

## CASE REPORT

### Lichenoid uvula mass as a rare cause of hot potato voice and progressive dysphagia

Ramaprabah KANDIAH<sup>1</sup>, Azliana AZIZ<sup>1</sup>, Nik Fatin Amirah NIK MIN<sup>2</sup>, Muhd Rifqi RAHIM<sup>3</sup>, Norzaliana ZAWAWI<sup>2,4</sup>, Khairil Amir SAYUTI<sup>3</sup>, Sanjeevan NADARAJAH<sup>5</sup>, Irfan MOHAMAD<sup>1</sup>

<sup>1</sup>Department of Otorhinolaryngology-Head & Neck Surgery, School of Medical Sciences, Universiti Sains Malaysia, Health Campus, Kota Bharu, Kelantan; <sup>2</sup>Department of Pathology, School of Medical Sciences, Universiti Sains Malaysia, Health Campus, Kota Bharu, Kelantan; <sup>3</sup>Department of Radiology, School of Medical Sciences, Universiti Sains Malaysia, Health Campus, Kota Bharu, Kelantan; <sup>4</sup>School of Dental Sciences, Universiti Sains Malaysia, Health Campus, Kota Bharu, Kelantan; <sup>5</sup>Department of Otorhinolaryngology, Trafalgar Memorial Hospital, Oakville, Ontario Canada

#### Abstract

Lichenoid uvula mass is an unusual entity and the clinicopathologic entities are important for the diagnosis. In this case report, we report an elderly lady presented with dysphagia and “hot potato voice” due to a lichenoid uvula mass. Apart from muffled voice, the other associated symptoms are dysphagia, sore throat, globus pharyngeus and snoring. She was a non-smoker, and not on any medications except for hypertension and dyslipidaemia. Apart from the enlarged uvula, other parts of the oral cavity, oropharynx and larynx were normal. She has no other signs such as cutaneous lesions. The mass was excised under general anaesthesia followed by resolution of symptoms. Awareness of this entity, the clinical and histopathological features should be highlighted to differentiate it from other diagnosis.

**Keywords:** Hot potato voice, dysphagia, lichenoid, uvula mass.

#### INTRODUCTION

The uvula is a midline soft tissue appendage formed by the fusion of two halves of the soft palate. The lesions involving uvula mass are divided into neoplastic, non-neoplastic and congenital in origin.<sup>1</sup> Neoplasm lesions are further subdivided into malignant and benign lesions from epithelial and mesenchymal origin. On the other hand, non-neoplastic lesions are infective or inflammatory, traumatic, cystic and developmental in nature.<sup>2</sup> Elongated, big and floppy uvula will be symptomatic and present with obstructive symptoms, irritation and voice change. Surgery is the mainstay of treatment in symptomatic patients as per our case report.

#### CASE REPORT

A 72-year-old female with underlying hypertension, hyperlipidaemia and degenerative spine disease presented with a history of

progressive dysphagia to solid food and foreign body sensation over the throat during swallowing for five months duration. She also had voice changes, snoring and dyspnoea on lying flat which was relieved when changed to lateral position. There was an absence of constitutional symptoms like loss of weight, appetite, alopecia or skin changes. There was no history of changes in current medications intake and she was not taking additional herbal medications or over-the-counter prescriptions. She is a non-smoker. Examination revealed she had a muffled voice. There was an enlarged broad-based uvula. The uvula was smooth surfaced, firm and not inflamed (Fig. 1). Other oral cavity examinations were unremarkable. We aspirated and sent for cytology but unfortunately, it was non-representative material. Computed tomography (CT) scan of the neck revealed a well-defined non-enhancing hypodense lesion in the uvula measuring 1.3 cm × 1.5 cm × 2.5

\*Address for correspondence: Irfan Mohamad, Department of Otorhinolaryngology-Head & Neck Surgery, Universiti Sains Malaysia Health Campus, 16150 Kubang Kerian, Kelantan, Malaysia. Tel: 609-7676420. Fax: 609-7676428. Email: [irfankb@usm.my](mailto:irfankb@usm.my)



FIG. 1: Intraoperative appearance of the uvula mass, showing a pale, smooth-surfaced, elongated uvula touching the base of the tongue, with a solid mass at the base of uvula.

cm, (Fig. 2), which was suggestive of a benign lesion. As the patient had progressive dysphagia, we then proceeded with excision of the mass under general anaesthesia. Postoperatively, her voice improved, she had no velopharyngeal insufficiency and symptomatically relieved from globus pharyngeus and dysphagia.

Grossly, it consists of a piece of greyish tissue partly covered by mucosa. No bullae, ulceration or surface discoloration was noted

(Fig. 3). Microscopically, it shows mucosal tissue covered by stratified squamous epithelium. The epithelium shows acanthosis with focal areas of hyperkeratosis and parakeratosis with ballooning degeneration of the basal cells. Area of bullae formation is also noted. There is moderate to dense lymphoplasmacytic infiltration at the subepithelial area with numerous ectatic lymphatic channels are also seen. The inflammatory infiltrates are also seen at the

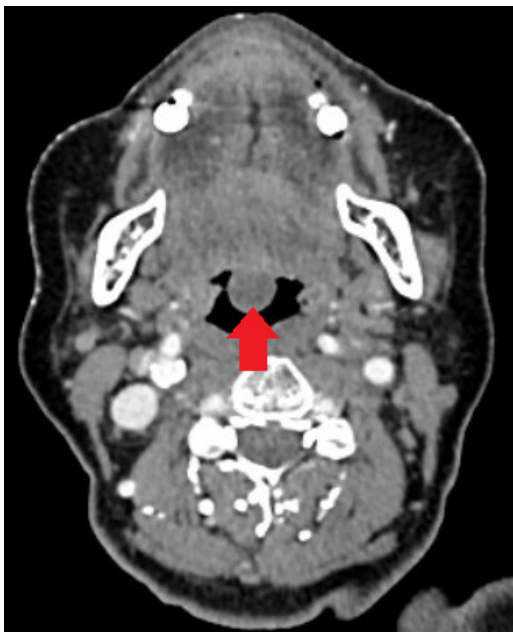


FIG. 2: CT scan neck revealing the uvula mass (red arrow) showing the well-defined non-enhancing hypodense lesion in the uvula.



FIG. 3: Specimen consists of an irregular piece of pink-greyish tissue patchily covered by mucosa.

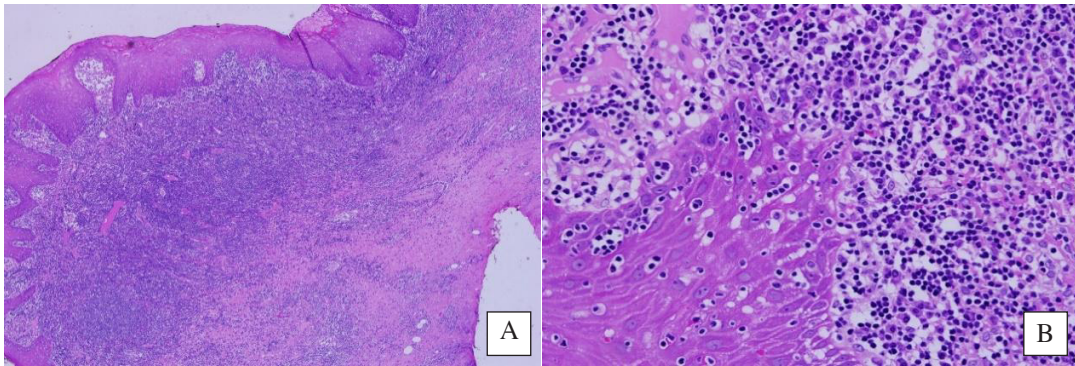


FIG. 4: Uvula lined by stratified squamous epithelium with areas of acanthosis and hyperkeratosis. An intense band-like lymphocytic infiltrate is seen beneath the epithelium (H&E, 4x) (A). High power view of the inflammatory infiltrates composed mainly of lymphocytes with lymphocytes exocytosis (H&E, 40x)(B).

periglandular area and also scattered in between the interstitial stroma. Occasional histiocytic aggregates are seen. The inflammatory infiltrates are composed of a polyclonal population on B and T cell markers (CD20 and CD3 staining). Macrophages and occasional multinucleated giant cells are also seen within the lymphatic channels. No fungal bodies or acid-fast bacilli were seen. There was also no evidence of dysplastic keratinocyte or malignancy seen (Fig. 4 & 5). The histological features are suggestive of the oral lichenoid lesion (OLL). The immunofluorescence study did not proceed in this case as it was a formalin-fixed specimen.

## DISCUSSION

Uvula plays an important role in articulation assisting in speech and prevents nasal regurgitation of food during swallowing. It is composed chiefly of racemose glands and

connective tissue, covered by mucous membrane and containing a slender prolongation of the azygos uvulae muscle in its upper part.

The differential diagnosis for hot potato voice is infections like acute epiglottitis, tonsillitis, peritonsillitis, peritonsillar and retropharyngeal abscess, acute angioedema, caustic ingestions, patients with tumours of the oropharynx or foreign body aspiration. Types of lesions from epithelial tissue are like myoepithelioma and pleomorphic adenoma. Examples of lesions from mesenchymal tissue are lipoma, fibroma, neurofibroma, schwannoma, angiokeratoma, angioleiomyoma and granular cell tumour.<sup>2</sup> Abscess, peripheral giant cell granuloma, pyogenic granuloma and squamous papilloma are infective or inflammatory types of lesions. The traumatic types are denture induce hyperplasia, fibroepithelial polyp and neuroma. Lesions such as the dermoid, epidermoid and mucocele and

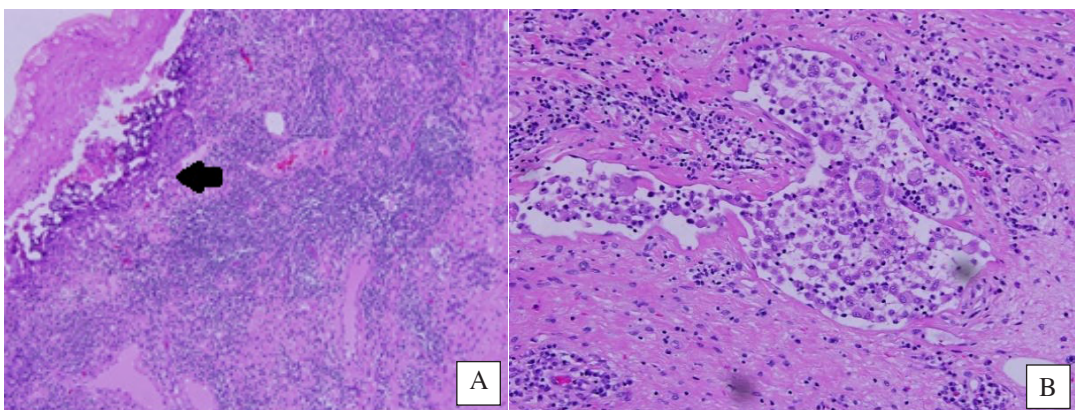


FIG. 5: A low power view of bullae formation (arrow) with separation of epithelial layer from underlying mucosa(H&E,4x)(A). High power view of inflammation within the lymphatic channels consist of macrophages, multinucleated giant cells and lymphocytes (H&E, 20x)(B).



haemangioma and lymphangioma are from cysts and developmental category respectively.<sup>2</sup>

An elongated or enlarged uvula causes irritation to the throat, dysphagia, sore throat, cough, hawking and vomiting. Some patients complain of foreign body sensations, tickle in the throat and gagging.<sup>3</sup> In severe cases, clinical features suggestive of respiratory distress<sup>4</sup> were reported.

In patients with sleep apnoea syndrome, there will be an increase in the amount of uvula muscle, the protein content and anaerobic metabolic activity.<sup>5</sup> Submucosal oedema and minor salivary glands hypertrophy were noticed in patients with sleep apnoea syndrome.<sup>6</sup>

Fine needle aspiration and cytology (FNAC) or aspirate is done first, to determine the nature of the swelling, whether it is benign or malignant prior to deciding the mode of treatment. When there is a mucosal lesion or ulcer, it is wise to take a punch biopsy and aspirate, on the contrary, is opted if the lesion is deep and has intact mucosa.

Imaging plays an important role in diagnosis and intervention. CT of the neck with the contrast of our patient, revealed a well-defined non-enhancing hypodense lesion in the uvula (about 20 Hounsfield Unit), causing narrowing of the oropharynx. No calcification or adjacent fat streakiness and no cervical lymphadenopathy. Hence, the radiological findings concluded that it is a benign cystic lesion and non-neoplastic in origin. Confirmatory diagnosis would be based on tissue diagnosis.

Diseases involving the uvula are very rare. There have been few isolated cases reporting oedema of the uvula with a variety of aetiologies such as allergy, drug reactions, infection of the upper respiratory tract, trauma or vascular alterations however, over half of the cases studied were found to be idiopathic. In the idiopathic group, usually patient presented with no symptoms or nonspecific such as urticaria, angioedema or anaphylaxis.<sup>7</sup> In this case, the patient presented with symptoms of dysphagia with no known predisposing insult to the oral area.

A lichenoid reaction is described histologically as an inflammatory disease indicated by the presence of basal cell damage in the form of cell death or apoptosis, liquefaction or the vacuolar change associated with a band-like mononuclear inflammatory cell composed of activated T-lymphocytes, macrophages and dendritic cells.<sup>8</sup> Oral lichenoid disease (OLD) is a group of pathological processes with an

immunologic component that usually presents as white papules on the oral mucosa, a gross feature otherwise absent in our case. In most cases, no known cause can be identified in this disease.<sup>9</sup>

Two main subtypes can be classified as OLD: oral lichen planus (OLP) and oral lichenoid lesions (OLL).<sup>9</sup> Oral lichenoid lesions can be further subdivided into Amalgam restoration, Drug-related OLL, OLL in chronic graft versus host disease and OLL, unclassified whereby there is the presence of erythematous change but it does not fulfil criteria for "classic" Lichen planus.<sup>10</sup> There is a lack of clear histopathological criteria in differentiating these two subtypes, however; some authors report criteria such as depth of inflammatory infiltrates, perivascular inflammation, inflammatory cells such as polymorphonuclear neutrophils and presence of epithelial dysplasia. However according to Alberdi-Navvaro *et al.* in his research shows no relation between the different clinical and histopathological subtypes of oral lichenoid disease subtypes.<sup>9</sup> In our case, a gross specimen is composed of uvula tissue partly covered by mucosa. The mucosa appears unremarkable with no white lines, bullae formation or ulceration. Microscopic examination revealed histopathological features of lichenoid reaction with no dysplasia or feature of malignancy noted. The lichenoid tissue reaction pattern which is seen in this case includes dense band-like inflammatory cell infiltrates at the juxtaepithelium junction, vacuolar basal cell degeneration, subepidermal bullae and lymphocytes exocytosis. In our case, no evidence of Civatte bodies, disordered maturation or dysplasia was identified.

Some studies have also discussed the relation between the histopathological aspect and clinical parameters such as clinical lesion of the biopsy showed that presence of thinning of the epithelium in erythematous lesion compared to papular lesions.<sup>11</sup> In another study, documented greater keratinization in reticulo-papular biopsies and predominant neutrophilic infiltration atrophic erosive lesions.<sup>12</sup> Recently, however, these clinical parameters have been suggested as possible modifying factors of the histopathological aspects. Thus, both clinical and histopathological assessment is necessary in order to obtain a proper diagnosis.

*Authors' contribution:* RK involved in drafting the initial manuscript. AA and SN involved

in writing the clinical history and patient's progression. MRR and KAS provided the radiological input. NFANM and NMN provided the microbiological content. IM provided critical input and finalize the draft.

*Conflict of interest:* The authors declare no conflict of interest.

## REFERENCES

1. Boveri T. Concerning the Origin of Malignant Tumours by Theodor Boveri. Translated and annotated by Henry Harris. *J Cell Sci* 2008;121(Supplement 1):1-84.
2. Al-Khateeb T. Benign Oral Masses in a Northern Jordanian Population-a Retrospective Study. *Open Dent J* 2009; 3(1):147-53.
3. Daghistani KJ. Conditions of the uvula: a 14 years' experience. *Auris Nasus Larynx* 2000; 27(3):261-4.
4. Shott SR, Cunningham MJ. Apnea and the elongated uvula. *Int J Pediatr Otorhinolaryngol* 1992; 24(2):183-9.
5. Sériès FJ, Simoneau SA, St Pierre S, Marc I. Characteristics of the genioglossus and musculus uvulae in sleep apnea hypopnea syndrome and in snorers. *Am J Resp Crit Care Med* 1996;153(6):1870-4.
6. Cimino A, Speciale R, Gallina S, *et al.* Morphologic and ultrastructural changes of soft palate in patients who underwent palatopharyngoplasty. *Acta Otorhinolaryngol Ital* 1995;15(2 Suppl 47):18-23.
7. Alcoceba E, Gonzalez M, Gaig P, Figuerola E, Auguet T, Olona M. Edema of the uvula: etiology, risk factors, diagnosis, and treatment. *J Investig Allergol Clin Immunol* 2010;20(1):80-3.
8. Khudhur A, Di Zeno G, Carrozzo M. Oral lichenoid tissue reactions: diagnosis and classification. *Expert Rev Mol Diagn* 2014;14(2):169-84.
9. Alberdi-Navarro J, Marichalar-Mendia X, Lartitegui-Sebastian M, Gainza-Cirauqui M, Echebarria-Goikouria M, Aguirre-Urizar J. Histopathological characterization of the oral lichenoid disease subtypes and the relation with the clinical data. *Med Oral Patol Oral Cir Bucal* 2017;22(3):307-13.
10. Hiremath SK, Kale AD, Charantimath S. Oral lichenoid lesions: Clinico-pathological mimicry and its diagnostic implications. *Indian J Dent Res* 2011;22(6):827-34.
11. Karatsaidis A, Schreurs O, Helgeland K, Axéll T, Schenck K. Erythematous and reticular forms of oral lichen planus and oral lichenoid reactions differ in pathological features related to disease activity. *J Oral Pathol Med* 2003; 32(5):275-81.
12. Fernández-González F, Vázquez-Álvarez R, Reboiras-López D, Gándara-Vila P, García-García A, Gándara-Rey JM. Histopathological findings in oral lichen planus and their correlation with the clinical manifestations. *Med Oral Patol Oral Cir Bucal* 2011;16(5):e641-6.