

Unusual Presentation of Inferior Thyroid Artery Aneurysm: Case Report

İnferiyor Tiroid Arter Anevrizmasında Beklenmedik Başvuru Şekli

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ABSTRACT We present a case with an aneurysm of inferior thyroid artery after minor trauma. A 44-year-old man was admitted with a left neck side swelling occurring after taking bath. Duplex ultrasound showed a mass of 24 x 25 mm with intracavitary turbulent flow in the left anterolateral supraclavicular region, suggesting an aneurysm possibly supplied by the origin of the left vertebral artery. However, selective angiography demonstrated a left inferior thyroid artery aneurysm formation. The patient was scheduled for coil embolization. However, he refused firstly. After detailed description more pros than cons, he is still considering this option. The reason for the aneurysm formation was possibly atherosclerotic in origin because he had risk factors for atherosclerosis. This case is important in that occurring after minor trauma in the presence of vascular insulating factors.

Key Words: Thyroid gland; arteries; injuries; aneurysm

ÖZET Minor travma sonrası inferior tiroit arterde anevrizma gelişen bir olgu sunulmaktadır. Kırkdört yaşındaki erkek hasta banyo yaptıktan sonra boynun sol tarafında gelişen şişlik nedeniyle başvurdu. Yapılan dubleks ultrason tetkikinde boynun sol supraklavikular anterolateral bölgesinde kavite içi türbülans akım gösteren 24 x 25 mm ebatlarında, olası sol vertebral arterden kaynaklanan anevrizmal kitle izlendi. Ancak selektif anjiyografide anevrizmal kitlenin inferior tiroit arterden kaynaklandığı tespit edildi. Hastaya kateterle koil embolizasyon önerilmesine rağmen tedaviyi reddetti. Yapılacak işlemin olumlu tarafları detaylı anlatılmasına rağmen hasta hala kararsızlığını sürdürmektedir. Anevrizma nedeni olarak, risk faktörleri varlığından dolayı ateroskleroz damar hastalığı düşünüldü. Bu olgu vasküler hasar yapıcı faktörlerin varlığında minor travma sonrası anevrizma oluşması açısından son derece önemlidir.

Anahtar Kelimeler: Tiroid bezi; arterler; yaralanmalar; anevrizma

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Aneurysms involving small peripheral arteries such as inferior thyroid artery and its branches are extremely rare. These aneurysms are usually atherosclerotic, traumatic, mycotic or occasionally due to angitis or syphilis.^{1,2} The presenting symptoms of inferior thyroid artery aneurysm reported in the literature are cervical swelling,¹ hoarseness of voice,³ vertigo,⁴ cervicofacial hemorrhage,^{5,6} dysphagia, mediastinal hemorrhage, and respiratory distress.⁷⁻⁹ In scientific literature only 28 cases have been reported, of which 32.9% regard ruptured aneurysms and 10.7% led to mortality.² Therefore, surgical exclusion or coil embolization is always recommended, even in asymptomatic cases, because of high mortality and

morbidity.^{2,7-9} We describe a rare case of an aneurysm of the left inferior thyroid artery presenting as acutely developed left neck swelling compressing the left cervical sympathetic nerves, causing left drooping eyelid, decreased eye pupil size and sunken globe.

CASE REPORT

A 44-year-old man admitted to our hospital for further evaluation of temporarily occurring a swelling in the left side of his neck. He had been good health until one week previously, when he developed acutely a swelling in the left neck after gentle massage for relieving of his neck pain in bath. Subsequently, the patient was admitted to a local hospital, where he was noted to be hypertensive (blood pressure of 190/120 mmHg) but no marked swelling was found in his neck and therefore no considerable diagnosis was put at that time. Blood pressure was controlled with antihypertensive agents in the emergency department and sent to home after prescribing a course of antihypertensive agents. However, the day after the patient went to the emergency department, he began to manifest a picture of smaller left eye pupil and sunken globe compared to his right eye. The patient was referred for further evaluation by his primary care physician for complicated situation. His medical history was only remarkable for hypertension, managed with an angiotension receptor blocker. He was a nonsmoker and lifelong nondrinker. He denied antecedent head and neck trauma, contact with sick persons and drug abuse.

On physical examination, his vital signs were as follows: temperature, 37°C; heart rate, 82 beats/min; respiratory rate, 16 breaths/min; blood pressure, 130/80 mmHg. Cardiovascular examination revealed a nonradiating mild diastolic murmur at the right sternal border. Examination of the neck revealed a very small, firm and slightly mobile mass of 0.5 x 0.5 cm size with smooth borders in the left anterolateral supraclavicular region. There was prominent ptosis, enophthalmos and meiosis in his left eye (Figure 1). No focal neurological deficits were identified.



FIGURE 1: Prominent clinic picture of left eye meiosis, ptosis and enophthalmos (R: right).

Findings on the rest of the physical examination were normal.

Echocardiography revealed only mild aortic regurgitation. Ultrasonography (USG) and Doppler USG of the neck showed a mass of 24 x 25 mm with intracavitary turbulent flow in the left anterolateral supraclavicular region, suggesting an aneurysm possibly supplied by the origin of the left vertebral artery. With an assumption of Horner syndrome, dynamic axial and coronal computed tomography (CT) and three-dimensional CT images of the neck and thorax performed with intravenous contrast material showed an aneurysmal mass of 25 mm x 28 mm in size with well-demarcated borders stemming from either left vertebral artery or other neck vessels including inferior thyroid artery (Figure 2). Aneurysm was in continuity with an ectatic small artery. No obvious malignant or infiltrative processes of the lung, mediastinum or neck were visualized. Although the CT scan showed evidence of an arterial aneurysm of one of the neck vessels, the exact origin of the aneurysm could not be clarified. Magnetic Resonance Imaging (MRI) of the neck vessel has also not delineated comprehensive information about aneurysm from which vessel was originated (Figure 3). At this stage, it would be appropriate to perform selective vascular angiography to identify the exact site and origin of the aneurysm. This study was performed, revealing an aneurysm stemming from mid region of inferior thyroid artery, a branch of thyrocervical truncus of the left subclavian artery (Figure 4). Postaneurysmal course of the inferior thyroid artery was ectatic (Figure 5). Left vertebral artery was normal.

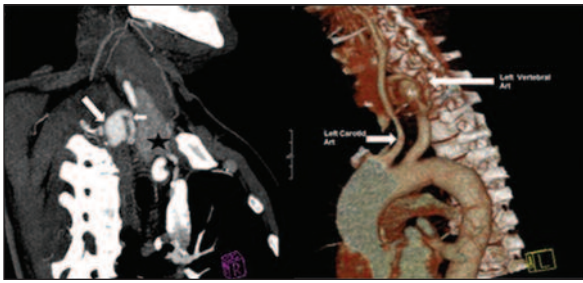


FIGURE 2: (on the left) Coronal CT image clearly shows the aneurysm (long white arrow) and postaneurysmal ectatic course of the vessel (short white arrow). The close association of aneurysm with thyroid is also seen clearly (asterix: shows left thyroid lobe). (on the right) 3D CT images demonstrates aneurysm and its neighboring vessels on the left lateral view (An: Aneurysm).



FIGURE 3: Definite diagnosis for the origin of the aneurysm images could not be found in MRI but it revealed close association of aneurysm with left thyroid lobe (An: Aneurysm; Th: Thyroid).

Laboratory data including collagen vascular disease, connective tissue disorders and haematological investigations were in the reference range and serological tests for syphilis were negative.

After definitive diagnosis of the aneurysm, the patient was scheduled for coil embolization. However, he is still considering this option.

DISCUSSION

This case emphasizes the need for careful clinical evaluation of patients presenting with equivocal symptoms that may be a clue for underlying

disease. The causes of inferior thyroid artery aneurysm reported in the literature are mainly trauma, arteriosclerosis, degenerative changes and iatrogenic injuries such as central venous cannulation.^{1,7,9} Atherosclerosis is present in the majority of the patients, but media necrosis and posttraumatic false aneurysm can also occur.⁷ Degenerative changes, especially of the media have been reported but no systemic features of Marfan’s syndrome have been noted in any of these cases. Hypertension was recorded in such patients and is a contributory factor in the pathogenesis of atherosclerosis.¹

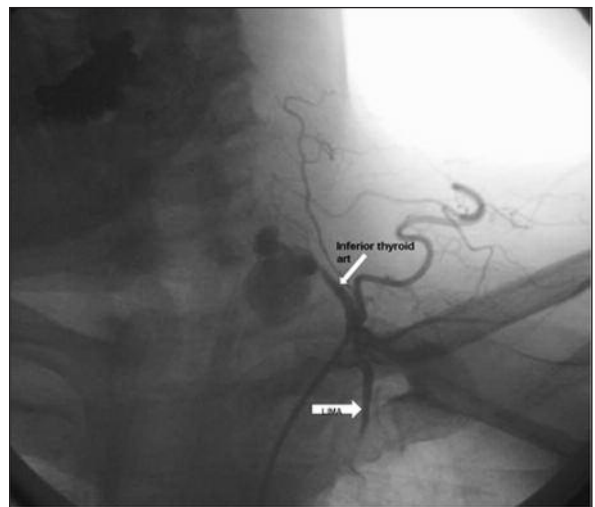


FIGURE 4: Selective angiography of the left thyrocervical trunk clearly demonstrated the aneurysm stemming from inferior thyroid artery (thin white arrow) (LIMA: Left internal mammary artery).

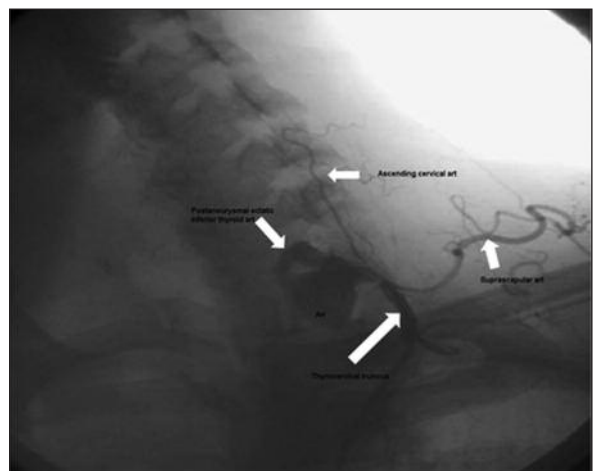


FIGURE 5: Selective angiography demonstrated ectatic postaneurysmal course of the inferior thyroid artery.

In our case, despite the absence of pathological analysis, we suspected that arteriosclerosis was the cause of aneurysm and hypertension could have been causative factor for atherosclerosis. It is assumed that atherosclerosis can no longer be thought of as a fixed model where plaque growth would always lead to luminal narrowing. On the contrary, the widely accepted thought is arterial remodelling where atherosclerosis may cause vessel obstruction or dilatation.¹⁰ In our patient postaneurysmal course of inferior thyroid artery was ectatic and atherosclerosis was suspected as a potential etiology of ectatic vessel wall. Minor trauma (gentle neck massage while taking shower) in the presence of ectatic arterial wall which was possibly resulted from atherosclerosis could explain aneurysm development

Systemic vasculitis was another consideration because it can involve any organ system and mimic other diseases. However, this patient's acute presentation would argue against vasculitis because vasculitis usually presents as a subacute illness that evolves over weeks to months. This patient had no history of vasculitis and laboratory investigations excluded this diagnosis.

Ruptures of such aneurysms may cause acute respiratory failure as a result of compression of the trachea, vocal cord paralysis caused by compression of recurrent nerve or hypovolemic shock as a result of mediastinal hemorrhage and dysphagia caused by compression of the esophagus.^{1,7,9} Review of the literature revealed that observation with tracheostomy after the spontaneous rupture led to the death of two patients.⁸ In these emergency settings, they carry high mortality and morbidity rate;⁷ therefore, endoluminal techniques including arterial embolization has to be preferred as the first choice. If any failure is encountered with these techniques immediate surgery must be performed.⁹ Patients without rupture symptoms included hoarseness caused by compression of recurrent nerve, paralysis of the third branch of the trigeminal nerve, C8/T1 paraesthesia, dysphagia and asymptomatic swelling.⁷

The most striking presentation of our patient involved left eye enophthalmos, meiosis and pto-

sis, suggestive of Horner syndrome. Review of the literature revealed that only two cases had complication of Horner syndrome occurring postoperatively.⁸ However, this patient's picture of Horner syndrome was present at first admission before he had received any interventional or surgical procedures. Finally, this could have been resulted from possible direct pressure of the aneurysm over the left paravertebral sympathetic chain and inferior and stellate ganglion because of close proximity.¹¹

For diagnosis, standard radiography which shows enlarged upper mediastinum, ultrasonography and Doppler USG, and computed tomography scanning.⁹ For verification and therapy, selective angiography with selective embolization is the method of choice.^{2,5,7,8}

It is advocated that all inferior thyroid artery aneurysms, because of high mortality and morbidity, must be treated actively at the time of the diagnosis.^{2,7} The two possible treatments for aneurysms are surgery or angiographic embolisation.² The definite strategy for treatment of true or pseudo aneurysm of inferior thyroid artery is surgery. Surgical therapy has been performed successfully in symptomatic and asymptomatic patients.⁸ Endoluminal techniques have now become the first line of therapy instead of surgery in most instances, especially in an emergency setting, but they have also been used successfully in elective patients.^{5,7,8} The most widely-used materials to embolise or exclude the aneurysm from visceral circulation are coils, gelfoam plugs, isobutyl 2-cyanoacrylate.⁷ A review of the literature indicates that true and pseudoaneurysms of the inferior thyroid artery are treated via endoluminal techniques, which have lower procedure morbidity and a high success rate.^{7,8} Coil embolization can be added expeditiously in an elective or urgent setting, avoiding the risk of nerve injury (including Horner syndrome and vocal cord paralysis) reported with surgical resection.⁸

No certain clinical consensus has been encountered in literature on the treatment strategy comparing endoluminal techniques with surgery.

Surgery must also be performed in such aneurysms if angiographic embolization has failed⁹ or in the presence of special conditions such as continuous compression by the coil-filled aneurysm to the adjacent structures. In our case, we believe that continuous compression on the nerve following the embolotherapy with no relief of Horner's syndrome necessitates surgery.

CONCLUSION

This case illustrates two important principles in the pathology of inferior thyroid artery aneurysm. First, its rarity can cause the difficulties such as misdiagnosis and secondly, aneurysm developed in inferior thyroid artery carries high morbidity and mortality rates.

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