Case Report / Olgu Sunumu

Management of lingual hemangioma in a case with von Willebrand disease

Von Willebrand hastalığı olan bir olguda lingual hemanjiyom tedavisi

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Hemangiomas are the most common vascular tumors and von Willebrand disease is the most common inherited bleeding disorder. In this article, we report a 21-year-old female who had von Willebrand disease presenting with 1x1 cm lesion of the tongue. The lesion was located at the anterior one-third in midline tongue throughout full-thickness. The patient was administered cryoprecipitate preoperatively. Then the lesion was excised under local anesthesia. Histopathological examination revealed a hemangioma. No complication such as bleeding or infection after the operation was observed.

Key Words: Hemangioma; surgery; tongue; von Willebrand disease.

Hemanjiomlar en sık görülen vasküler tümörler olup, von Willebrand hastalığı en sık görülen kalıtımsal kanama bozukluğudur. Bu yazıda, dilinde 1x1 cm lezyonu olan ve von Willebrand hastalığı bulunan 21 yaşında bir kadın olgu sunuldu. Lezyon, tam kat boyunca dilin orta çizgisinde üçte bir oranında ön kısımda yerleşmişti. Ameliyat öncesi hastaya kriyopresipitat uygulandı. Daha sonra lezyon lokal anestezi altında eksize edildi. Hitopatolojik inceleme sonucunda hemanjiyom görüldü. Ameliyat sonrasında kanama veya enfeksiyon gibi herhangi bir komplikasyon izlenmedi.

Anahtar Sözcükler: Hemanjiyom; cerrahi; dil; von Willebrand hastalığı.

Von Willebrand disease (vWD) is an autosomally inherited congenital bleeding disorder involving deficiency of von Willebrand factor (vWF). Von Willebrand disease is the most common inherited bleeding disorder with prevalence of 0.6-1.3%.[1] There are three types of vWD. Type 1, deficiency of vWF, the most common, usually is mild; type 2, abnormal vWF, is less common; type 3, complete absence of vWF is rare. People with this condition often experience menorrhagia, epistaxis, gingival bleeding, and prolonged bleeding or oozing following an injury or surgery.[2]

Hemangioma is a benign vascular lesion. Lingual hemangiomas are rare tumors which cause spontaneous hemorrhage from the tongue.[3]

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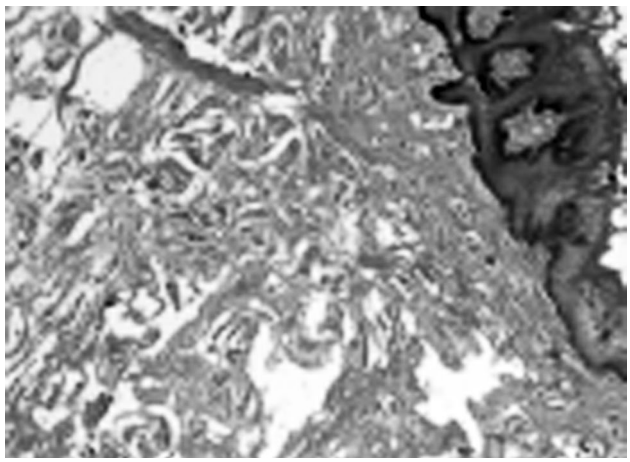

Figure 1. *Uncapsulated wide vascular lumen beneath squamous epithelium, between myofibrils (H-E x 4).*

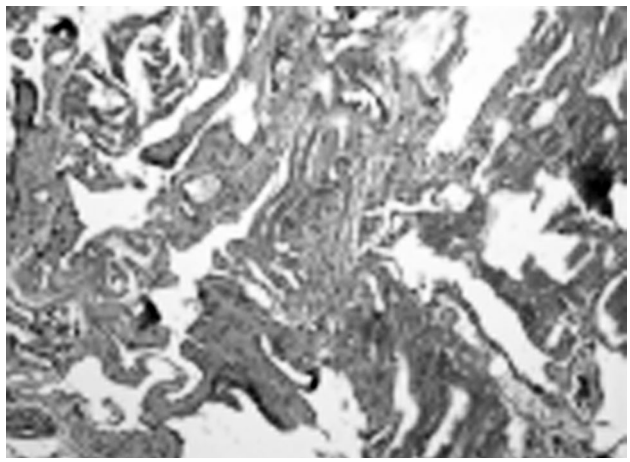

Figure 2. *Wide vascular lumen with erythrocytes and some connective tissue stroma between vessels (H-E x 10).*

In this article we report management of lingual hemangioma in a patient with vWD.

CASE REPORT

A 21-year-old female was admitted with a lesion of her tongue that was growing for the last two months. The lesion was causing pain when moving the tongue. There was a 1x1 cm purple full-thickness midline mass at the anterior one third of the tongue. She also had type 1 vWD managed by a hematologist. Prothrombin and partial thromboplastin time were prolonged. Von Willebrand disease, factor 8 and von Willebrand ristocetin cofactor activity were low. Before the operation, the patient was administered cryoprecipitate (6 units), feniramin hydrogen maleate (intravenous) and paracetamol 500 mg (oral). The lesion was totally excised with number 15 blade under local anesthesia with lidocaine hydrochloride 40 mg and epinephrine hydrochloride 0.025 mg. There was minor bleeding during the operation which was controlled with bipolar cauterization and suture ligation. Histopathological diagnosis was hemangioma (Figure 1, 2).

The wound healed in two weeks. No complication such as bleeding or infection occurred after the operation.

DISCUSSION

Hemangiomas are the most common benign tumors of vascular origin of the head and neck region.[4] The possible sites of occurrence in the oral cavity are lips, tongue, buccal mucosa, and palate. Despite its benign origin and behavior, it is always of clinical importance to the otolaryngologist and requires appropriate management.[5] Some of childhood hemangiomas may have spontaneous regression but some of these lesions need urgent treatment.[6]

Particularly in a patient who has vWD, a hemangioma may cause life-threatening bleeding. It is treated urgently and carefully.

Cryoprecipitate, derived from plasma, has been used to treat hemophilia A and vWD. It has been suggested before surgical intervention in vWD patients.[7] We administered cryoprecipitate and paracetamol before surgery and this prevented bleeding during the operation. Aspirin is contraindicated in patients with vWD, because of inhibitory effect of platelet aggregation, and may exacerbate vWD.[8]

Embolization, excision, cryotherapy, sclerotherapy, plasma knife surgery and laser photocoagulation methods have been used for the treatment of hemangiomas.[3] Because of the small lesion, we preferred excision with a cold knife. We did not observe any significant bleeding during surgery or after the operation. The wound healed in two weeks.

Von Willebrand disease is a common cause of bleeding disorder. Excision of the hemangioma of the tongue in that disease may cause life-threatening bleeding. The patient who has VWD should be evaluated carefully before surgery.

Declaration of conflicting interests

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