

## Short Case Report

# Influence of a short lingual frenulum and a lack of tonicity of the lingual and suprahyoid muscles in apneic teenager: report of a case

Manfredi Bertrand<sup>\*</sup>, Berquet Alexandre

Oral Surgery and Oral Medicine Department, Hospitall François Mitterrand, Dijon, France

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**Abstract – Observations:** A short lingual frenulum and a lack of tone of the lingual and supra-hyoid muscles could influence a OSA in a teenager, refractory to conventional treatment. The therapy is based on surgery and rehabilitation with polysomnographic surveillance. **Comments:** The aggravating factors were to be noted during the treatment. No article in the literature put this relationship into perspective. **Conclusion:** The results obtained were inconclusive. This line of research could interest the scientific community in order to guide the physiopathological knowledge of the disorder and to optimize the possibilities of therapeutic response in case functions.

## Introduction

Currently, the standard treatment for obstructive sleep apnea (OSA) is continuous positive nasal airway pressure (CPAP) therapy [1], which is often poorly tolerated, particularly by younger patients. Mandibular advancement orthoses (MAO) represents an alternative treatment; however, its use is limited in edentulous or growing patients. Furthermore, oral hygiene and dietary measures are always recommended in patients undergoing treatment with MAO. Here we present a case of a possible influence of a short lingual frenulum with a lack of lingual and suprahyoid muscle tone on the existence or severity of OSA in a teenager aged 16 years.

## Observation

A 16-year-old patient had been referred by her pulmonologist, who was treating her for OSA, for treatment with MAO, following a refusal to undergo CPAP therapy.

She had moderate OSA [apnea/hypopnea index (AHI) at 10.3] that was unmanaged, severe obesity (BMI, 37 kg/m<sup>2</sup> or 96 kg for 161 cm), depression, and school phobia; furthermore, she had several voluntary drug intoxications between 2016 and 2017. Active smoking, estimated at 2 pack-years (PY), as well as allergies to pollen; cat hair; and grasses, which had been monitored and treated, was noted.

Her treatments included benzodiazepine anxiolytics, an H1 antihistamine, antiepileptic drugs, and nicotine replacement therapy.

An extraoral examination revealed extensive lumbar and cervical protrusions and extended craniofacial posture.

Intraorally, the tongue was in the lower and posterior position, with indentations on its lateral edges; its mobility was significantly limited during elevation and protraction by the presence of a short lingual frenulum (Figs. 1 and 2). Palpation of the lingual and suprahyoid muscles showed a weaker than normal muscle tone.

The patient exhibited atypical swallowing with the contraction of the labial muscles.

Regarding OSA, she was found to be snoring with pauses in breathing almost every night while maintaining a subjectively qualitative sleep. During the day, the patient had frequent problems related to concentration and morning headaches, but without drowsiness. The Epworth Sleepiness Scale score was estimated at 13/24.

A lingual frenectomy, followed by a 3-month rehabilitation performed by a physiotherapist, represents the main framework of our therapeutic management. Polysomnography (PSG) performed before the start of treatment acted as the control. A second control PSG was to be performed at the end of the physiotherapy sessions. A significant improvement in PSG findings after the treatment was hypothesized.

Lingual frenectomy (Fig. 2) was performed under local anesthesia using a classical technique (for short lingual frenulum, simple dissection following the lingual muscular

\* Correspondence: [myjbertrand@yahoo.fr](mailto:myjbertrand@yahoo.fr)



**Fig. 1.** Lingual protrusion limited by a short lingual frenulum (right photograph) and the protrusion immediately after surgery (left photograph).



**Fig. 2.** Presence of a short lingual frenulum limiting the elevation of the tongue (right photograph) and a postoperative photograph of the lingual frenectomy (left photograph).

plane, in the raised state, guiding the procedure until there is a significant freedom of movement), thus, making it possible to release the constraints preventing lingual mobility. Physiotherapy began after 15 days of healing. It focused on the rehabilitation of swallowing and phonation as well as toning exercises of the floor and tongue [2]. The number of sessions was left to the discretion of the physiotherapist, but the duration was fixed at 3 months.

During the first weeks following the frenectomy, the patient's friends and family and her pulmonologist noted a decrease in snoring.

The control PSG, delayed to 7 months postoperatively instead of the originally planned 3 months postoperatively due to school holidays and the patient not coming for follow-ups, showed AHI to be 15/h, i.e., an increase of 4.7/h compared with baseline PSG. The Epworth Sleepiness Scale score was estimated at 4/24, indicating a marked improvement in sleep quality.

## Comments

The purpose of the comprehensive management was to provide physiological, or even etiological, treatment to a patient who did not wish to undergo the usual treatment for OSA and who could not benefit from a mandibular advancement

orthosis because she was still growing. The results obtained in the present case do not allow us to draw a conclusion because of several elements.

First, the patient gained approximately 15 kg during the time between the two PSGs, between January and September 2018, with no changes in the body and a metabolic and non-athletic profile. This represents a major confounding factor.

Second, the Epworth Sleepiness Scale is a questionnaire containing eight questions about the subjective elements of everyday life; between the two PSGs, the period of school holidays, which is spent outside the school, is not the same as the active scholastic period regarding the circadian rhythms. This may explain the low score obtained in September 2018.

Third, the follow-up observations by the physiotherapist are not reliable in this patient who proved to be less compliant with follow-up visits, in general.

Fourth, the use of an H1 antihistamine and a benzodiazepine (independent sources of sleepiness and sleep pattern disruption), whose dosage and adherence varied, represent as many confounding factors as possible to the relationship that was sought to be established in the present case.

In the literature, there are no studies on a direct association between tongue mobility disorders and OSA. However, ankyloglossia can cause, among other things, malocclusion as well as a projection of the alveolar processes and thus of the basal bone and their relationships, influencing the dysfunction of the adjoining strap muscles. These phenomena are all the more significant during the growth phase. These elements could be related to OSA [3].

## Conclusion

On the basis of biological knowledge, it is plausible that anatomical and functional factors promote OSA onset or aggravation. However, the present case does not allow us to draw any preliminary conclusions corroborating this statement, but it may help gain the attention of the scientific community to study this relationship.

**Conflicts of interests:** The author declare that they have no conflicts of interest in relation to this article.

## References

1. Vecchierini MF, Laaban JP, Desjobert M, Gagnadoux F, Chabolle F, Meurice JC, *et al.* Stratégie thérapeutique du SAHOS intégrant les traitements associés? *Rev Mal Respir* 2010;27(Suppl 3):S166–S178.
2. Breton Torres I, Fournier M. Rééducation du temps buccal de la déglutition salivaire et des dyspraxies orofaciales. Bilan et rééducation – *EMC-Odontologie* 2016;12:1–14; 23–490-D-15.
3. Guilleminault C, Huseni S, Lo L. A frequent phenotype for paediatric sleep apnoea: short lingual frenulum. *ERJ Open Res* 2016;2:pii.00043-2016.