

# AN UNUSUAL CASE OF ANKYLOGLOSSIA WITH SUBLINGUAL SIALOLITHIASIS

Head and Neck Surgery

Submitted : 01.04.2020

Accepted : 14.07.2020

Published : 14.07.2020

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## Özet

### DİL ALTI SIALOLİTİAZİSLE BİRLİKTE OLAĞANDIŞI BİR ANKİLOGLOSSİ OLGUSU

Ankiloglosi, anormal derecede kısa bir lingual frenulum ile karakterize konjenital bir oral anomali. Ankiloglosi yetişkinlerde bebeklere göre daha az görülür. Sialolithiasis genellikle yaşamın üçüncü ve beşinci yılları arasında görülür. Submandibular bez en sık tutulan tükürük bezidir. Bu literatürde erişebildiğimiz sialolithiasis'e eşlik eden ilk ankiloglossia olgusudur. Ankiloglosi uzun süre tedavi edilmezse, bu durum submandibular sialolitiazise neden olabilir. Bu nedenle ankiloglosi tedavisinde submandibular sialolitiazis düşünülmalıdır.

**Anahtar kelimeler:** Ankiloglossi, submandibuler, sialolitiazis, submandibuler sialolitiazis

## Abstract

### AN UNUSUAL CASE OF ANKYLOGLOSSIA WITH SUBLINGUAL SIALOLITHIASIS

Ankyloglossia is a congenital oral anomaly characterized by an abnormally short lingual frenulum. Ankyloglossia is less common in adults as compared to infants. Sialolithiasis usually occurs between the third and fifth decades of life. The submandibular gland is the most frequently involved salivary gland. This is the first case of ankyloglossia accompanying sialolithiasis in literature which we could access. If ankyloglossia is left untreated for a long time, this condition may cause submandibular sialolithiasis. Therefore in ankyloglossia treatment, submandibular sialolithiasis must be considered.

**Keywords:** Ankyloglossia, submandibular, sialolithiasis, submandibular sialolithiasis

## Introduction

The incidence of ankyloglossia has been reported between 0.02% and 10.7% [1]. In adolescents or adults the prevalence ranges from 0.1% to 2.08%. Ankyloglossia affects males more commonly [2]. Ankyloglossia is a congenital oral anomaly characterized by an abnormally short lingual frenulum [3]. Ankyloglossia is characterized by short vertical length of the lingual frenulum, which may result in a varying degree of decreased tongue tip mobility [4]. Ankyloglossia is less common in adults as compared to infants and some authors have hypothesized that ankyloglossia improves with age [3]. The clinical consequences of ankyloglossia include infant breastfeeding difficulties, speech disorders, various mechanical and social issues related to disability of tongue movement, and orthodontic anomalies [4]. About 79 % of salivary calculi are in the submandibular glands and 34% of them are localized in the duct of Wharton's, 57% in the hilum and 9% inside the gland [5,6]. Sialolithiasis usually occurs from the third to fifth decades of life, and is slightly more often in males [7]. The submandibular gland is the most frequently involved salivary gland because of the longer length of its major duct with a narrow orifice, as well as the nature of the submandibular gland secretion, which can have a consistency as thick as a gel [8]. This gel becomes a framework for deposition of salts, which leads to development of calculi [9]. Small calculi of approximately 3–7 mm are common either in the submandibular duct or gland. Single giant calculus of size 33 mm [9] 10 mm in the submandibular duct or multiple calculi of 7 in number of an average diameter of 7–8 mm within the submandibular gland are extremely rare 10.

## Case Report

A 17-year-old teenager presented to our clinic with complaints of a sublingual swelling for 3 months and pus discharge from the left Wharton's papilla of 2 weeks duration (Figure 1).



**Figure 1**

Figure 1: Intraoral view of the left sublingual swelling and short lingual frenulum.

The patient's history revealed previous recurrent left submandibular swelling and pain after eating. In oral examination, there was a short lingual frenulum and a swelling of 1x1 cm dimension in the left sublingual region. On the cervical CT scan, a hyperdense lesion of 11 mm diameter was observed in the left sublingual region (Figure 2).



**Figure 2**

Figure 2: Axial section of CT with left intraductal sialolith.

The patient underwent frenuloplasty and transoral excision of the calculus in the left Wharton's duct. (Figure 3)



**Figure 3**

Figure 3: Frenuloplasty was performed, followed by intraoral excision of the ductal sialolith.

The dimensions of the calculus were approximately 20x10 mm (Figure 4).



**Figure 4**

Figure 4: Submandibular sialolith

## Discussion

Predisposing factors for submandibular sialolithiasis are salivary stasis, increased alkalinity of the saliva, infections of the oral cavity, physical trauma to the submandibular duct or gland, smoking, and diuretic use [10,11]. Ankyloglossia or tongue-tie is a short attachment of the lingual frenulum. The lingual frenulum connects the ventral tongue to the floor of the mouth. In the case of a short lingual frenulum, forward and downward pressure of the tongue to the mandible may cause growth of the mandible in a more prognathic manner and different consequences on face development [4,12,13]. Also, the increased pressure of the tongue may lead to a decrease or even halt of the submandibular salivary flow.

The incidence of ankyloglossia is low in adults [2]. Submandibular sialolithiasis may be seen in children also with low incidence, since relatively long time is required for the formation of a salivary calculus, and sublingual papilla is very small in children, which blocks the ingress of foreign material required for the initial nidus formation. However, a calculus is formed more easily in adults than in children [9]. Although submandibular sialoliths are usually asymptomatic, pain and swelling which increase with food anticipation may be the cardinal signs and symptoms [9,14,15]. The presented case was a teenager who had ankyloglossia and an intraductal sialolith. This is infrequent in teenager.

Although CT, magnetic resonance imaging, and X-ray can be used to diagnose sialolithiasis, the most effective diagnostic imaging modality is sonography [16]. In our case CT was preferred.

Surgical repair may be considered in patients with a tight lingual frenulum, as outlined below, and a history of associated feeding, speech, or mechanical/social difficulties. The diagnosis of ankyloglossia is confirmed by characteristic examination findings. The tongue is often heart-shaped at its tip. Tongue protrusion is limited, and the tip may not extend past the lower lip. When the tongue protrudes past the teeth, the midportion of the tongue typically rolls or curls forward, but the tip itself is drawn inferiorly by the frenulum with little forward extension. Tongue elevation is also characteristically impaired. Frenuloplasty and frenotomy are the most commonly used surgical procedures for treatment of ankyloglossia [17]. The primary treatment of submandibular sialolithiasis consists of stimulation of saliva production and massage of the gland. The other treatments include submandibular

gland excision, sialoendoscopy or transoral removal of the calculi [6,18]. Surgical treatment for the submandibular calculi is chosen according to location and palpability of the sialolith. Intraoral removal is more suitable for sialoliths which are palpable in the mouth and are located in the distal duct at a distance of no more than 2 cm from the ostium of Wharton's duct [19]. Intraoral excision of the ductal calculi is managed with linear incision on the mucosa along the duct on the place of the calculus bulge and is removed<sup>10</sup>. In our case, firstly frenuloplasty was performed, followed by intraoral excision of the ductal sialolith.

To our knowledge, this is the first case of ankyloglossia accompanying sialolithiasis in the accessible literature. If ankyloglossia is not treated for a long time, this condition may cause submandibular sialolithiasis. Therefore in ankyloglossia treatment, submandibular sialolithiasis must be considered.

## References

1. Walsh J, Links A, Boss E, Tunkel D. Ankyloglossia and lingual frenotomy: national trends in inpatient diagnosis and management in the United States, 1997-2012. *Otolaryngol Neck Surg.* 2017;156(4):735-740.
2. Ballard JL, Auer CE, Khoury JC. Ankyloglossia: assessment, incidence, and effect of frenuloplasty on the breastfeeding dyad. *Pediatrics.* 2002;110(5):e63-e63.
3. Messner AH, Lalakea ML. Ankyloglossia: controversies in management. *Int J Pediatr Otorhinolaryngol.* 2000;54(2-3):123-131.
4. Jang S-J, Cha B-K, Ngan P, Choi D-S, Lee S-K, Jang I. Relationship between the lingual frenulum and craniofacial morphology in adults. *Am J Orthod Dentofac Orthop.* 2011;139(4):e361-e367.
5. Zenk J, Constantinidis J, Al-Kadah B, Iro H. Transoral removal of submandibular stones. *Arch Otolaryngol Neck Surg.* 2001;127(4):432-436.
6. Juul ML, Wagner N. Objective and subjective outcome in 42 patients after treatment of sialolithiasis by transoral incision of Warthon's duct: a retrospective middle-term follow-up study. *Eur Arch Oto-Rhino-Laryngology.* 2014;271(11):3059-3066.
7. Parkar MI, Vora MM, Bhanushali DH. A large sialolith perforating the Wharton's duct: review of literature and a case report. *J Maxillofac Oral Surg.* 2012;11(4):477-482.
8. Jadu FM, Jan AM. A meta-analysis of the efficacy and safety of managing parotid and submandibular sialoliths using sialendoscopy assisted surgery. *Saudi Med J.* 2014;35(10):1188.
9. Trivedi BD. Surgical removal of submandibular gland sialolithiasis in a 9-year-old girl: A case report. *Pediatr Dent J.* 2014;24(2):111-114.
10. Hazarika P, Punnoose SE, Singh R, Arora S. Deep and unusual sialolithiasis of submandibular duct and gland: a surgical dilemma. *Indian J Otolaryngol Head Neck Surg.* 2013;65(4):309-313.
11. Huoh KC, Eisele DW. Etiologic factors in sialolithiasis. *Otolaryngol Neck Surg.* 2011;145(6):935-939.
12. Petit H, Davis W. The role of the tongue in facial development. *J Pedod.* 1986;10(3):199.
13. Defabianis P. Ankyloglossia and its influence on maxillary and mandibular development.(A seven year follow-up case report). *Funct Orthod.* 2000;17(4):25-33.
14. Dalkiz M, Dogan N, Beydemir B. Sialolithiasis (salivary stone). *Turkish J Med Sci.* 2001;31(2):177-179.
15. Alkurt MT, Peker I. Unusually large submandibular sialoliths: report of two cases. *Eur J Dent.* 2009;3(2):135.
16. Kirkendall J. Sialolithiasis: sonographic detection of salivary duct stones. *J Diagnostic Med Sonogr.* 2013;29(1):36-39.
17. Lalakea ML, Messner AH. Frenotomy and frenuloplasty: If, when, and how. *Oper Tech Otolaryngol Neck Surg.* 2002;13(1):93-97.
18. Maresh A, Kutler DI, Kacker A. Sialoendoscopy in the diagnosis and management of obstructive sialadenitis. *Laryngoscope.* 2011;121(3):495-500.
19. Park JS, Sohn JH, Kim JK. Factors influencing intraoral removal of submandibular calculi. *Otolaryngol Neck Surg.* 2006;135(5):704-709.

