

Rare Causes of Chronic Cough: Aberrant Right Subclavian Artery: A Case Report

Kronik Öksürüğün Nadir Nedenleri: Aberran Sağ Subklavian Arter: Olgu Sunumu

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Abstract

Aberrant right subclavian artery (ARSA) is a congenital anomaly with a reported frequency of 0.5-1% and generally shows an asymptomatic clinical course. Being rare and usually asymptomatic complicates diagnosis and treatment. Although respiratory symptoms are rare in ARSA, chronic cough may be a symptom for diagnosis. Cases with chronic cough that persists despite treatment should be evaluated using posteroanterior chest X-ray and, if necessary, thorax computed tomography. In our article, we presented a case of a patient who applied to our family medicine outpatient clinic with a complaint of chronic cough and was diagnosed with ARSA.

Keywords: Subclavian artery, congenital abnormalities, cough, primary health care

Öz

Aberran sağ subklavian arter (ASSA), sıklığı %0,5-1 arasında bildirilen ve genellikle asemptomatik klinik seyir gösteren konjenital bir anomalidir. Nadir görülmesi ve genellikle asemptomatik olması tanı ve tedavi sürecini zorlaştırmaktadır. ASSA'da solunum semptomları nadir olmakla birlikte kronik öksürük, hastaların tanı almasını sağlayan semptomlardan biri olabilir. Tedaviye rağmen devam eden kronik öksürüğü olan olgular posteroanterior akciğer grafisi ve gerekiyorsa toraks bilgisayarlı tomografisi ile değerlendirilmelidir. Yazımızda aile hekimliği polikliniğimize kronik öksürük şikayetiyle başvuran ve ASSA tanısı alan bir olguyu sunduk.

Anahtar Kelimeler: Subklavyen arter, konjenital anormallikler, öksürük, birinci basamak sağlık hizmeti

Introduction

A cough that lasts longer than eight weeks in adults is called chronic cough⁽¹⁾. Chronic cough is a symptom that can be seen in most chronic lung diseases and some non-pulmonary diseases.

While the most common causes include upper airway cough syndrome, asthma, and gastroesophageal reflux, there

are many causes of upper respiratory tract origin (such as allergic rhinitis, chronic sinusitis), lower respiratory tract origin [such as asthma, chronic obstructive pulmonary disease (COPD), foreign body], and non-respiratory origin [such as gastroesophageal reflux disease (GERD), drug use, and cardiovascular diseases]⁽²⁾. Aberrant right subclavian artery (ARSA) is clinically the most common embryological abnormality of the aortic arch. This abnormality was first



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described by Hunauld in 1735. Bayford described dysphagia lusoria as a clinical entity in a woman with a long history of dysphagia and ARSA at autopsy in 1787. Therefore, it is also known as Bayford-Autenrieth dysphagia⁽³⁾. ARSA can cause dysphagia, dyspnea, or chronic cough due to compressive mechanisms with adjacent organs⁽⁴⁾.

In our article, we present a case of a patient who presented to our family medicine outpatient clinic and was diagnosed with ARSA.

Case Report

A 59-year-old male patient was admitted to our family medicine outpatient clinic with a cough complaint. The patient said that he works in the food industry. However, he is not working now. The patient had a 60 pack/year smoking history. He had no history of alcohol or substance abuse. The cough has been going on since childhood. The patient's cough was accompanied by colorless sputum. He added that he had shortness of breath on exertion for the past few years. When his additional complaints were questioned, he said that he had a reflux complaint that had been going on since childhood.

The patient had a history of diabetes, hypertension, and chronic obstructive pulmonary disease.

The diagnosis of hypertension was present for approximately 6 years, and the diagnosis of diabetes was present for 10 years.

The diagnosis of COPD was made 3 years ago by the physician to whom he applied due to cough.

The patient had a stent because of myocardial infarction 12 years ago.

Drugs used by the patient: Metformin 2x1000 mg/day, lercanidipine hydrochloride, salbutamol 100 mcg inhaler (if needed), salmeterol fluticasone propionate inhaler, montelukast 1x10 mg/day, and desloratidine 1x5 mg/day.

However, the drugs he used did not improve the patient's cough and shortness of breath complaints. The patient's family history was unremarkable.

The patient had previously applied to the chest diseases outpatient clinic because of cough.

When the patient's past examinations were examined, it was seen that the thorax computed tomography (CT) imaging requested by the chest diseases outpatient clinic on 09.07.2019 was reported as the right subclavian artery with aberrant origin.

However, he stated that he was not told anything about the patient's condition, but only that he had a problem with his lungs. In addition, he did not apply to the cardiovascular surgery outpatient clinic for this reason.

Physical examination: Fever: 36.5 °C, pulse 100/min, blood pressure 114/70 mmHg.

Lung sounds are natural. Both lungs accompany breathing. There were no al rhonchus.

Posteroanterior chest X-ray (Image 1) and laboratory tests [complete blood count, kidney function tests, liver function tests, glycosylated hemoglobin (HbAlc), thyroid stimulating hormone (TSH), vitamin B12, ferritin] were requested from the patient.

Laboratory Findings

Glucose -toughness: 150 mg/dL, creatinine: 0.86 mg/dL, blood urea nitrogen: 9.7 mg/dL, estimated glomerular filtration rate: >60 mL/min/1.73 m², alanine aminotransferase: 18 U/L, aspartate aminotransferase: 15 U/L, alkaline phosphatase: 124 U/L, gammaglutamyl transferase: 56 U/L, lactate dehydrogenase: 136 U/L, ferritin: 29.8 μ g/L, vitamin B12: 84 ng/L, hemoglobin: 14.8 gr/dL, leukocytes: 9.0x10³/µL, platelets: 300x10³/µL, sedimentation: 20 mm/h, TSH: 0.517 uIU/mL, free T4: 11.51 pmol/L, HbA1c: 6.9%, C-reactive protein: 3.67 mg/dL.

An enlargement of the distal esophagus was observed on chest X-ray. Lung fields were normal.

When the current condition of the patient was evaluated, the patient was informed that the cough and reflux complaints that did not go away for a long time were connected to the right subclavian artery with aberrant origin. The patient was referred to the cardiovascular surgery outpatient clinic. Surgery was recommended to the patient by cardiovascular surgery.

However, the patient did not accept the operation because of the high risk of mortality in the operation.

Discussion

Typically, three major arteries arise from the aortic arch: The brachiocephalic trunk (divided into the right common carotid artery and right subclavian artery), the left common carotid artery, and the left subclavian artery (Figure 1). However,

in the abnormality of ARSA, the brachiocephalic trunk is absent and four major arteries arise from the aortic arch; right common carotid artery, left common carotid artery, and left subclavian artery. The last one is the most distal left-sided origin, the right subclavian artery, also called arteria lusoria (Figure 2). This artery goes to the right arm, crosses the midline of the body, and usually runs behind the esophagus⁽³⁾. ARSA may follow a retroesophageal course, a course between the trachea and esophagus, or a pretracheal pathway. The atypical vessel may compress the trachea and esophagus while forming an incomplete vascular ring around them⁽⁵⁾.

The frequency of ARSA is 0.5-1% and varies all over the world, but in Europe, it occurs in 0.11% (England), 0.16% (Greece), 0.3% (France), or 0.36% (Netherlands) of the population, depending on the country detected⁽⁶⁾. Studies have also been conducted on other continents, with Asia accounting for 0.1-0.2% of cases (China and Japan, respectively); North America, 0.5% of cases (United States); and Australia and Oceania, 0.8% of cases (New Zealand)⁽⁷⁾. In a study, it was determined that women have two times more isolated ARSA than men⁽⁵⁾.

Although 60-80% of patients can remain asymptomatic throughout their lives, in a study by Michał et al., $^{(3)}$ who

presented a systematic review of 141 reports, the most frequently reported symptoms due to compression of adjacent structures by the ARSA were dysphagia (71.2%), dyspnea (18.7%), retrosternal pain (17.0%), cough (7.6%), and weight loss⁽⁷⁾. In the patient who applied to our clinic, we observed the complaint of cough that has continued since childhood. In this retrospective study, the mean age of all patients evaluated was 49.9 years; however, statistically significant differences were found between the mean ages of male and female subjects (54.0 years and 44.9 years, respectively), and the patient who applied to our clinic was 59 years old⁽³⁾. In a publication on the subject in our country, in which 8 cases with ARSA anomaly are presented, 2 cases applied to our clinic with the complaint of chronic cough that has lasted for several years. Similar to our patient, various tests were performed on both patients, and antacid treatments were started with the diagnosis of GERD. Inhaled corticosteroid and long-acting bronchodilator treatment was administered to one of the two patients diagnosed with asthma. However, two patients did not benefit from these treatments. No pathology was detected in the physical examination and chest X-ray, and thoracic CT was performed to detect the etiology of chronic cough and ARSA was detected⁽⁸⁾. In the patient who visited our clinic, the physical examination was









Image 1. PA chest radiograph

normal, and the distal esophagus was enlarged on the chest X-ray.

Chest radiographs should be obtained to rule out most infectious, inflammatory, and malignant thoracic conditions, unless a possible cause is identified⁽⁹⁾. In our patient, we first requested chest radiography to rule out these causes.

Family medicine is a department with six core competencies. One of these core competencies is authentic problem-solving skills. Within the framework of this competence, the family physician can make a unique clinical decision according to the prevalence and incidence of diseases in the society to which he/she is affiliated. It has a unique clinical decisionmaking feature because serious diseases are encountered less frequently in the primary care setting compared with the hospital environment, and the prevalence and incidence of diseases differ according to the hospital environment. In addition, with a comprehensive approach, which is another core competence, disease management is performed with limited information in the period when the disease is not differentiated yet⁽¹⁰⁾. As in our case, the important thing in the first approach to a patient with ARSA, which is an incompletely differentiated and rarer disease in primary care compared with the hospital, is to examine the patient regardless of the

complaint, to evaluate the patient in detail, and to quickly refer the patient to the necessary departments to prevent complications. This situation was probably overlooked in this study because evaluations were made only in their own fields by other clinics in the hospital environment. Thanks to the comprehensive evaluation in the family medicine outpatient clinic, the cause of persistent cough was revealed. Thus, unnecessary hospital admissions and drug use have been eliminated.

Conclusion

Structural anomalies, such as ARSA, should be considered in the differential diagnosis of patients presenting with chronic and persistent cough, and further investigation should be performed.

Ethics

Informed Consent: Informed consent was obtained.

Footnotes

Authorship Contributions

Concept: H.A., İ.F., Data Collection or Processing: K.K., E.S., H.Ö., Analysis or Interpretation: H.A., İ.F., D.A.B., Literature Search: K.K., E.S., H.Ö., Writing: H.A.

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