Case report

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DYSPHAGIA LUSORIA: A CASE REPORT OF A PATIENT WITH ABERRANT RIGHT SUBCLAVIAN ARTERY AND TRUNCUS BICAROTICUS

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Aberrant right subclavian artery (ARSA) is a rare vascular anomaly. It is even more rarely associated with dysphagia and common carotid artery trunk—truncus bicaroticus (TB). Bayford described ARSA in 1794 and called it arteria lusoria, and this type of dysphagia—dysphagia lusoria. Infundibular extension to deviate ARSA was first described by Kommerell in 1936, therefore, it is called a Kommerell's diverticulum.

A 69-year-old patient, MD, suffered from dysphagia and hoarseness, showing a weight loss of 9.5 kg, two months before the examination. Esophagoscopy revealed external compression of the posterior wall of the middle third of the esophagus. MSCT examination of the chest, as the most reliable non-invasive diagnostic tool, showed ARSA and Kommerell diverticulum with TB.

The presence of ARSA is usually asymptomatic and accidentally located in examinations and autopsies. Dysphagia with a loss of body weight, if present, originate from the outer compression of ARSA to esophagus, while hoarseness occurs within Ortner cardiovocal syndrome. The patient was advised to undergo surgical treatment in case of deterioration of dysphagic disorder, and until then more frequent food consumption during the day, in small bites, and the use of proton pump inhibitor.

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Key words: *dysphagia lusoria, arteria lusoria, aberrant right subclavian artery, Kommerell's diverticulum*

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Introduction

Aberrant right subclavian artery (ARSA) is a rare vascular anomaly (0.2–2.5% of people) (1–4). Even more rarely is it associated with dysphagia and a common carotid arterial trunk that deviates from the aortic arch—truncus bicaroticus (TB). Bayford described it in 1794 and named it arteria lusoria, and this type of dysphagia, dysphagia lusoria (5). Infundibular extension in the deviation ARSA was first described in 1936 by Kommerell, and consequently it was named Kommerell's diverticulum (KD) (6).

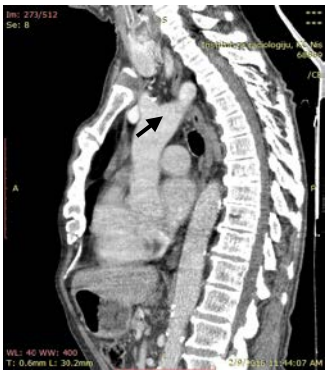
Usually, the right subclavian artery (SA) stems from the truncus brahiocephalicus while ARSA stems directly from the aortic arch behind deviated left SA. Truncus bicaroticus accopmanied with ARSA is present even more rarely (7). In most cases, ARSA is asympotomatic but there may occur dysphagia, dyspnea, stridor, hoarseness, loss of body weight and pain in the chest (7–9). In the presence of ARSA, certain problems may occur requiring cannulation of the right subclavian artery in the interventional cardiology and cardiovascular surgeries. Unrecognized ARSA represents the risk of major bleeding during transhiatal esophagectomy and operations on the aortic arch and the desceding aorta.

Case Presentation

A 69-year-old patient, MD, complained of difficulty swallowing solid food and hoarseness for a month, or a month and a half. During that period he lost around 9.5 kg. For two years now, the patient has not been consuming alcohol, which he previously used in large quantities. He reported no other diseases or previous surgeries. Suspecting the presence of pseudoachalasia, or motility disorders of the esophagus, esophagoscopy was done during which a neoplastic process was not found, but the impression was created of the external compression in the middle third of the esophagus

at the back. Since the lungs' Rtg P-A findings were correct, differential diagnosis assumed the presence of vascular abnormalities in the mediastinum. A chest multi-slice computed tomografy (MSCT) was performed, which revealed ARSA resulting from a descending aorta KD, located below the left SA deviation. ARSA moves back and to the right, passing behind the trachea and esophagus, and in front of the spinal column. In the course of its aberration, it puts pressure on the esophagus in its middle third. Both carotid arteries are derived from TB, which is another rare variety of blood vessels of the aortic arch branching (Figures 1–5).

The patient was advised to undergo surgical treatment in case of dysphagic disorder deterioration, and until then, eat more often during the day, in small bites, if necessary, purée foods, and use proton pump inhibitors (PPI).



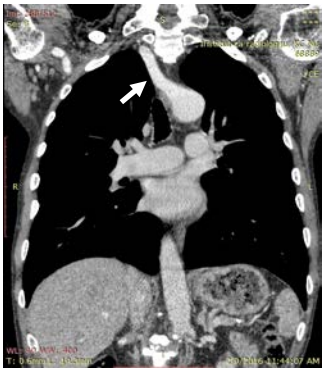
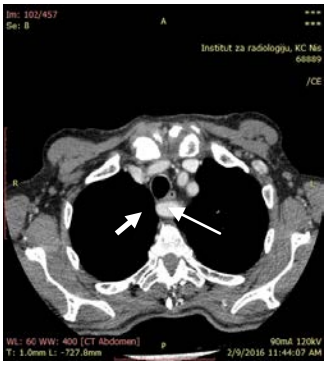


Figure 1. MSCT chest shows sagittal image of arteria lusoria
Figure 2. MSCT chest shows coronal image of arteria lusoria



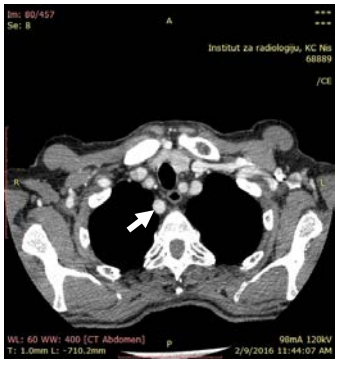


Figure 3. MSCT chest shows (thick arrow), compressed esophagus (thin arrow)
Figure 4. MSCT chest shows arteria lusoria (arrow)

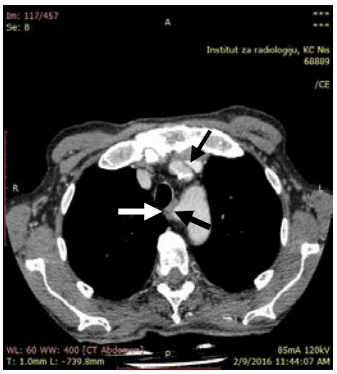


Figure 5. MSCT chest shows truncus bicaroticus (black arrow), Kommerell's diverticulum (thicker black arrow), compressed esophagus (white arrow)

Discussion

Aberrant right subclavian artery is a rare anomaly, but also the most common anomaly of the aortic arch (1, 10). First description of this arterial abnormality was cited by Hunauld in 1735 (11). A term "dysphagia lusoria" was first used in 1794 by Bayford, who described this type of dysphagia in an older woman who died of starvation due to the consequent esophagus obstruction (2, 5, 12, 13). On that occasion, ARSA was described as a "freak of nature" (L. lusus nature) and named arteria lusoria (5).

Arteria lusoria occurs due to disturbances during embryogenesis, when it comes to the involution of the fourth aortic arch on the right side along with the cranial part of the right dorsal aorta, whereby the seventh intersegmental artery remains annexed to the descending aorta (12). If there is no involution of the right dorsal aorta (in about 60% of cases) there occurs aneurysmal, infundibular expansion at the origin of ARSA—Kommerell's diverticulum (according to B. Kommerell, who first described it in 1936) (6, 12). ARSA can be accompanied by other anomalies of the heart and blood vessels, usually in the tetralogy of Fallot (14). Aberrant left subclavian artery and right-sided aortic arch branching are far less common (7).

The classification of ARSA types is based on the ways of anomalous branching from the aortic arch (13). Adachi–Williams classification of the anomalous ARSA branching describes four basic types of branching: 1) Type G-1, ARSA branching from distal aortic arch as the last branch, other branches of the aortic arch are normally separated; 2) Type CG-1, branching as in G-1, with a deviation of the left vertebral artery directly from the aortic arch; 3) Type H-1, ARSA separates as G-1, and the left and right common carotid arteries branch off from TB, which deviates from the aortic arch. This is found in 0.16% of cases and is usually detected as an incidental finding (15). It is even more rare in patients with dysphagia associated with ARSA (as in the presented case); 4) Type N-1 of branching is the mirror image of type G, is rarely encountered and represents right aortic arch and the left subclavian artery (LSA) which deviates beyond the right subclavian artery (15).

In the case of the presence of ARSA right n. laryngeus recurrens separates directly from the ipsilateral n. vagus and does not make a loop or a return arch around ARSA, but in the shortest route enters the laryngeal area (16). This must be taken into account when operating on the thyroid gland and trachea.

The presence of ARSA is usually asymptomatic and/or is found in autopsy as incidental findings. Rarely, the presence of ARSA creates difficulties with different degree dysphagia, dyspnea, stridor and hoarseness, usually after a long asymptomatic period and in old age. The appearance of symptoms in the elderly is explained by ARSA stretching around the esophagus, the appearance of atherosclerotic changes with increasing rigidity of the ARSA or its aneurysms or the aorta arch (7). Hoarseness as a sign occurs within Ortner's cardiovocal syndrome, similar to the dilation of the left atrium in mitral estuary stenosis (17, 18). Other signs and symptoms that occur less frequently include: retrosternal pain, cough, loss of body weight, asymmetrical pulse on the hands, trophic changes on the right hand, cyanosis, arterial insufficiency and erosion of thoracic vertebrae (11, 19). Problems related to the presence of ARSA in the showcased patient can be partly explained by the fact that due to the previous excessive alcohol consumption, a development of alcoholic polyneuropathy occurred, which has engulfed the submucosus plexus and plexus myentericus, thus causing a weakening of the esophagus wall resistance to already existing ARSA. Since alcohol is a good source of energy, the question is how well and how often the patient ate, and what quality the patient's food intake was in the previous period, and whether he was noticing and attributing any significance to the dysphagia occurrence.

The diagnosis of this anomaly is performed by esophagoscopy, barium contrast esofagography, MSCT, MRI and angiography (20, 21). ARSA diagnosis (with non-invasive methods) is set with high probability using MSCT (21). For diagnosing the existence of ARSA, it is essential to suspect its presence. The patient had previously visited many doctors because of the symptoms related to difficulty swallowing—otolaryngologists, neurologists, cardiologists, endocrinologists, hematologists and gastroenterologists. Interestingly, the first association of our team on the endoscopic findings of the extramural compression on the esophagus was the existence of ARSA, then descending aorta aneurysm and mediastinal/lung tumor. A performed MSCT of the thorax has confirmed the presence of ARSA.

Surgical therapy, endovascular and hybrid procedures in dysphagia lusoriae are considered a treatment option when difficulties are present and get worse over time (22, 23). The risk of complications and side events during the operative treatment of these patients does exist, since the surgical procedures are non-standardized, and are rarely performed in a small number of centers in the world.

Knowledge of the anatomy and anatomical anomalies of the blood vessels of the aortic arch and the timely detection of ARSA when planning operations on the esophagus, aortic arch, trachea and thyroid are essential for the prevention of possible complications during the surgery itself. One of the most serious complications is the injury of ARSA itself and the abundant intraoperative bleeding that is difficult to treat surgically (24). As one of the chronic complications of the transhiatal esophagectomy and the presence of the unrecognized ARSA arterio-esophageal fistula is described (11, 25).

If the complaints are mild and do not get worse, the changes in the hygienic-dietary regimen of life and a diet with the use of PPI are recommended (22). For this patient, we opted for a combined PPI therapy, also applying hygienic and dietary life regime, recommending frequent meals with small bites of food, use of mixed and liquid food, and the outcome was: a loss of body weight stopped, stable general condition and acceptable troubles for the time being. The patient was advised that, in case of symptoms worsening, the possibility of surgical, endovascular and/or hybrid treatment is available. Gastrostomy as a form of treatment was dismissed because the oral food intake is one of the major "sources of pleasure", and the patient is not acutely life threatened by dysphagia.

Conclusion

Aberrant right subclavian artery with truncus bicaroticus is a rare clinical entity. It should be suspected in the occurrence of dysphagia accompanied by hoarseness and loss of body weight. MSCT proved to be the most reliable non-invasive diagnostic tool. Asymptomatic patients should be monitored and their hygienic-dietary regimen of life modified, and the PPI should be used. Active surgical, endovascular and/or hybrid treatment should be performed in symptomatic patients whose ailments worsen.

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DYSPHAGIA LUSORIA: PRIKAZ PACIJENTA SA ABERANTNOM DESNOM POTKLJUČNOM ARTERIJOM I ZAJEDNIČKIM STABLOM KAROTIDNIH ARTERIJA

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Aberantna desna arterija subklavija (ADAS) predstavlja retku vaskularnu anomaliju. Još se ređe dešava da bude udružena sa disfagijom i zajedničkim stablom karotidnih arterija – *truncus bicaroticus* (TB). Bejford je 1794. godine opisao ADAS i nazvao je tu pojavu *arteria lusoria*, a ovaj tip disfagije *dysphagia lusoria*. Infundibularno proširenje na odstupu ADAS-a prvi je opisao Komerel 1936. godine, pa se naziva Komerelov divertikulum.

Kod šezdesetdevetogodišnjeg pacijenta M. D. potvrđene su disfagija i promuklost. Dva meseca pre pregleda došlo je do gubitka težine (9,5 kg). Ezofagoskopski je uočena spoljna kompresija na zadnjem zidu srednje trećine jednjaka. Pregled grudnog koša urađen pomoću višeslojne kompjuterizovane tomografije (engl. *Multi-slice Computed Tomography*), kao najpouzdanijeg neinvazivnog dijagnostičkog sredstva, ukazao je na ADAS i Komerelov divertikulum sa TB-om.

Prisustvo ADAS-a je najčešće asimptomatsko i slučajno se otkriva prilikom pregleda i obdukcija. Disfagične tegobe praćene gubitkom telesne težine (ako su prisutne) potiču od spoljne kompresije ADAS-a na ezofagus, dok se promuklost javlja u okviru Ortnerovog kardiovokalnog sindroma. Bolesniku je dat savet da se uputi na operativno lečenje ako dođe do pogoršanja disfagičnih tegoba. Preporučeno mu je da dotad jede češće u toku dana, da zalogaji budu mali, kao i da koristi inhibitore protonske pumpe.

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***Ključne reči**: dysphagia lusoria, arteria lusoria, aberantna desna arterija subklavija, Komerelov divertikulum*