

Retro-oesophageal right subclavian artery in association with thyroid ima artery: a case report, clinical impact and review of the literature

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A 37-year-old female Caucasian cadaver with an aberrant right subclavian artery extending from the left side of the aortic arch and following a retro-oesophageal course is presented. A non-recurrent right laryngeal nerve and a thyroid ima artery arising from the lower part of the middle third of the right common carotid artery coexisted. The brachiocephalic trunk was absent, while both common carotid arteries and left subclavian artery followed their normal course. The aim of the current study is to highlight the clinical impact of the above abnormalities providing useful and practically applicable knowledge to interventional clinicians, thoracic and neck surgeons, since the vast majority of documented cases with an arteria lusoria are clinically silent and in most cases discovered incidentally. Clinical manifestations such as dysphagia, chronic cough, and acute ischaemia to the right upper limb may occur, leading to misinterpretation in radiographic examination and complications during neck and thoracic surgery. Review of the literature was also performed and the embryological background of the aberration is highlighted. (Folia Morphol 2016; 74, 1: 130–135)

Key words: aortic arch variations, arteria lusoria, non-recurrent laryngeal nerve

INTRODUCTION

Numerous variations in the branching pattern of the human aortic arch exist [39, 41, 50]. “Normal” or “classic” type gives rise to three branches: (a) the brachiocephalic trunk (BT) as the largest branch that gives off the right subclavian artery (RSA) and the right common carotid artery (RCCA), (b) the left common carotid artery (LCCA) and (c) the left subclavian artery (LSA) [23].

This pattern referred to as “type A” according to Adachi [2] is detected in a variable incidence of 64.9–94.3% [4, 50] among several populations, with higher incidences in Japanese (94.3%) [52], Indians (91.4%) [51] and Caucasians (83%) [50]. Following the systematic classification of the variability of the

aortic arch branches [52], an aberrant RSA may occasionally arise next to a normally positioned LSA, as the last branch of the aortic arch, in 0.16–25% of the individuals [9, 50]. Hunald first described the aberrant vessel on a necropsy in 1735 and Bayford coined the term ‘dysphagia lusoria’ in 1761, after discovering a retro-oesophageal RSA — RERSA (arteria lusoria) compressing the oesophagus in a 62-year-old woman who suffered from swallowing difficulty and eventually died of emaciation [5]. The BT absence and the arteria lusoria presence are the findings used to identify the non-recurrent right laryngeal nerve (NRLN) [70]. Occasionally, a thin anomalous feeding vessel, the thyroid ima artery (IMA) or artery of Neu-

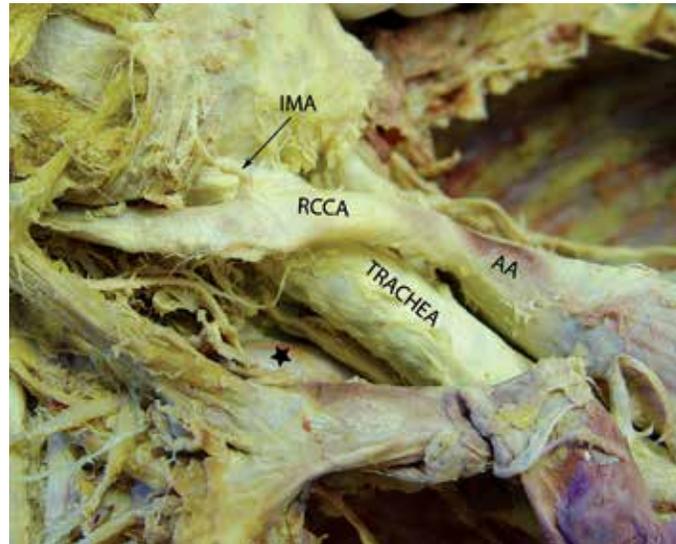


Figure 1. View of the aortic arch and its branches showing the retro-oesophageal right subclavian artery (asterisk); AA — aortic arch; RCCA — right common carotid artery; IMA — thyroid ima artery.

bauer ascending to the thyroid gland is detected in 1.5–12.2% according to Faller and Schärer (1947) as cited by Krudy et al. [37]. This inconstant artery, usually detected on the right side, may be given off from the BT (0.5–3%) [18, 43], the RCCA (0.5–1%) [41, 43], the aortic arch (0.1–2.22%) [35, 43], the internal thoracic artery, the pericardiophrenic artery, the subclavian artery, the thyrocervical trunk, the inferior thyroid artery or the transverse scapular artery. An IMA replacing the left inferior thyroid artery [42] or an IMA arising from the BT associated with bilateral absence of the inferior thyroid arteries were detected [73]. In both cases the right and left recurrent laryngeal nerves (RLNs) had a typical course. Locating IMA is of paramount importance during neck surgery due to the high risk of iatrogenic injury to the artery and during parathyroid arteriography when detecting adenomas or other gland lesions [37]. In the present study we report a rare case of a RERSA associated with an IMA arising from the RCCA and a right NRLN. Moreover, the embryological background of the coexistent variations is highlighted.

CASE REPORT

During a routine dissection of a 37-year-old formalin embalmed female Caucasian cadaver at the Department of Anatomy (Medical School of the Aristotle University of Thessaloniki), a very rare aberration of the aortic arch was detected. After the careful dissection of the anterior part of the neck and the thoracic

cavity, 4 branches of the aortic arch were identified (proximal to distal): the RCCA giving rise to an IMA from the lower part of its middle third (below the CCA bifurcation), the LCCA, the LSCA and a RERSA as the last branch (Figs. 1, 2). Consequently no BT was observed. The IMA after its emergence, passed inwards and directly upwards, supplying the right lobe and the lower part of the isthmus of the thyroid gland. The vessel had 1 bifurcation and multiple terminal branches. The RERSA coursed horizontally between the oesophagus and the vertebral column, on its way to the right upper limb. The perimeter of the RERSA diminished gradually as the artery coursed towards the right side of the neck. The right recurrent laryngeal nerve branched directly from the vagus nerve, while the contralateral nerve (left RLN) appeared to loop around the aortic arch normally. The lumen of all great vessels was free of any aneurysmal dilatation. Except for the RERSA, both CCAs and LSA followed their typical course to the neck, adopting their usual origin and distribution pattern. After a detailed investigation of the cadaver's medical records, no relevant clinical manifestations, such as right upper extremity ischaemia or dysphagia lusoria were retrieved. The thoracic duct followed the classic course and drained normally into the jugular-subclavian junction. Trachea and oesophagus had normal structure and position. No malformations or anomalies were observed in the rest of the thoracic and abdominal organs. The cause of death was unrelated to the encountered findings.

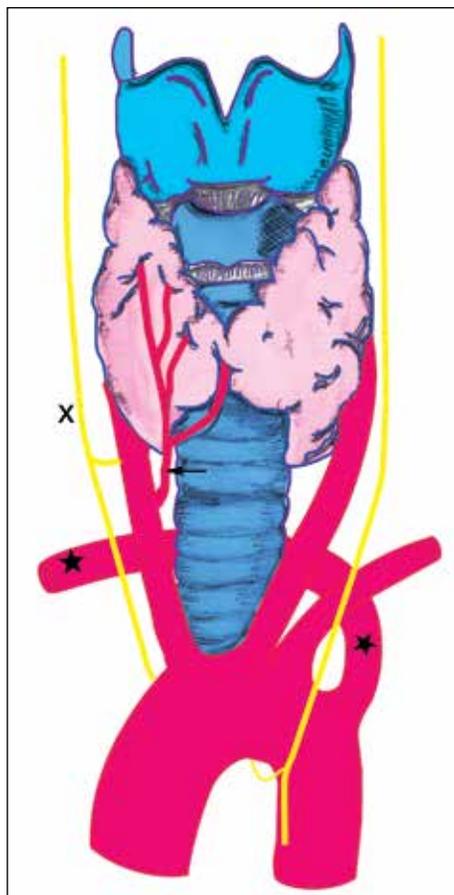


Figure 2. A scheme presenting the retro-oesophageal right subclavian artery (asterisk) and the thyroid ima artery (arrow). All branches of the aortic arch are demonstrated; X — vagus nerve.

DISCUSSION

A plethora of classifications of the human aortic arch branches has been reported. Most likely these variations appear due to altered development of certain arteries of the branchial arch [7, 68]. Formation of an aberrant RSA is the result of the 4th vascular arch degeneration along with the dorsal aorta, while the 7th intersegmental artery remains attached to the descending aorta [20]. The persistent intersegmental artery will eventually become the RERSA or arteria lusoria in 80–84% of the cases, or the aberrant RSA passes behind the trachea in 12.7–15% or in front of the trachea in 4.2–5% [5, 18]. The Kommerell's aortic diverticulum, a remnant of the distal right aortic arch may be detected at the RSA origin. As regards the morphometric details, the diameter of the RERSA is usually greater at the proximal than the distal part [20], while its wall is thinner than that of normal arteries of the same diameter [3].

The occurrence of the RERSA varies widely between 0.8% and 37% among various populations, the higher figure corresponds to the incidence in Down syndrome patients with congenital heart disease [9, 29, 41, 58, 70]. The higher reported incidence with a wide range of 2.46–9.7% were found in African Americans by Bean (1905) and Loth (1912) as cited by De Garis et al. [21], followed by Germans (4%) [55], Japanese (3.95%) [48] and South Africans (1.4%) [17]. Contrariwise, a very low incidence (0.30%) was detected in a European population [1]. The arteria lusoria is associated with other congenital anomalies, such as the formation of a bicarotid trunk (BCT), an aortic coarctation, a replaced right or left vertebral artery, a persistent left superior vena cava [72], a ventricular septal defect, the tetralogy of Fallot [44], pulmonary atresia and major aortico-pulmonary collateral arteries [40]. Reinshagen et al. [55] emphasized that the arteria lusoria was present in 14.8% of patients with BCT, while patients with classic anatomy of the neck vessels presented the artery in 2.1%. Natsis et al. [49] highlighted that the coexistence of arteria lusoria and BCT appears in up to 2.78% in Caucasians. Moreover, higher incidence of arteria lusoria has been reported in patients with Down syndrome (12–37%) [16, 29, 57, 74], Edwards and DiGeorge syndrome and congenital heart disease [45, 63]. The artery may also coexist with long gap oesophageal atresia and trachea-oesophageal fistula in neonates [13].

Abnormal aortic arch development is related with variation in the course of the RLN [17]. Arteria lusoria is usually associated with right NRLN, due to the fact that during foetal development, the vessel fails to drag the nerve caudally when the heart descends and neck elongates [3, 22]. Simmons et al. [61] and Tateda et al. [65] reported a RERSA associated with a right NRLN. Tartaglia et al. [64] and Wang et al. [69] found an arteria lusoria in coexistence with a right NRLN in 0.4% of their sample and Toniato et al. [67] in 0.5%. De Garis et al. [21] mentioned the presence of a right NRLN in all cases of RERSA, except of one specimen where the RLN followed a recurrent course under the inferior thyroid artery. The right NRLN appears in 0.3–1.6% [66], while the left NRLN is extremely rare with an incidence of up to 0.04% [32]. The atypical course of the recurrent laryngeal nerve, usually on the right side is clinically important during thyroidectomy and parathyroidectomy, where the nerve cannot be identified at the lower pole of the thyroid gland and may be injured during the procedure [67]. Hence, the

Table 1. Literature overview pertaining to a thyroid ima artery (IMA) in combination with a retro-oesophageal right subclavian artery

Study	Year	Gender	Sample	Race/ /Material	IMA/Origin	RSA Course	RLN
Hammer and Meis [31]	1941	Female	Case report	African American/ /Cadaver	Thymica/LCCA	Retro- -oesophageal	NRLN
Hammer and Meis [31]*	1941	Female	Case report	Caucasian/ /Cadaver	IMA/RCCA	Retro- -oesophageal	RLN under IMA
Simmons et al. [61]**	1987	Female	Case report	Not mentioned/ /Angiography	IMA/RCCA	Retro- -oesophageal	NRLN
Loukas et al. [42]	2006	Male	Case report	Not mentioned/ /Cadaver	IMA/Common trunk with the RVA	Retrotracheal	RLN
Present case	2015	Female	Case report	Caucasian/ /Cadaver	IMA/RCCA	Retro- -oesophageal	NRLN

*IMA is mentioned by authors as inferior thyroid artery; **According to Pratt's recognition of IMA [54], authors incorrectly describe their finding as being an inferior thyroid artery instead of an IMA; LCCA — left common carotid artery; RCCA — right common carotid artery; NRLN — non recurrent laryngeal nerve; RLN — recurrent laryngeal nerve; RVA — right vertebral artery

NRLN preservation is extremely challenging, even to experienced hands. To avoid nerve injury, a preoperative identification with computed tomography scan of the neck area and the intraoperative neuromonitoring has commonly been applied [28].

IMA supplies the thyroid gland when the superior and/or the inferior thyroid arteries are absent (3–6%) or mal-developed or when the blood flow increases due to pathological conditions. The artery has a small calibre when passes to thymus (thymica accessoria) and although the gland is almost completely atrophied, total degeneration never occurs [54]. Sporadic reports exist as regards the coexistence of arteria lusoria and IMA with a variable course of the recurrent laryngeal nerve (Table 1).

Most documented cases of RERSA are clinically silent, due to the fact that the vessel does not form a complete vascular ring around oesophagus and trachea [14]. However when clinical manifestations occur, cardiac symptoms or silent ischaemia may appear [1] with prominent symptoms the progressive dysphagia and dyspnoea, particularly when the RERSA wall is calcified and the advanced atherosclerosis and hardening of the vessel compressing the oesophagus [34]. Persistent cough may be the main symptom due to the pulsatile action of the aberrant artery on the tracheal wall [57] or the compression exerted by an existing aneurysm on the adjacent structures. Dyspnoea, stridor, feeding difficulty, recurrent respiratory infections are the main symptoms in childhood, due to the vulnerable trachea, while dysphagia is the primary symptom in adults [27, 53, 59, 63]. Clinicians should not forget that in paediatric population, serious respiratory discomfort may appear without

dysphagia [36]. Feeding or swallowing difficulties in patients with Down syndrome should immediately raise the suspicion of a RERSA, although only 10–20% of them will develop dysphagia [56]. Patients with undetected arteria lusoria and prolonged nasogastric intubation may develop spontaneous oesophageal fistulisation, after the pulsatile arterial compression on the oesophageal wall. A limited necrosis may progressively appear on the lumen of both structures, in association with the rigid intubation catheter. Fistula formation is a rare fatal event. Since trans-oesophageal ultrasound is usually diagnostic, the long-term placement of nasogastric tube should precede in intensive care patients [24–26]. Other manifestations like aneurysm formation, acute ischaemia of the right upper limb may occasionally develop [10, 47]. The RERSA is an incidental finding on imaging studies [20] that needs further examination based on plain chest roentgenogram [11, 71] and oesophagography findings [14]. Once the anomaly is diagnosed, the precise vascular anatomy and other associated cardiac defects are investigated with echocardiogram or trans-oesophageal echocardiography [46, 71]. Contrast enhanced computed tomography scan, magnetic resonance imaging or conventional angiography will provide further information about the type and extent of lesion, which is extremely helpful in preoperative planning [8, 30, 47, 59]. In cases of RERSA, the barium swallow of the oesophagus is a very helpful diagnostic tool, which will reveal the typical impression defect, while endoscopically a pulsatile mass may be discovered [33]. Treatment is indicated especially in cases of paediatric dysphagia. Surgical repair is mandatory in aneurysm cases [59]

in order to avoid further serious or even fatal complications [12]. A combined endovascular and surgical treatment was reported [38], while minimally invasive surgical and endovascular techniques have also been suggested as repair methods [6, 17]. Davidian et al. [19] accomplished the first successful treatment of an RERSA aneurysm using a polytetrafluoroethylene covered stent graft. Shennib and Diethrich [60] introduced innovative hybrid approaches, but long term results are not yet available. New endovascular techniques without thoracotomy are very promising, especially in immunocompromised patients [62]. Head and neck surgeons should always keep in mind all possible vascular anomalies in the region, especially when performing tracheostomy, employing either the open or the closed percutaneous dilatation. An aberrant RSA may be encountered unexpectedly during the procedure [15].

CONCLUSIONS

Patients with vascular anomalies are at higher risk of iatrogenic injuries intraoperatively. Head and neck and thoracic surgeons, interventional radiologists and cardiologists should always be aware of all possible variations of the aortic arch in order to achieve the best possible and uneventful surgical outcome when dealing with thyroid gland diseases. General practitioners, paediatricians, gastroenterologists, should always keep in mind the possibility of a vascular anomaly when a patient is recurrently diagnosed with wheezing, upper respiratory tract infections, cough and failure to thrive.

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