VIA MEDICA

CASE REPORT

Folia Morphol.
Vol. 65, No. 3, pp. 236–241

ISSN 0015–5659
www.fm.viamedica.pl

A retrotracheal right subclavian artery in association with a vertebral artery and thyroidea ima

M. Loukas[1, 2], R.G. Louis Jr.[3], J. Gaspard[3], M. Fudalej[4], R.S. Tubbs[5], W. Merbs[1]

[1]Department of Anatomical Sciences, St George's University, School of Medicine, Grenada, West Indies
[2]Department of Education and Development, Harvard Medical School, Boston, MA, USA
[3]Department of Anatomy, American University of the Caribbean, Sint Maarten, Netherlands Antilles
[4]Department of Forensic Medicine, Warsaw Medical University, Warsaw, Poland,
[5]Department of Cell Biology, University of Alabama at Birmingham and Children's Hospital, Birmingham, Alabama, USA

[Received 1 December 2005; Revised 25 April 2006; Accepted 25 April 2006]

The retro-oesophageal right subclavian artery is an anatomical abnormality encountered by anatomists and pathologists and, more recently, interventional cardiologists and thoracic surgeons with an incidence of 0.2–2% in the population. We report a case of a retrotracheal right subclavian artery which originated distally along the left aortic arch and coursed between the trachea and the oesophagus. Additionally, the aortic arch gave rise to a common trunk, which subsequently bifurcated to yield to a right vertebral artery and a left thyroidea ima, replacing the left inferior thyroid artery. Consequently the right and the left recurrent laryngeal nerves were found to recur normally. The possible embryonic development of these branching patterns and their clinical significance is discussed.

Key words: retro-oesophageal right subclavian artery, retrotracheal course, aortic arch, coronary angiography, recurrent laryngeal nerve, vertebral artery, thyroidea ima

INTRODUCTION

With the increasing use of imaging studies, anomalies of the aortic arch have been identified more frequently [6]. However, the clinician should be aware of their existence to better appreciate the wide range of anomalies that occur in the arch and the great vessels. This could help in adequately managing these variations in emergency approaches to the arch and the great vessels when imaging studies are not available [10].

In approximately 80% of individuals, three branches arise from the aortic arch: the brachiocephalic trunk, the left common carotid artery and the left subclavian artery [9]. Adachi first described this branching pattern as type A [2]. Another 11% of reported cases exhibit Adachi's pattern type B, which consists of a common trunk for the left common carotid artery and the subclavian artery. This branching pattern results in only two trunks originating from the aortic arch. The third most common pattern, type C, is characterised by the vertebral artery, originating proximally to the left subclavian artery as a fourth branch of the aortic arch. The remaining 1% of cases are composed of numerous other aortic arch branching pattern variations [23].

Thomson was able to identify nine different variations in the mode of origin of the branches arising from the aortic arch in 500 specimens [32]. In five of the specimens (1%) he was able to identify a retro-oesophageal right subclavian artery (RRSA) and

Address for correspondence: Dr. M. Loukas, MD, PhD, Assoc. Prof., Department of Anatomical Sciences, St George's University, School of Medicine, Grenada, West Indies, tel: 473 444 4175 x 2014, fax: 473 444 2887, e-mail: edsg2000@yahoo.com, mloukas@sgu.edu

classified it as type D. Similar important contributions to the classification and description of the branching pattern of the aorta have been made by Quain [28] and Henle [17]. According to these authors, the RRSA was reported from 1791 until 1868 as occurring in 13 individuals. From 1868 until recently more than 100 cases of RRSA have been described.

In this report we describe an unusual case in which a left aortic arch gave rise to a right retrotracheal subclavian artery (RtRSA) and a common trunk, which subsequently bifurcated to yield to a right vertebral artery and a left thyroidea ima, replacing the left inferior thyroid artery.

CASE REPORT

During a routine medicolegal autopsy of a 12 year-old male at the Department of Forensic Service at Warsaw Medical University a unique branching pattern of the aortic arch was discovered. The young male had died from brain injuries sustained during a motor vehicle accident. While no obvious gross pathological changes or traumatic rupture of the vessels involved was evident, it is unknown whether the anomaly was a contributory cause of death. During the examination a RtRSA anomaly was recognised and the aorta (along with the proximal parts of its branches and its associated thoracic viscera) was removed *en bloc* from the body. The specimen was then cleaned, dissected to expose silent features and photographed. After careful examination the examiners returned the specimen to the body. The young male did not possess any abnormalities apart from the anomalous right subclavian artery, which originated from the descending aortic arch, and the common trunk giving rise to a right vertebral artery and a thyroidea ima. The aortic arch appeared unremarkable. The branches of the aortic arch (proximal to distal) were as follows: right common carotid artery, left common carotid artery, common trunk with a right vertebral artery and to the left a thyroidea ima, left subclavian artery and RtRSA. The RtRSA crossed between the oesophagus and the trachea posteriorly to reach the right upper limb (Figs. 1, 2). Consequently, the brachiocephalic trunk was absent. Additionally, the common trunk, which arose between the left common carotid and the left subclavian artery, subsequently bifurcated to yield the right vertebral artery and the left thyroidea ima. The left vertebral artery arose normally. The thyroidea ima appeared to be replacing the left inferior thyroid artery.

There were no noticeable abnormalities in the heart or the remaining thoracic organs. The trachea and oesophagus were positioned normally. All abdominal viscera were normally located without any malformation or disease. Both right and left recurrent laryngeal nerves looped typically around the right subclavian artery and aortic arch, respectively.

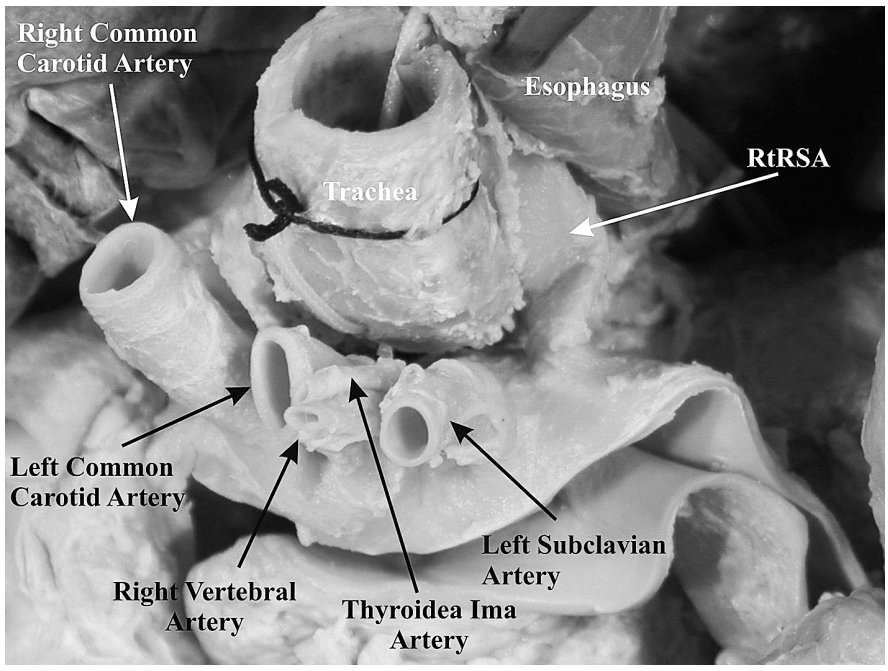

Figure 1. This figure shows a left aortic arch giving rise to a common trunk, in which a right vertebral artery and a left thyroidea ima arise as well as a retrotracheal right subclavian artery.

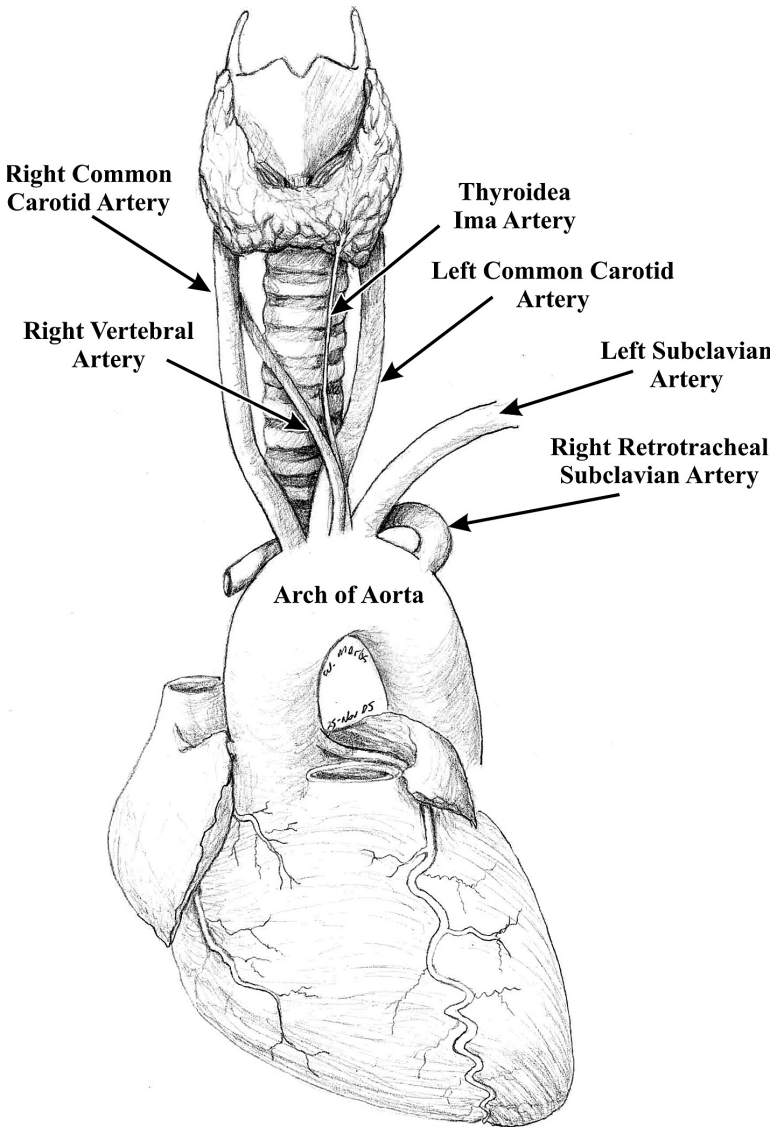

Figure 2. This scheme demonstrates the retrotracheal right subclavian artery arising distally from the aortic arch. In addition the scheme demonstrates all the additional branches such as the right vertebral artery and a thyroidea ima, replacing the left inferior thyroid artery.

DISCUSSION

The origin of the RRSA as the last branch of the aortic arch is a common congenital aortic arch anomaly with a reported prevalence of 0.4–2% [18, 20]. Of these cases 15% exhibit a retrotracheal course [21], the remaining specimens resulting in the retro-oesophageal variant.

It is important to make a clear distinction between RRSA as reported by Klinkhamer [18] and Léger [20] and RtRSA as described here. In the excellent review paper of Nizanowski et al. [24] 25 types of abnormal origin from the arteries of the aortic arch are described, in ten of which a RRSA is exhibited. However, we were unable to find a similar case to ours according to Nizanowski's classification [24].To the best of our knowledge this is a unique case in which a left aortic arch gave rise not only to an RtRSA but also to a common trunk from which a vertebral artery and a thyroidea ima arose.

Five paired arches develop in the first five months of embryonic life. Several segments degenerate, leading to the normally branching left aortic arch. These correspond to: the distal portion of the right dorsal aortic root, the first and second aortic arches on both sides, both dorsal aortae between the third and fourth aortic arches, resulting in the common carotid arteries, the distal right sixth arch and the upper dorsal intersegmental arteries with the exception of number seven on both sides [7]. There are even variations within the "normal" aortic arch. The vertebral artery

may originate, as in our case, from the aortic arch. Normally the vertebral arteries originate from an artery paralleling the dorsal aorta and connected by multiple intersegmental arteries. The origin of the vertebral arteries from the aortic arch is explained by degeneration of the normal origin from the seventh intersegmental artery with persistence of a higher intersegmental artery, such as the sixth [12]. The developmental RtRSA anomaly occurs when degeneration of the fourth vascular arch, along with the dorsal aorta, leaves the seventh intersegmental artery attached to the descending aorta [1]. This persistent seventh intersegmental artery assumes a retro-oesophageal or retrotracheal position as it proceeds out of the thorax into the right arm and becomes RRSA or RtRSA in the adult. Alternatively, the descent of the cranial part of the trachea and the ascent of the caudal part may fuse anteriorly to the subclavian artery, forming an RtRSA [21, 22].

The embryological basis of the development of a thyroidea ima still remains largely unknown. A recent report by Vasovic et al. [33] claims that the thyroidea ima artery probably represents an example of the arterial self-differentiation and induced differentiation of the arteries of the aortic arch. However, we agree with the theory proposed by Robinson that the thyroidea ima artery is the principal embryonic blood source to the third and fourth pharyngeal pouches [29]. As a result, anomalous or late morphogenesis of the third and fourth pharyngeal pouches may favour the persistence of a thyroidea ima artery to adulthood [29].

Interestingly, cases of RtRSA may arise in *situs inversus* and a double aortic arch. Apart from these cases, Bialowas et al. [8] described four cases of rare aortic arch anomalies in a study including 1700 cadavers. These included two cases of a left-sided aortic arch with aberrant right subclavian artery, a case of a left-sided aortic arch with a retro-oesophageal course and a right side descending aorta and, finally, a case of a right-sided aortic arch with a retro-oesophageal bulge and an aberrant subclavian artery. Despite the fact that none of these cases were identical with ours, all of them share the same clinical symptoms.

There are no reports in the literature describing the precise clinical symptomatology of an RtRSA. We suggest that owing to the specific location of RtRSA posterior to the trachea, it may result more often in dyspneic conditions as opposed to dysphagia, which is more common with RRSA. However, we cannot exclude the possibility of a patient with RtRSA expressing the same symptoms as a patient with RRSA. These include dysphagia, and the condition of an RRSA or RtRSA is thus termed "dysphagia lusoria". RRSA or RtRSA compress the oesophagus, causing painful swallowing. Usually symptoms are intermittent and do not need any specific treatment. Surgery is reserved for patients with severe and progressive symptoms. In addition, a vascular ring formation known as Kommerell's diverticulum can present with significant tracheo-oesophageal compression [4, 5]. In elderly patients the RtRSA could be become tortuous and ectatic, resulting in oesophageal or tracheal compression, for which surgery is indicated if the symptoms are severe [27].

The inferior right recurrent laryngeal nerve is an asymptomatic variation or anomaly, which can be an important obstacle and may be seriously damaged during cervicotomy, thyroid and parathyroid surgery. In such cases the inferior right recurrent laryngeal nerve is a classic risk and must be eliminated by location and routine dissection of the nerve [11]. The importance of the diagnosis of a RRSA and/or inferior right recurrent laryngeal nerve differs according to whether the patient is a child or an adult. This is of particular importance when the diagnosis concerns an asymptomatic neural anomaly [11, 22] discovered by dissection or a vascular anomaly whose symptoms are quite variable [11, 31].

According to Hara et al. [16] and Proto et al. [26], the RtRSA course was present in 43–44% of the arteries examined with CT and radiographs. This course is able to create a posterior tracheal imprint which appears as a vascular retrotracheal opacity. As a result of this course Hara et al. [16] suggest that the blood vessels themselves and the oesophagus lying between them have been compressed anteriorly and cause constant displacement of the trachea. Barium oesophagography, angiography and MR angiography are reported to be useful for confirming the diagnosis [3]. An interesting report by Parker et al. revealed four cases of symptomatic vascular rings, including one case of RtRSA, causing asthma [25]. The asthma could possibly be due to anterior displacement of the trachea from RtRSA.

RtRSA is also clinically important to the angiographer who uses the right axillary, brachial or radial approach to the ascending thoracic aorta [1, 13]. The presence of an RtRSA could be suspected in cases in which catheterisation of the ascending aorta proves difficult. Using the right radial approach, access to the ascending aorta is usually easy, as the brachiocephalic trunk is the first branch of the aortic arch

permitting direct access to the ascending aorta. As a result, angiography in the presence of RtRSA could be very challenging [11].

The incidence of an anomalous course of the vertebral artery is low [15]; nevertheless, failure to recognise it promptly may result in iatrogenic injury during surgical procedures of the neck. Four recent detailed reports of abnormal origin of the vertebral artery associated with variations of the aortic arch are described in the literature. Gluncic and Marusic [15] described a left vertebral artery arising from a common trunk of the aortic arch that also gave rise to the left subclavian artery [15]. Best and Bumpers [10] described an angiogram finding of a RRSA and a right vertebral artery originating from the right carotid artery. Fazan et al. [14] reported a case of a right vertebral artery originating from the right common carotid artery with the addition of a RRSA. Koenigsberg et al. [19] described three cases of right vertebral artery abnormalities: the vertebral artery arising as the fourth and most distal branch of the aortic arch, as a distal to the right thyrocervical trunk and as a branch of the right common carotid artery in addition to a RRSA. All the aforementioned cases present similarities with our case. However, our case identified a thyroidea ima artery which none of the previously mentioned studies identified.

With regard to the common trunk with a vertebral artery and a thyroidea ima arising from the aortic arch, the clinical consequences are also unknown. One possible problem could arise in the case of aortic dissection. During aortic arch dissection the orifices of the common carotid arteries may often become occluded, drastically decreasing cerebral blood flow. In the present case the ostium of the vertebral artery may also have become occluded, thus leading to a decrease in blood flow to the brain.

Yilmaz et al. [34] reported a case of a thyroidea ima arising from the brachiocephalic trunk and the left vertebral artery from the aortic arch. Similarly, Schwarzacher and Krammer [30] described a case in which the aortic arch gave rise to five primary branches: the brachiocephalic trunk, giving rise to a thyroidea ima, the left common carotid artery, the left vertebral artery, the right subclavian artery and the right vertebral artery.

In the field of clinical anatomy it continues to remain important to report unusual or rarely seen anatomical variations. By constantly adding to our wealth of knowledge, physicians and anatomists can ensure forward progress and, ultimately, better clinical outcomes for our patients.

CONCLUSIONS

Fortunately, most variations of the aortic arch and great vessels are not a reason for patient complaints. However, in order to decrease the risk of iatrogenic injury, clinicians dealing with the arch and the great vessels should be aware of this variation, as more imaging studies and catheter-based techniques are performed every year. Furthermore, knowledge of the embryology of the cardiovascular system could be a useful tool for understanding the pathophysiology of the variation and recognising it better.

REFERENCES

1. Abhaichand RK, Louvard Y, Gobeil JF, Loubeyre C, Lefevre T, Morice MC (2001) The problem of arteria lusoria in radial transradial coronary angiography and angioplasty. Catheter Cardiovasc Interv, 54: 196–201.
2. Adachi B (1928) Das Arteriensystem der Japaner. 1st ed. Vol. 1. Verlag der Kaiserlich-Japanischen Universitat, Kenyusha Press, Kyoto pp. 29–41.
3. Avisse C, Marcus C, Delattre JF, Marcus C, Cailliez--Tomasi JP, Palot JP, Ladam-Marcus V, Menanteau B, Flament JB (1998) Right nonrecurrent inferior laryngeal nerve and arteria lusoria: the diagnostic and theurapeutic implications of an anatomic anomaly. Review of 17 cases. Surg Radiol Anat, 20: 227–232.
4. Aubertin JM, Iseni MC, Tordjman T, Bloch F, Petite JP (1995) Dysphagia lusoria. Discussion physiopathologique a propos d'un cas clinique. Gastroenterol Clin Biol, 19: 629–632.
5. Balaji MR, Ona FV, Cheeran D, Paul G, Nanda N (1982) Dysphagia lusoria: a case report and review of diagnosis and treatment in adults. Am J Gastroenterol, 77: 899–901.
6. Baron RL, Guitierrez FR, Sagel SS, Levit RG (1981) CT of anomalies of the mediastinal vessels. Am J Roentgenol, 137: 571–576.
7. Bary A (1951) The aortic arch derivations in the human adult Anat Rec, 111: 221–238.
8. Bialowas J, Hreczecha J, Grzybiak M (2000) Right-sided aortic arch. Folia Morphol, 59: 211–216.
9. Bergman RA, Thompson SA, Afifi AK, Saadeh (1988) Compendium of human anatomic variation. Text, Atlas and World Literature. Bergman, Urban & Schwarzenberg, Munchen, Wien, Baltimore, pp. 61–63, 68–70.
10. Best IM, Bumpers HL (2002) Anomalous origins of the right vertebral subclavian and common carotid arteries in a patient with a four vessel aortic arch. Ann Vasc Surg, 16: 231–234.
11. Bogliolo G, Ferrara M, Pietropaolo V, Pizzicannella G, Miscusi G (1982) Dysphagia lusoria: proposal of a new treatment. Surg Endos, 1: 225–227.
12. Cairney (1925) The anomalous right subclavian artery considered in the light of recent findings in arterial development; with a note on two cases of an unusual relation of the innominate artery to the trachea. J Anat, 59: 265–296.

13. Campeau L (1999) Percutaneous radial approach for coronary angiography. Cathet Cardiovasc Diagn, 16: 3–7.
14. Fazan VPS, Caetano AG, Filho OAR (2004) Anomalous origin and cervical course of the vertebral artery in the presence of a retroesophageal right subclavian artery. Clin Anat, 17: 354–357.
15. Gluncic V, Marusic A (2000) Association of the truncus bicaroticus, common trunk of the left subclavian and vertebral arteries, and retro-esophageal right subclavian artery. Case report. Ann Anat, 182: 281–283.
16. Hara M, Satake M, Itoh M, Ogino H, Shiraki N, Taniguschi H, Shibamoto Y (2003) Radiographic findings of aberrant right subclavian artery initially depicted on CT. Radiat Med, 21: 161–165.
17. Henle J (1868) Handbuch der Gefaesslehre des Menschen. In: Henle J (ed.) Handbuch der systematischen Anatomie des Menschen. Braunschweig, Vieweg and Son, pp. 247.
18. Klinkhamer AC (1966) Aberrant right subclavian artery. Am J Roentgenol, 54: 438–446.
19. Koenigsberg RA, Pereira L, Nair B, McCorming D, Schwartzman R (2003) Unusual vertebral artery origins: Examples and related pathology. Cathet Cardiovasc Intervent, 59: 244–250.
20. Léger L (1971) Repérage du nerf récurrent droit. Presse Med, 79: 2383–2384.
21. Lippert H, Pabst R (1985) Arterial Variations in Man. JF Bergmann, Munich, pp. 6–7.
22. Loukas M, Giannikopoulos P, Fudalej M, Dimopoulos C, Wagner T (2004) A retroesophageal right subclavian artery originating from the left aortic arch — a case report and review of the literature. Folia Morphol, 63: 141–145.
23. McDonald JJ, Anson BJ (1940) Variations in the origin of arteries derived from the aortic arch, in American whites and negroes. Am J Phys Anthrop, 27: 91–107.
24. Nizanowski C, Rajchel Z, Ziolkowski M (1975) Abnormal origin of arteries from the aortic arch in man. Folia Morphol, 34: 109–116.
25. Parker JM, Cary-Freitas B, Berg BW (2000) Symptomatic vascular rings in adulthood: an uncommon mimic of asthma. J Asthma, 37: 275.
26. Proto AV, Cuthbert NW, Raider L (1987) Aberrant right subclavian artery: further observations Am J Roentgenol, 148: 253–257.
27. Proye C, Dumont HG, Depadt G, Lagache G (1982) Le nerf récurrent non récurrent danger en chirurgie thyroidienne. Ann Chir, 36: 454–458.
28. Quain R (1844) The anatomy of the arteries of the human body. Taylor and Walton, London, pp. 152–155.
29. Robinson HB JR (1975) DiGeorge's or the III-IV pharyngeal pouch syndrome: pathology and a theory of pathogenesis. Perspect Pediatric Pathol, 2: 173–206.
30. Schwarzacher SW, Krammer EB (1989) Complex anomalies of the human aortic arch system: unique case with both vertebral arteries as additional branches of the aortic arch. Anat Rec, 225: 246–250.
31. Skandalakis JE, Gray SW (1994) Embryology for surgeons, the embryological basis for the treatment of congenital anomalies, 2nd ed. Williams & Wilkins, Baltimore, pp. 976–1018.
32. Thomson A (1893) Third Annual report of the committee of selective investigation of the Anatomical Society of Great Britain and Ireland for the year 1891––1892. J Anat Physiol, 27: 183–194.
33. Vasovic L, Arsic S, Vlajkovic S, Zdravkovic D (2004) Morphological aspect of the thyroid ima artery in human fetuses. Ital J Anat Embryol, 109: 189–197.
34. Yilmaz E, Celik HH, Durgun B, Atasever A, Ilgi S (1993) Arteria thyroidea ima arising from the brachiocephalic trunk with bilateral absence of inferior thyroid arteries: a case report. Surg Radiol Anat, 15: 197–199.