CASE REPORT

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Hybrid Treatment of Unresectable Sublingual Hemangioma

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ABSTRACT Vascular anomalies have categorized as two groups, hemangiomas and vascular malformations. Most of vascular anomalies have located superficially in the regions of the head and the neck. The growth period of deep hemangiomas occurs later than that of superficial hemangiomas. The accepted method of treatment of vascular malformations and hemangiomas, which can be life-threatening and can not be resected in this way, is the method of ligation or occlusion of blood vessels. The feeding vessels usually originate from the branches of external carotid artery or the main trunk of the external carotid artery. The aim of this report is to describe our approach to a patient with symptomatic non-resectable sublingual hemangioma.

Keywords: Carotid; ligation; hemangiomas; endovascular theraphy; hybrid theraphy

Hemangiomas and vascular malformations are neoplasms that usually locate on the head and neck and are considered benign. The main factors affecting the clinical course of hemangiomas are growth rate, location, histopathology and resectability. In this case report, we described a symptomatic patient with sublingual hemangioma who was not suitable for resection.

CASE REPORT

A 29-year-old male patient was admitted to our hospital with the complaints of speech disorder, tongue asymmetry, multiple raised bluish spots on the tongue and progressive tongue enlargement, moderate swallowing difficulty and shortness of breath. There were swellings on the left side of the patient's neck, which was evaluated as cervical lymphadenopathy. These lymphadenopathies were examined non-pulsatile and non-fluctuating masses. In the patient's anamnesis, it was learned that a biopsy was performed from these masses before and a diagnosis of hemangioma was made. Color doppler ultrasonography for lesions has been reported to be compatible with hemangioma. Computed tomographic angiography (CTA) confirmed the presence of sublingually located vascular-associated lesions fed from bilateral carotid external arteries (CEA) via collateral vessels (Figure 1, Figure 2).

Due to the presence of multiple lympha denopathies in the left cervical region, the option of surgical intervention for left carotid external artery (LCEA) ligation was considered in the foreground. In the operation, it was observed that the mass was vascularized with the collateral that developed from the LCEA. Direct ligation was performed without arterial clamping. Excisional biopsy was then performed on the lymphoid tissues.

In the postoperative period, the existing speech disorder improved. The volume of the mass occupying space in the oral cavity gradually decreased. Patho-

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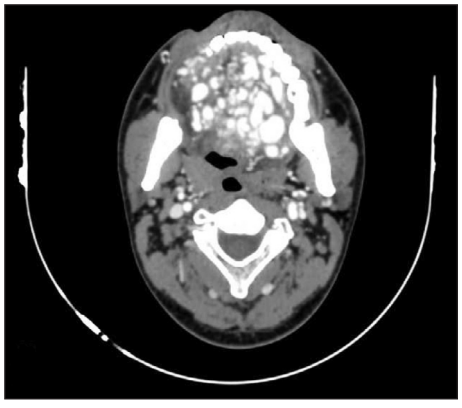

FIGURE 1: Sublingual mass with increased vascularization, computed tomographic angiography image.

FIGURE 2: Computed tomographic angiography image showed the feeding arteries of the hemangioma (arrows).

logical analysis of excisional lymphoid biopsy (CD3, CD20 and CD79a immunohisto-chemistry) reported as benign reactive hyperplasia. The patient was discharged on the 5th postoperative day.

Two months after surgery, significant reduction in the size of the tongue was observed. Control angiography showed that LCEA was occluded (Figure 3A). In the control CTA, it was observed that the hemangioma continued to be fed by collaterals originating from the right carotid external artery (RCEA). Thereupon, a balloon-expandable stent-graft (Advanta V12) of 7.0×38×120 mm was implanted in the angiography unit, starting from the right common carotid artery (RCCA) and covering the RCEA ostium (Figure 3B). In the 6th month postoperatively, it was determined that there was a complete reduction in the size of the mass and that the tongue functions were preserved. Consent was obtained from the patient.

DISCUSSION

Mulliken and Glowacki divided vascular anomalies into two main categories as hemangiomas and vascular malformations.[1] There are differences between the histopathological patterns, clinical features and prognosis of hemangiomas and vascular malformations. New techniques and treatment procedures are developing in the management of hemangiomas.[2] Hemangiomas are benign neoplasms usually located in the head and neck region. They often appear at birth. Growth of a hemangioma in the early stage can be rapid and unpredictable. After this process, slower and gradual involution is seen.[3] Hemangiomas can be life-threatening when they involve the larynx and trachea. They can also lead to ulceration, infection, and necrosis.[2]

The vast majority of hemangiomas are not candidates for conventional excisional surgery as first-line therapy. Isolated and small lesions can be treated with laser or simple surgery to prevent their growth. With these simple interventions, the regression of the mass can be achieved and it can create an advantageous situation for subsequent treatments. If the mass is enlarged despite laser therapy, the addition of drugs to the treatment (steroids or B-blockers) should be considered.[4] However, more experience is needed to understand the effects of these drugs added to the treatment on hemangiomas. Embolization has proven to be an effective treatment method for unresectable lesions. However, it should be kept in mind that embolic material may move from the CEA to the internal carotid artery (ICA) during this procedure and stroke may develop as a result.[5] Similarly, it should be remembered that embolic material may pass through the extracranial collateral pathways to the ICA and cause a stroke.

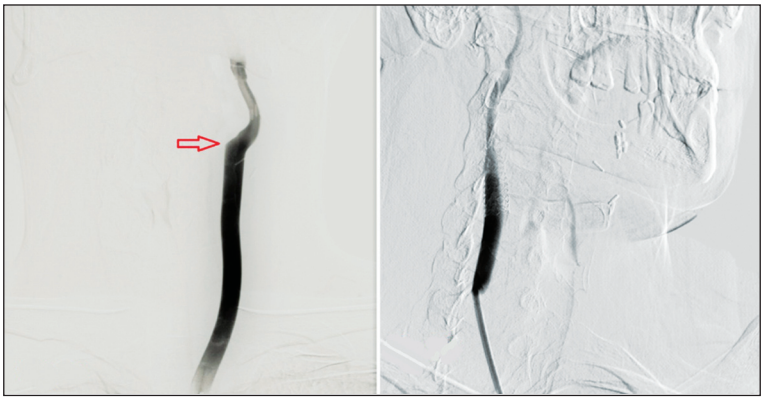

FIGURE 3: A) Selective angiographic image of the left carotid artery, ligation of left carotid external artery (arrow). **B)** Selective angiographic image of the carotid stent-graft implantation of the right carotis artery, occlusion of the right carotid external artery.

In this case report, we presented a case of hemangioma that was found to be non-resectable. Surgical intervention was planned primarily due to the presence of multiple lymphadenopathies in the surgical area in the left neck region. For this reason, both excision of lymphadenopathies and LCEA ligation were performed with surgical intervention. In the postoperative 2nd month follow-up of the patient, it was observed that there was a significant reduction in the tongue volume and the tongue returned to its normal dimensions. At the same time, a 7.0×38×120 mm balloon expandable stent-graft (Advanta V12) was implanted starting from the RCCA to the right ICA in this control. Thus, the RCEA was occluded at its origin. Since the bilateral occlusion procedure to be performed in the same session may cause tissue ischemia, we implemented a two-stage intervention plan. In the 6-month clinical follow-up of this case, it was observed that normal tongue functions were preserved and no additional pathology developed.

Ligation of the bilateral external carotid arteries is among the surgical treatment options for sublingual hemangiomas. The only known complication of this procedure is the development of tongue tip necrosis.[6]

In the approach to neck masses, imaging methods should be preferred in order to determine the treatment option. These methods such as ultrasonography, magnetic resonance angiography, computed tomography angiography can be used. As in the history of this patient, diagnostic biopsy should be avoided due to the high risk of bleeding, especially in the presence of a hemangioma is suspected. Multidisciplinary approach should be prioritized in the treatment of such cases. In addition, life-long follow-up is required because of the high risk of potential recurrence of such masses. Close interdiciplinary cooperation is neccessary for treating cases like our's. Due to potential recurrence risks of the mass, life time follow up is required.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Ahmet Fatih Özyazıcıoğlu;* ***Design:*** *Ahmet Kağan As, Burak Erdolu;* ***Control/Supervision:*** *Ahmet Fatih Özyazıcıoğlu, Burak Erdolu;* ***Data Collection and/or Processing:*** *Ahmet Kağan As;* ***Analysis and/or Interpretation:*** *Burak Erdolu, Ahmet Kağan As;* ***Literature Review:*** *Sadık Ahmet Sünbül, Peruze Çelenk;* ***Writing the Article:*** *Ahmet Kağan As;* ***Critical Review:*** *Ahmet Fatih Özyazıcıoğlu;* ***Materials:*** *Ahmet Fatih Özyazıcıoğlu.*

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