



Case Report

ULTRASOUND CHARACTERIZATION OF A LYMPHATIC MALFORMATION OF THE TONGUE: A CASE REPORT

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ABSTRACT

Lymphangioma is an infrequent benign lesion in the family of vascular malformations classified as lymphatic malformation (LMs). These, in most cases, have onset in the first years of life but can sometimes occur during growth. Clinical severity can range from the absence of symptoms to mild discomfort to the point of determining bone malformations of the entire head and neck area. The diagnosis can be addressed from the clinical examination, which the histopathological examination will confirm. The imaging tests used in this area are computed tomography, magnetic resonance, and ultrasounds. Ultrasounds can be used with both intraoral and extraoral approaches. The intraoral approach is best suited when the mass is on the tongue. A case report of a lymphangioma of the back of the tongue is presented. The clinical and histological characteristics of the lesion were analyzed. An ultrasound examination (US) was performed to determine the characteristics of the lesion. Information obtained in the US was useful as a diagnostic aid. Intraoral ultrasonography can be a promising diagnostic aid in the context of LMs of the tongue.

KEYWORDS: *Lymphangioma, tongue, clinical and histopathological examination, ultrasound*

INTRODUCTION

Lymphangiomas are infrequent lymphatic malformations (LMs), usually involving the head and neck region. This group of lesions has been known and described for a long time, but the pathogenesis is still unclear, and it is not easy to categorize and classify them (1). These lesions can be described as congenital malformations of the lymphatic system with different sizes of lymphatic channels and cystic spaces (2). The head and neck region is most affected (75% of cases)

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and represents about 5% of benign tumors in children (3). The skin is the most common site, but oral lesions are frequent. In particular, the dorsum of the tongue is reported as the major affected site, followed by lips, buccal mucosa, soft palate, and floor of the mouth (4). The reported incidence ranges from 1:2500 to 1:16000, with no difference in gender distribution. Morbidity and mortality are related to the size and site of the lesion (2, 5). The prognosis is generally good, and the clinical course can be asymptomatic in some cases, although larger LMs may provide airway obstruction and death in children (6).

In many cases, the early development of these masses at the lingual level and in all oral districts can cause alterations in the growth of the lower third of the face and considerable orthodontic problems (7). In addition, LMs can enlarge with an upper respiratory infection or hormone changes at puberty (8). In the past, LMs were classified according to their clinical presentation and site of onset. In 1982, histological and cellular characteristics were integrated into the clinical classification. Furthermore, this classification divided the vascular anomalies into tumors and malformations, improving the management of this category of lesions (9). In this work, LMs were classified as low-flow vascular malformations of lymphatic origin and subcategorized into macrocystic, microcystic, and mixed (10). Subsequent classifications adopted this nomenclature in 1996 and in the classification proposed and approved at the International Society for the Study of Vascular Anomalies (IVVSA) workshop in 2014 (11).

The latter intended to evolve the understanding of the biology and genetics of vascular and lymphatic malformations (12). From a histological point of view, LM consists of lymphatic channels surrounded by endothelial cells. The channels' size and shape are variable, determining the classification (13). Histopathologic features consist of lymphatic vessels with marked dilatations (7), and the vessels can infiltrate the closest soft tissues and exhibit lymphoid aggregates. The endothelium is slight, with fluid rich in proteins and lymphocytes; secondary haemorrhage may occur in the lymphatic vessels. Lymphatic fluid, red blood cells, lymphocytes, macrophages, and neutrophils characterize the lymphatic spaces. Surrounding connective tissue consists of loose fibrotic tissue and inflammatory cells (14). Usually, the tumor is superficial, shows a lobulated surface, and may appear translucent. When the lesion is deeper, it appears as a soft consistency mass with irregular edges. Superficial lesions may also have a papillary appearance, with no changes in colour in the surrounding healthy mucosa, while the deeper lesions appear as nodules without showing a significant change from the surrounding mucosa (4). The differential clinical diagnosis for LMs in the oral cavity includes all the exophytic lesions of the tongue, from granular cell tumors to lingual thyroid and mesenchymal tumors. All vascular malformations of mesenchymal origin should be considered.

Also, pyogenic granuloma should be inserted in the differential diagnosis. For these reasons, the histopathological analysis will be decisive for the final diagnosis as the listed lesions often present a clinical history similar to LMs (15). Therefore, surgical excision is the treatment of choice (13, 16). Some diagnostic tools can be used as diagnostic aid, particularly computed tomography (CT) and magnetic resonance imaging (MRI) (17). Ultrasonography (US) can be used for prenatal diagnosis of large masses in the head and neck region and other areas. However, some authors emphasize the importance of the intraoral US in managing oral LMs (16, 17). This study presents a case of tongue lymphangioma using the US for diagnostic characterization.

CASE REPORT

A thirteen-year-old male presented at the Oral Medicine and Pathology Division of the University of Campania, "Luigi Vanvitelli", for a swelling on the left dorsum of the tongue. The patient's parents reported that the lesion had appeared for about one year following infection with a fever of the upper respiratory tract. The young patient reported that he had always had discomfort in that tongue area. Intraoral examination showed harmonic growth of the bone structures with a mild Angle first-class malocclusion and crowding of the lower incisors, all teeth present in the arch (except wisdom teeth), excellent oral hygiene, and no caries. The lesion appeared as an elastic mass of about 3x1.5 cm, occupying the dorsum's entire central portion, extending posteriorly up to the left lingual tonsil. The surface appeared raised and characterized by nodules alternating with a papular appearance and micro-vesicles without any significant change in surface texture or colour.

Despite the size of the lesion, the patient denied any pain and reported being able to breathe and speak normally, with mild discomfort in swallowing. US examination was performed with an 18MHz linear hockey stick probe in B-mode. One focus was placed in correspondence with the lesion, and the harmonic mode was set. In the US, the lesion appeared hypo-iso-echoic with a thickened hyperechoic epithelium on the surface and an anechoic band that delimited it from the

underlying connective tissue. The thickness measured was about 2.5 cm. In the context of the lesion, well-defined and anechoic ovoid areas were present, well delimited by a hyperechoic border to form septa. An acoustic shadow was present under those areas (Fig. 1). In power Doppler mode, it was possible to observe the flow of microvessels under the lesion. The incisional biopsy was performed. Histological analysis confirmed the clinical diagnosis of lymphangioma. The patient decided to wait for surgical therapy due to the extent of the lesion and the paucity of symptoms. An annual follow-up was scheduled.

DISCUSSION

Despite being a benign lesion, managing intraoral LMs is challenging for the clinician. In particular, the clinical examination can address the diagnosis, but other information can be obtained with imaging tools. CT and MRI are the gold standards, especially in surgical planning; however, some information may be lost in smaller lesions. MRI imaging, for example, could be affected by metal artefacts in dentures or dental reconstructions. Furthermore, CT and MRI are less appropriate for superficial lesions without obtaining clinically relevant images with these techniques (17). US, on the other hand, is a non-invasive and reproducible technique, which allows obtaining real-time images of the scanned tissues with appropriateness comparable to the techniques mentioned above, especially in terms of size and in lesions <5mm.

Furthermore, some studies suggest that in the US, it is possible to obtain images comparable to macroscopic histopathological sections (18, 19). In the context of LMs, US is used with an extraoral approach, but in some lesions, in certain areas, such as the tongue, this may not be as effective as an intraoral approach: air spaces can attenuate acoustic waves, and the bone could represent an obstacle for the penetration of soundwaves. Therefore, the intraoral approach is preferable to diagnose tongue mass lesions (17, 20, 21). In this case report, images obtained by the intraoral US provided numerous additional information compared to the examination alone:

- a) it has provided information on the thickness of the lesion and its relationship with the surrounding tissues;
- b) it was possible to determine the ultrasound characteristics in terms of echogenicity and homogeneity;
- c) it was possible to recognize some structures in the context of the mass, such as the presence of lobules and well-defined anechoic areas;
- d) the ultrasound artefacts gave information on the density of the tissues;
- e) an anechoic capsule has been identified, which separates the mass from the surrounding context and could correspond, on a histopathological level, to the thick fibrous capsule surrounding the perimeter of LMs (Fig. 1).

It is precisely this characteristic, according to Sugawara et al, that allows for distinguishing a benign lesion from a neoplastic lesion from an ultrasound point of view (17). Furthermore, even if the authors use a different ultrasound system, the US analysis returns an image with similar characteristics, such as the presence of the ovoid anechoic areas within the lesion.

CONCLUSIONS

Diagnosis and management of LMs represent a challenge for the clinician. Most of the time, the diagnosis is directed by clinical examination, but imaging tools such as the US can provide valuable additional information. Further studies are needed to establish the feasibility of using the intraoral US to manage oral LMs.

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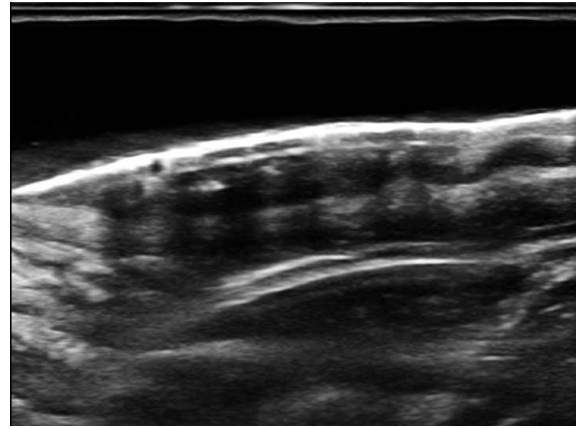


Fig. 1. *Acoustic shadow present in anechoic ovoid and hyperechoic border areas*

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