



# Spontaneous Venous Thrombosis of the Tongue

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## Abstract

Acute swelling of the tongue most commonly is due to inflammation, abscess or secondary to undesired drug side-effects (e.g. ACE-inhibitors). Less common are benign or malignant growths, foreign bodies or zoonoses. Acute thrombophlebitis as a cause for a significant swelling of the tongue has only been reported twice in literature before. We report the third case, in which Enoxaparin failed to resolve the thrombus. The patient was then successfully treated by unfractionated Heparin. Since knowledge about acute thrombophlebitis of the tongue is rare, we dare to recommend treatment of this condition with unfractionated Heparin, although this recommendation is founded on the successful treatment of only one case.

## Introduction

Acute swelling of the tongue is a rare condition. We describe a case of a 38 year old lady with a swollen tongue. The cause for the swelling was initially unclear, until repeated questioning of the patient revealed that she had been treated in an outside institution for an oral vascular malformation twenty years before [1]. Based on this key information, targeted diagnostics allowed making the diagnosis of an acute thrombophlebitis of the tongue. The patient could then be treated successfully with unfractionated Heparin [2].

## Case Presentation

We would like to report on a case of a 38 years old female patient, who was referred to our department because of left-sided swelling of the tongue (Figure 1a, 1b). She also reported about difficulties in swallowing. The tentative diagnosis was "abscess of the tongue" [3]. There were no signs of dyspnoea, stridor, fever or inflammation. The past medical history was normal; she only had received an operation for the treatment of varicose veins. Clinical examination showed a swollen tongue and an edema of the left-sided floor of the mouth, flexible endoscopy of the larynx did not show any airway compromise [4] (Figure 2). Diagnostics showed that inflammation parameters were in the normal range. Ultrasound imaging of the neck did not show any abnormalities, except the swelling of the tongue, no signs of abscess. An MRI was performed, which confirmed the swelling of the tongue, but no signs of abscess. Further laboratory parameters were negative for rheumatic disease and auto-antibodies as well as for Quincke's edema [5]. Symptomatic therapy was initiated with i.e. Dimetinden (Fenistil), Ranitidin and Prednisolon. This therapy did not result in any improvement. Instead a generalized swelling of the tongue occurred. The tongue was hard on palpation and changed color to a purple shade [6]. The patient was repeatedly asked in more detail about her past medical history, which revealed that she had been treated by laser therapy for a vascular malformation of the soft palate and the floor of the mouth in an outside institution in 1999. With respect to these information's we changed our tentative diagnosis to "vascular malformation

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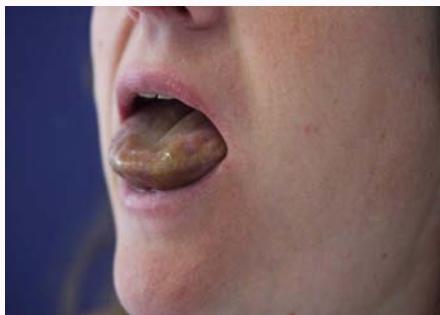
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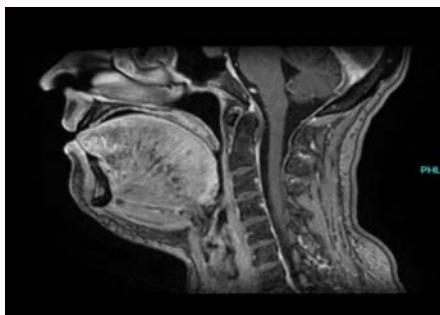
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Figure 1a: The tongue is swollen and shows reduced mobility. Pressure marks from the teeth are evident.



**Figure 1b:** Mobility of the tongue is reduced; it can hardly be moved from left to right and vice versa.



**Figure 2:** Mobility of the tongue is reduced; it can hardly be moved from left to right and vice versa.



**Figure 3:** Findings after therapy with unfractionated Heparin. The swelling has resolved the color of the tongue returns to normal. Mobility of the tongue has significantly improved as well as speech and swallowing.

of the tongue with spontaneous thrombosis". Lab-tests for D-dimers were elevated (1.20 mg/l). Another MRI with MR-angiography was initiated and showed a large venous malformation of the tongue and floor of the mouth with spontaneous venous thrombosis [7]. Factor-V-Leiden-thrombophilia was ruled out by laboratory tests. Initially we tried to treat the thrombosis with Enoxaparin, which did not bring any improvement. Therapy was therefore changed to full-dose heparinisation with unfractionated heparin. Within a few hours the swelling resolved and the patient improved dramatically (Figure 3 and 4).

## Discussion

When confronted with a case of a swollen tongue, in which the cause is unclear, every physician should also consider "spontaneous thrombosis of the tongue" in his differential diagnosis. Such a thrombosis may occur spontaneously or based on a pre-existing vascular malformation, even if previous imaging studies (e.g. MRI-scans) did not show any abnormalities [8]. Vascular malformations



**Figure 4:** Findings after therapy with unfractionated Heparin. The swelling has resolved the color of the tongue returns to normal. Mobility of the tongue has significantly improved as well as speech and swallowing.

develop congenitally and are classified according to their morphology and vascular components. In more than 50% of the cases the initial diagnosis is wrong, even if the correct imaging studies have been performed. Venous malformations are the most common vascular malformations with an incidence of 1-2/10000 and a prevalence of 1%. The tongue is the most common location of venous and arterio-venous malformations in the head and neck region [9]. If there are any signs of vessels abnormalities like phleboliths, superficially ectatic vessels or a significant past medical history, imaging studies should be initiated. Surgeries, like drainage of tentative abscesses should be postponed [10].

In our case thrombosis of the tongue had developed on the basis of a previously undiagnosed venous malformation of the tongue and the floor of the mouth. Enoxaparin therapy did not bring any improvement [11]. When therapy was switched to full-dose therapy with unfractionated heparin, thrombosis of the tongue resolved within a few hours [12].

## Conclusion

There is not much knowledge about spontaneous thrombosis of the tongue. Based on our case, we conclude, that imaging studies should be initiated early on and D-dimers should be included in the lab works. Enoxaparin did not work, only therapy with unfractionated heparin brought a significant improvement.

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