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Letter to Editor

Pathology Section

Hypermobility of Tongue: A Clinical Curiosity

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BY-NC-ND

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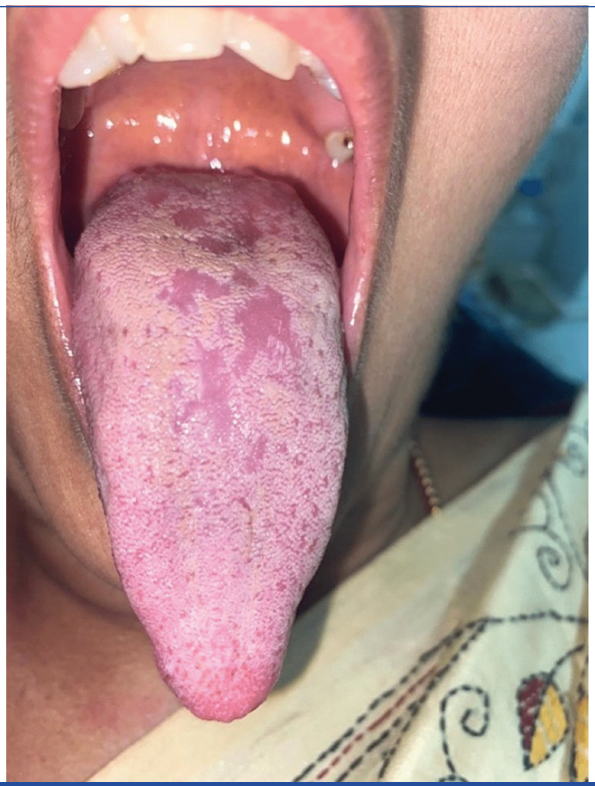
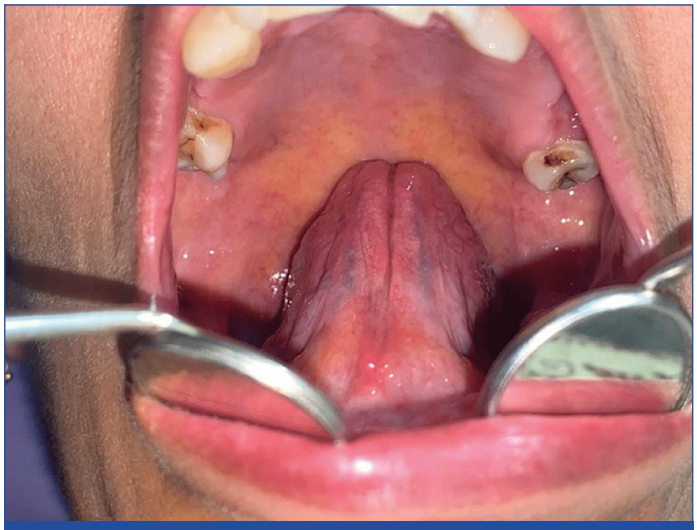
Dear Editor,

A 43-year-old female patient presented to private dental clinic with a chief complaint of accumulated dirt and stains on teeth for the past two years. The patient expressed aesthetic discomfort and reported experiencing bad breath for approximately six months. No relevant medical, surgical, or habitual history was disclosed. During the general examination, the patient appeared well-oriented, co-operative, moderately built, and overall normal. Intraoral examination revealed generalised extrinsic stains, plaque, and calculus. No evidence of gingival pockets or recession was observed, leading to a diagnosis of generalised chronic gingivitis. The lingual surface of the tongue exhibited the absence of a lingual frenulum, resulting in an "abnormally moving tongue." The dorsal surface of the tongue appeared depapillated and coated with food debris. Consequently, a scaling procedure was recommended. During scaling, the patient was instructed to retract her tongue to clean the lingual surface of the lower incisors. Unexpectedly, the patient folded her tongue back and placed it behind the uvula region. When asked to protrude the tongue forward, it extended beyond the chin [Table/Fig-1,2].



[Table/Fig-1]: Dorsal surface of the tongue.
Forward protrusion of the tongue showing Gorlin's sign-tongue touching the chin

Deformities in the lingual frenulum are typically of systemic, developmental, or genetic origin. Various conditions, such as Ehlers-Danlos Syndrome (EDS), Infantile Hypertrophic Pyloric Stenosis (IHPS), and non syndromic ankyloglossia, are commonly associated



[Table/Fig-2]: Ventral surface of the tongue.
Abnormal tongue fall back- touching the uvula due to absence of lingual frenum

with the absence of the lingual frenulum [1-3]. IHPS occurs in three out of 1000 live births, with a male predilection at a ratio of 3:1. It also shows a high familial predisposition. Infants between 3 and 6 weeks of age start exhibiting symptoms such as increased episodes of vomiting after each feed [2]. However, since the patient in present case did not report any history of bowel obstruction or forceful vomiting, IHPS was ruled out. EDS is a connective tissue disorder that is usually of genetic origin. The prevalence of EDS is 1 in 400,000 people, with no prominent gender prevalence observed [3]. The syndrome is divided into six types: classical, hypermobile, vascular, kyphoscoliotic, dermatosparaxis, and arthrochalasis. It involves mutations in multiple genes, including COL5A1, COL5A2, COL3A1, COL1A1, PLOD1, TNXB, and COL1A2. EDS affects multiple organs and can be life-threatening. One of its characteristics is the absence of the lingual frenulum. Other structures affected by this syndrome include joints, skin, and blood vessels. A hypermobile tongue is an important clinical feature, present in approximately 71% of the cases [4]. Existing literature supports a hypermobile tongue as a unique diagnostic criterion for EDS. In present case, there were no signs or symptoms associated with EDS other than a hypermobile tongue. The ability to touch one's nose tip or chin with the tongue is referred to as Gorlin's sign. It is common in 5% of the general population [5] and in 50% of individuals with EDS [6]. Interestingly, Gorlin's sign was positive in the presented case as well.

The first case series on hypermobile tongue was reported in 1742 [6]. Petit brought excessive tongue mobility to the medical world's attention in three children, two of whom died due to suffocation caused by it. In 1845, Fairbairn reported a similar case where the patient died due to airway obstruction. In 1877, Henning also reported a case of obstructing hypermobile tongue associated with mortality. Later, in 1958, Schiff reported a case without any symptoms during a routine Ear, Nose and Throat examination in the

Navy [6]. Cinar F et al., presented a case of a healthy 45-year-old Turkish man who could pass the tip of his tongue beyond the uvula and into his nasal cavity to clear nasal secretions [1]. They referred to this condition as "idiopathic hypermobile tongue". Cincik H et al., discovered a hypermobile tongue while examining a 16-year-old Turkish boy who came for evaluation of loud snoring [7]. The boy's tongue was capable of reaching the posterior nasal cavity. In 2012, Surendran S et al., reported a case series of 12-year-old and 16-year-old with hypermobile tongue that extended beyond the uvula during routine examinations [8]. More recently, in 2019, Felemban R and Mawardi H reported a case of congenital absence of the lingual frenulum in a 21-year-old female patient without any associated syndrome [9]. Idiopathic hypermobile tongue does not usually cause any functional disturbances. Hence, they are often ignored, but in some cases, they can result in choking and even death. These rare conditions have significant diagnostic value and can help prevent mortality caused by ignorance.

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