degenerated), while the distal portion is formed by the seventh right intersegmental artery [2]. As the aortic arch arises, the differentiated grow modifies the origin of the RSRA and the origin of the left subclavian in cranial direction [6]. The fact that the main portion of the right subclavian artery is derived from a dorsal right aorta portion explains the retroesophageal direction in which the artery follows while it targets the superior limb.

RSRA classification is made through a system described by Adachi about the emission pattern of the aorta branches [4]. There are several morphologic types described for a RSRA. According to Adachi–Williams' classification, the anomalous patterns of the RSRA ramification can be constituted of one of the four basic types represented in Fig. 3.

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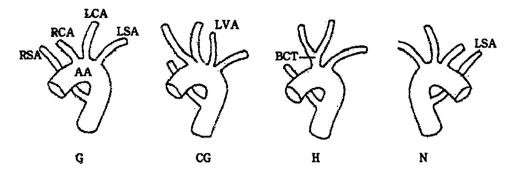


Fig. 3. Main RSRA morphologic types. The classification can be described as below:

1) Type G-1

The right subclavian artery arises from the distal portion of the aortic arch as its last branch. The other main branches (common right and left carotid arteries and the left subclavian artery) follow they normal trend.

2) Type CG-1

The right subclavian artery is anomalous (as in type G) and the left vertebral artery originates itself directly from the aortic arch.

3) Type H-1

The right subclavian artery is anomalous (as in type G), and the common right and left carotid arteries arise from a unic trunk named bicarotidic trunk, as in the presented case. Furthermore, there are other morphological types described and they are rarer. It occurs in about 0.16% of the aortic arch angiographic exams, being in most of the times an incidental finding [7]. It is even rarer when the patient relates symptoms of dysphagia, as the patient presented in our case [7]. 4) Type N-1

This pattern is a mirror image of type G. There is a right aortic arch and the left subclavian artery origin succeeds both corotid arteries and the right subclavia. It is rarer than the RSRA occurrence.

The aberrant artery can cause esophageal and tracheal compression resulting in dysphagia and respiratory problems. These symptoms more frequently occur in children. The appearance of this symptoms or its recurrence throughout adult life may result in atherosclerosis which can make the artery wall more stiff and then compress the esophagus [8,9]. Other symptoms less frequents are the presence of asymmetric pulses of the upper limb, trophic alterations on the right upper limb, thoracic spine erosion, cyanosis, arterial insufficiency and several other vascular anomalies.

To understand the normal aortic arch morphology and its anatomic variants (mainly of the RSRA) is important for the angiographist and the interventional cardiologist who make use of right axillar, brachial or radial approaches to reach the ascending Aorta. The presence of the anomaly ought to be suspected in cases in which the ascending Aorta catheterization is difficult.

Management of this condition should be performed in the presence of complications, usually due to the compression of adjacent structures. Therefore, surgical approach is indicated when a RSRA is symptomatic or has evidence of aneurysm [10].

Conflict of interest

There are no conflicts of interest in this work.

Funding

There are no funding sources in this work.

Ethical approval

This work has been approved by ethics committee.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy

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of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Every author have contributed equally in this work.

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