Case Report

Intravascular papillary endothelial hyperplasia of the masseter muscle

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ABSTRACT

Intravascular papillary endothelial hyperplasia (IPEH) is a rare, benign, non-neoplastic vascular lesion which usually occurs in the skin, subcutaneous tissue, and mucous membrane originating from any place where the vessel is located. It is most commonly seen in the head and neck region, while oral cavity involvement is rare. The clinical significance of IPEH is its histopathological similarity with Kaposi sarcoma, intravenous atypical vascular proliferation, spindle-cell hemangioendothelioma, and particularly with angiosarcoma. Surgical resection is the main treatment option. Herein, we present a case of IPEH of the masseter muscle which was resected successfully.

Keywords: Head and neck tumor, intravascular papillary endothelial hyperplasia, masseter muscle, Masson's tumor.

Intravascular papillary endothelial hyperplasia (IPEH) is a rare benign tumor originating from the skin, subcutaneous tissue, and mucosa in any part of the body.[1] It is most commonly seen in the head and neck region, while it may involve the extremities and trunk.[2]

Although the exact etiology is still unknown, reactive proliferation of the endothelial cells with papillary formations related to an abnormal process of organization in the preexisting vessel or vascular malformation or within an organizing hematoma are thought to be the main reasons of IPEH.[1,2]

The pathological diagnosis can be difficult, as it may be confused with other malignant and benign vascular tumors or malformations, such as angiosarcoma, angiomatous meningioma, cavernous hemangioma, arteriovenous angioma, and capillary hemangioma.[3,4]

In this report, we present a case of IPEH of the masseter muscle which was resected successfully.

CASE REPORTS

A 32-year-old male patient was admitted to the otolaryngology clinic of Liv Hospital with a complaint of swelling on the left cheek for two months. The mass was painless and there was no swallowing difficulty. His medical history was unremarkable with no history of alcohol, smoking, or trauma. On physical examination, a painless, well-rounded, and approximately 2-cm

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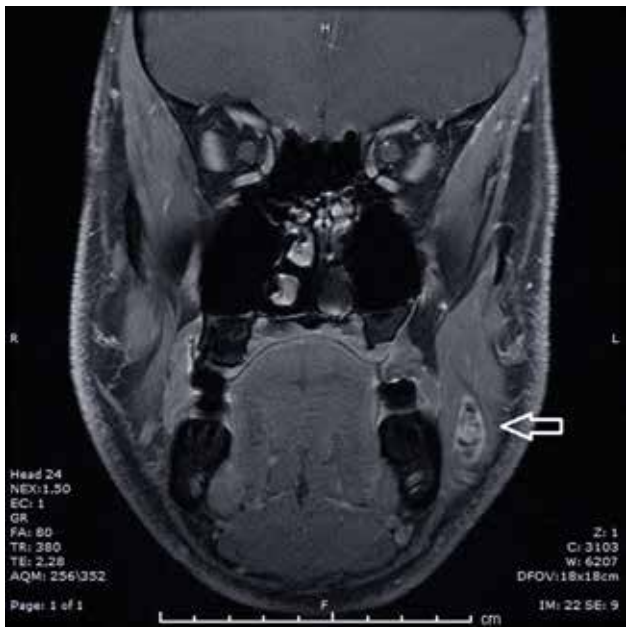

Figure 1. *Coronal contrast-enhanced magnetic resonance imaging showing a lesion approximately 20×12×8 mm in anteroinferior of masseter muscle.*

mass was revealed on the left masseter muscle. Other ear, nose and throat (ENT) examination findings were normal.

Doppler ultrasonography (US) showed a heterogeneous 13×6-mm solid lesion in the left masseter muscle with a significant certain

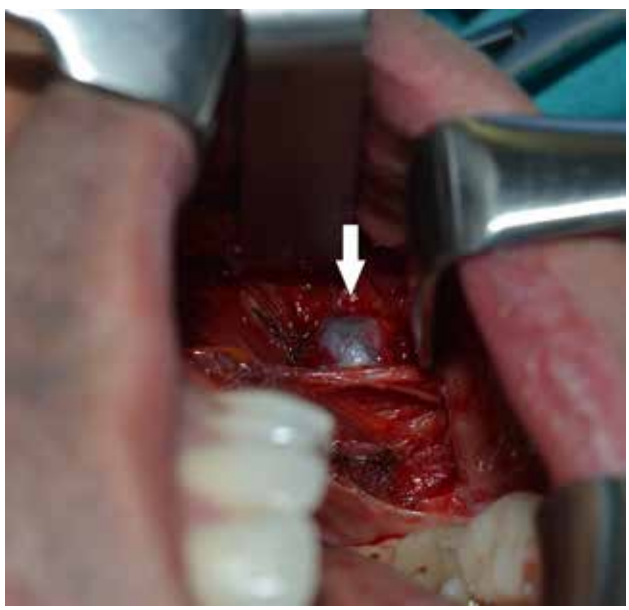

Figure 3. *Bluish color of the mass.*

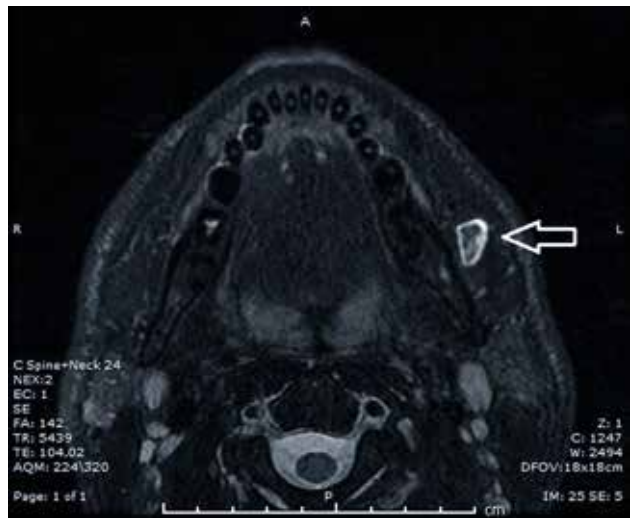

Figure 2. *An axial non-contrast magnetic resonance imaging view of the mass.*

vascularization. Contrast-enhanced magnetic resonance imaging (MRI) showed a lesion approximately 20×12×8-mm in the anteroinferior of the left masseter muscle, heterogeneous hyperintense in the T2A, isointense in the T1A, and a dense heterogeneous contrast enhancement in the post-contrast examination (Figures 1, 2). Biopsy was not performed due to vascular imaging findings and surgical excision was planned. A written informed consent was obtained from the patient.

The operation was performed under general anesthesia through intra-oral approach after a 4-cm linear incision for reaching the masseter muscle (Figure 3). The tumor was completely dissected from the surrounding tissues

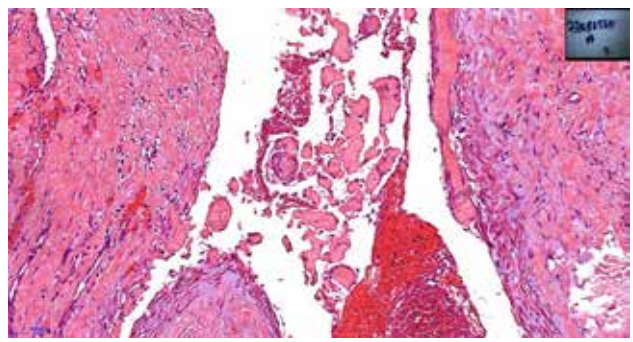

Figure 4. *Intravascular thrombosis and papillary endothelial hyperplasia. H-E ×150.*

with a solid margin and sent to pathological examination. On histopathological examination of the resected mass, numerous papillae with hyaline cores surrounded by monolayered endothelial hyperplasia was observed with an organized thrombus (Figure 4). The endothelium showed no malignant features such as anaplasia or necrosis. Immunohistochemical staining with CD34 and CD31 showed positive reactivity with a complex vascular network of the tissue mass. Based on these findings, a diagnosis of IPEH was made.

Postoperative recovery was unremarkable. Outpatient follow-up for two weeks and six months has not shown any evidence of recurrent or residual disease.

DISCUSSION

Intravascular papillary endothelial hyperplasia was first identified by the French pathologist Pierre Masson in 1923 through the detection of endothelial papillary proliferation in the lumen of the inflamed hemorrhoidal plexus and named as vegetant intravascular hemangioendothelioma.[5] Since then, it has been described under many different names and finally IPEH has been increasingly used, as it explicitly describes the disease pathology.[6]

Until now, three subtypes of IPEH have been described in the literature. The primary (pure) form is a lesion arising within a dilated vessel, most often venous (55.8%). The secondary (mixed) form arises in hemangiomas, arteriovenous malformations, aneurysms, pyogenic granulomas, and lymphangiomas (39.9%). The third variant is an extravascular lesion arising within hematoma (4.3%).[3]

Intravascular papillary endothelial hyperplasia is an unusual benign vascular lesion which accounts for 2% of all soft tissue and skin vascular tumors with a predilection for the head and neck, trunk, and upper extremities.[7] It is rarely seen in the oral cavity and most frequently involved in the lower lip (40.5%), followed by the tongue, buccal mucosa, upper lip, gingiva, labial commissure, mandibular vestibule, hard palate, floor of the mouth, and angle of mouth.[6]

The clinical presentation of IPEH usually includes as a soft-to-firm, painless mass, sometimes tender, ranging in size from 0.5 to 1.8 cm with a reddish-blue color to the overlying skin or mucous membrane. In our case, there was no discoloration of the facial skin and buccal mucosa, as the mass was located in the masseter muscle. It was painless, smooth-surface mass in the left masseter region, which was able to be only noticed by palpation. It is important to differentiate these tumors from parotid located tumors. The MRI, computed tomography, and Doppler US are helpful tools to establish the diagnosis. The tumors of the masseter muscle are quite rare. Schwannoma and hemangioma are the most common tumors located in the masseter muscle.[8] In our case, IPEH was located in the left masseter muscle. To the best of our knowledge, this is the second case of IPEH of the masseter muscle reported in the literature.[9]

The incidence of IPEH in women is slightly higher than men with a ratio of 1.14:1. The mean age varies from nine months to 79 years.[10] While the exact pathogenesis of IPEH is still unclear, an unusual form of thrombus organization following a trauma is considered to play a role. A history of minor trauma has been reported in 4 to 10% of cases. In oral lesions, Tosios et al.[10] reported the contribution rate of minor trauma to be 7%. Unlikely, our case was a young adult male with no history of trauma.

Histopathological image of IPEH is considerably pathognomonic, and a thorough knowledge of this lesion is of utmost importance for clinicians and pathologists for the differential diagnosis. Histological differential diagnosis of IPEH includes angiosarcoma, hemangioma, mucocele, traumatic fibroma, lymphangioma, pseudomyogenic hemangioendothelioma, Kaposi sarcoma, intravenous atypical vascular proliferation, spindle-cell hemangioendothelioma, malignant papillary angioendothelioma, and intravenous pyogenic granuloma.[3-6,7,9-11] In the majority of cases, it can be difficult to distinguish IPEH from angiosarcomas. The histopathology of IPEH shows intravascular proliferation of numerous papillae with close association with the thrombotic material, does not show tissue necrosis, cellular pleomorphism, and a high mitotic rate. Angiosarcomas, on the other hand, usually exhibit an invasive growth pattern,

infiltration, cellular pleomorphism, mitoses, and necrotic foci. The definitive diagnosis of the lesion is necessary to prevent further aggressive and inappropriate treatment. The prognosis of IPEH is mostly very good and the removal of the mass within intact limits is adequate in the treatment. The majority of patients remain recurrence- or metastasis-free.

In conclusion, IPEH should be considered in patients presenting with swelling in the face, subcutaneous tumors, masseter muscle masses and, particularly parotid masses. Radiological imaging modalities such as US, CT, and MRI are helpful to establish the diagnosis. Hemangioma and schwannoma tumors are frequently observed in the masseter muscle tumors. Although IPEH of the masseter muscle is rare, it should be always kept in mind in the differential diagnosis.

Declaration of conflicting interests

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