Venolymphatic Malformation of the Tongue and Response to Sclerotherapy Using Sodium Tetradecyl Sulfate

Dear Editor

A 28-years-old healthy woman presented with a bluish swelling on the dorsum of tongue, which was present for almost two decades. Her parents noticed bluish discoloration of tongue when she was approximately 8 years old, which enlarged at puberty to a pea-sized swelling. The lesion increased progressively to the present size in the last 2–3 years. Examination revealed a bilobed swelling, 3 × 4 cm in size [Figure 1A], present over the dorsum of middle one-third of the left half of the tongue. The swelling was soft, partially compressible with no thrill or bruit. The patient was majorly asymptomatic with no pain or difficulty in mastication but gave a history of occasional bleeding while chewing hard food. Rest of the mucocutaneous examination and physical examination was normal. Her hematological investigations and blood biochemistry revealed no abnormality. High-resolution ultrasonography (USG) showed an anechoic lesion on the dorsum of the tongue on the left side. The superficial, irregular, anechoic, cystic component reached up to the tongue surface. Lesion had a vaguely echogenic deeper component, which showed mild vascularity on color Doppler imaging [Figure 2A]. Contrast-enhanced computed tomography (CECT) of the face showed a non-enhancing, low attenuation mass of water density (Hounsfield Unit was 10-12 HU) with a deeper well-defined, irregular, homogenously enhancing component [Figure 2B] with enhancement persisting in the delayed phases as well. These findings were consistent with combined venolymphatic malformation (VLM) of tongue.

The patient was treated by sclerotherapy using intralesional injections of 3% sodium tetradecyl sulfate (STS). The lesion was injected with insulin syringe at multiple sites, using approximately 0.3–0.4 mL of solution. Postinjection, manual compression using gauze piece was given for 3 min. She reported transient pain and burning sensation immediately after the injection that resolved spontaneously in the next 15 min. No delayed adverse effects were noted. Two such injections at 3-week interval led to complete resolution of the lesion [Figure 1B] with no recurrence after 1 year.

Vascular malformations are structural malformations of vascular development. Depending on their predominant vessel type, they have been categorized as arterial, venous, lymphatic, or mixed types.[1] On the basis of flow characteristic, vascular malformations are generally subdivided into slow-flow lesions (venous, capillary, and lymphatic malformations) and fast-flow lesions (arterial malformations as aneurysm and arteriovenous malformation).[2] VLMs are tumorlike lesions combining dysplastic lymphatic and venous vessel structures.[3] Of the slow-flow lesions, venous malformations (VMs) are the most common ones accounting for two-third of all such cases. VMs and VLMs are generally present at birth and grow in size proportionately to the body growth. Approximately 40% of the lesions occupy head and neck region. Symptoms may range from asymptomatic to mild discomfort and pain, bleeding, disfigurement, and functional impairment. Clinically, it may not be possible to differentiate VMs and VLMs. Diagnosis of VLM is

Figure 1: A 28-year-old woman with VLM of the tongue. (A) Pre-op. (B) Post-op after two doses of STS
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generally based on USG and color Doppler imaging as was apparent in this case. CECT or magnetic resonance imaging is useful to correctly delineate the extent of the lesion, which is required to plan the treatment. A specific feature of VM is the presence of phleboliths that was not observed in this case. It is possible that many cases of VLMs are treated as VMs in the absence of radioimaging. VLMs do not involute spontaneously and require surgical intervention or treatment with sclerosing agents. Various treatment modalities include supportive therapy such as compression, sclerotherapy, lasers, and surgery or combination of the two modalities. Sclerotherapy is economical and efficacious and forms the first-line nonsurgical intervention, it is not widely used. The treatment has to be tailored for individual lesion based on the size, location, symptoms, and proximity to the vital structures. Multiple sessions may be required. A variety of sclerosing agents have been used, namely 95% ethanol, polidocanol, and STS. Other agents include sodium morrhuate, Picibanil (OK-432), bleomycin, and intravenous doxycycline. We report this case to document the following: (1) occurrence of VLM of the tongue and (2) excellent response of sclerotherapy with STS.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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