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Case Report
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Anatomical Variations of the Uvula: Considerations based on Two Cases

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ABSTRACT

Bifid uvula is a malformation reported as a variation of the uvula that is rarely observed in the population. The aim of the present study is to describe 2 patients with uvula malformation: a bifid uvula was identified in a 20-year-old male patient, and aplasia of the uvula was identified in a 24-year-old female patient, in the Dental Clinic located at Faculdade de Odontologia de Bauru, Universidade de São Paulo (FOB-USP). In the two cases reported, no comorbidities were associated with the uvula malformation, and the patients did not present any functional alterations. Thus, despite the fact that both were cases of anatomical variations, it is important to differentiate uvula malformations from the common structure, since, in some cases it is involved in other syndromes or genetic disorders.

Keywords: Bifid uvula; Cleft palate; Uvula; Anatomy.

Introduction

The uvula is a structure found in the posterior edge of the middle soft palate, and it is an appendicular structure resembling a teardrop (Fig. 1, 2A). Indeed, the term "uvula" means "small grape" in Latin because of its morphology.[1–4]

Among its functions, the uvula prevents the soft palate from invading the nasopharynx or mouth in situations such as coughing or sneezing.[2] Moreover, histological studies show that the uvula has glands that are able to produce a large volume of saliva.[5] Due to its location in an area prone to high antigen exposure, caused by exposure to air and food, this structure is also supposed to have a local immune protective function, with a notable count of leukocyte traffic.[6]

Bifid uvula is a malformation, a variation that is rarely observed in the population.[7] The term bifid indicates a partial or total bifurcation (Fig. 2B, C).[8,9] Additionally, abnormalities of the uvula and its variations are described as notched, bifida or broad (which represents cases in which the uvula is extremely wide).[10] The most severe consequence of bifid uvula is paralysis of the glossopharyngeal nerve, which is responsible for the movements of the palate and innervates the pharyngeal muscles.[11]

The incidence of bifid uvula is very uncertain, depending on the population, the country, and other associated anom-alies. Its incidence has been described as 1 in every 80 Caucasian individuals,[12] and it occurs in 0.36 to 1.54 per 1,000 births in Brazil.[13] However, a higher incidence in certain ethnicities has also been reported, which indicates the possibility that bifid uvula can have a genetic factor.[4,14]

Bifid uvula can also be present in cleft palate patients, as described by the Calnan triad: posterior palatal notch, zona pellucida, and bifid uvula.[10] Bifid uvula can also be one of the signs of Loeys-Dietz syndrome, a rare autosomal-dominant connective tissue disorder that is also associated with cardiac problems.[15] In this context, the accurate identification of anatomic abnormalities and/or variations in the clinical practice, such as bifid uvula, is useful to help clinicians identify other diseases or syndromes in the diagnosis.

In the present study, the authors aimed to describe one case of bifiduvula (Fig. 2B) and one case of aplasia of the uvula (Fig. 2D), found in patients in the Dental Clinic at Faculdade de Odontologia de Bauru, Universidade de São Paulo (FOB-USP).

Case Report

Case 1: A, 20-year-old Caucasian Brazilian male patient pre-sented to the dental clinic at FOB-USP, in the city of Bauru, state of São Paulo, Brazil, to perform a routine health care visit.

During the examination, the dentist noticed an

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unusual anatomical structure located in the soft palate region, a bifurcation and duplicity of the uvula (Fig. 3). The dentist contacted the Department of Anatomy of FOB-USP to report the case. After a complete clinical examination of the oral cavity, the patient was diagnosed with the anatomical variation known as bifid uvula. Moreover, the color and consistence of the tissues surrounding the structure were compatible with those of a healthy individual. The patient did not report difficulties in swallowing or any functional alterations in speech.

Case 2: A 24-year-old Caucasian Brazilian female patient presented to the dental clinic of FOB-USP to perform a routine health care visit. The dentist observed an unusual morphologic feature on the soft palate. After contacting the Department of Anatomy, a complete clinical examination of the oral cavity was performed. The patient was diagnosed with aplasia of the uvula (Fig. 4). The color and consistence of the tissues surrounding the structure were compatible with those of a healthy individual. The patient was referred to the Department of Speech Therapy for examination due to the possibility of oropharyngeal dysphagia. However, the patient did not present any functional alterations.

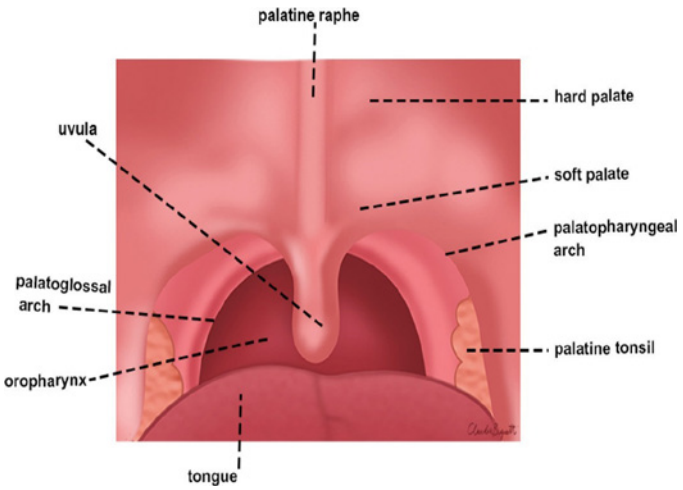

Figure 1. Common anatomy of the uvula and its location and adjacent structures.

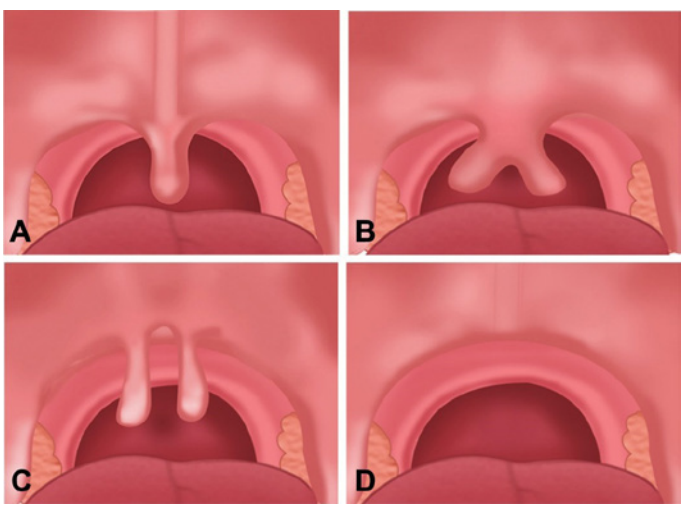

Figure 2. The uvula and its morphological anatomical variations: (A) common uvula; (B)bifid uvula; (C) notched uvula; and (D) aplasia of the uvula.

Discussion

The present study is a report of a case of bifiduvula (Case 1) and a case of aplasia of the uvula (Case 2), with no other associated alterations, such as cleft palate. The prevalence of bifid uvula is frequently associated with cleft palate, and it ranges from 1.5% to 10%.2 However, isolated cases of bifid uvula are rarely reported.[2]

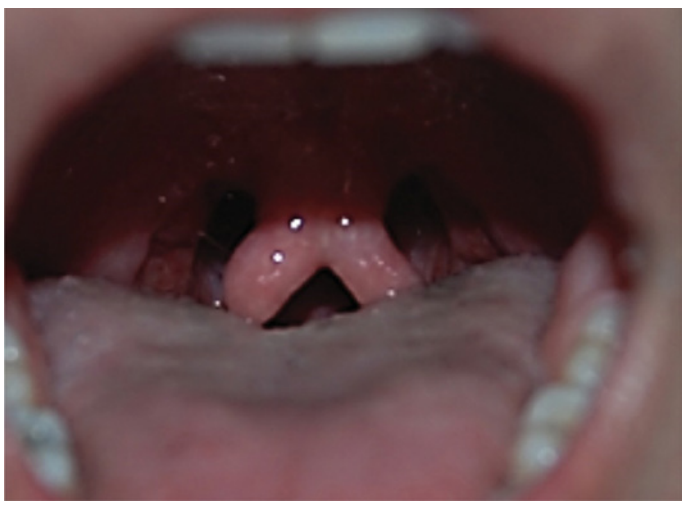

Figure 3. Case 1: patient with bifiduvula.

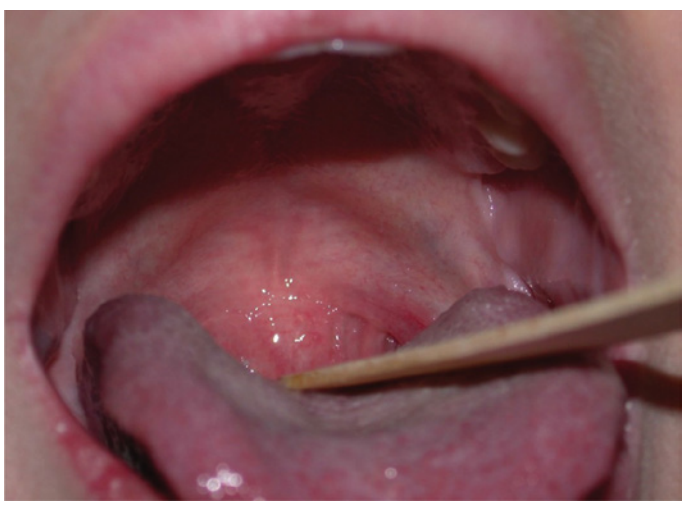

Figure 4. Case 2: patient with aplasia of the uvula aplasia.

The anatomical variations presented in the uvula are extremely rare, and usually occur due to genetic factors in the following proportion: for every 50 families that present the trait, 20% to 40% of the parents show the same anatomical variation.[2] In a study[9] that analyzed 1,206 children, only 6 (0.49%) presented bifid uvula. In spite of the evolution in the identification of the environmental and genetic risk factors associated

with cleft palate and the consequent occurrence of bifiduvula,the true etiology in most cases remains unexplained.[16]

Bifid uvula is described as more common in males than in females.[2] In the present case report, case 1 was a male patient with bifid uvula, and case 2 was a female patient with aplasia of the uvula. Other studies[17,18] show that the presence of cleft palate is more common in females, and a higher incidence of bifid uvula is possible. However, in a study by Sales *et al*,[9] out of 1,026 individuals analyzed, only 6 had bifid uvula, and 5 of those patients were male.[9] In a study by Archer *et al*,[10] 62.3% of the patients with variations of the uvula, from bifida to notched, were male as well.

Interestingly, the two patients in the present case report did not present oropharyngeal dysphagia or other functional alterations, and other systemic comorbidities associated with this local anatomical variation were not observed as well. In a study conducted by Achalli *et al*,[2] one of the patients had total absence of the uvula, a variation known as aplasia of the uvula, similar to case 2 in the present report. Aplasia of the uvula was also reported in six cases in a study conducted by Gopal *et al*.[4] However, in the case reported by Achalli *et al*,[2] the patient had difficulty speaking and swallowing due to his small tongue. In situations in which the uvula is surgically removed, Back *et al*[5] described complications such as pharyngeal dryness. However, those complications are not observed in patients who apparently were born with aplasia of the uvula.

Finally, the diagnosis of this anatomical variation, in a routine visual examination of the oral cavity, may not be as simple as it seems.[9] The viscosity of the mucus in the oral cavity may contain a bifid uvula, or leave it grossly bifid, making diagnosis difficult.[9]

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