Spontaneous sublingual hematoma: Surgical or non-surgical management?

Evgeni Brotfain, Leonid Koyfman, Schwartz Andrey, Efim Rusabrov, Amit Frenkel, Alexander Zlotnik, Moti Klein

ABSTRACT

Introduction: Spontaneous sublingual hematoma (SSH) is an uncommon, but emergency state in oral cavity. A few series of SSH are seen in the literature. There is no available consensus about clinical strategy and management of such patients. Case Series: The authors present two cases of spontaneous sublingual hematoma after anticoagulation therapy with sintrom (case 1) and warfarin (case 2). The different clinical management was applied in each case (surgical and supportive treatment respectively). The surgical approach was complicated by septic state and multiple organ failure which led to patient’s death. Conclusion: The main priority of physician’s strategy is based on airway protection. Otherwise, there are some different clinical points of view about management SSH (correction of anticoagulation state with blood products/vitamin K or aggressive surgical drainage). Based on previously reported series and our clinical experience (case 2) we consider that surgical intervention has a lot of complications and unsafe for the patients. The non-operative plan of treatment is more benefit and optimal.

Keywords: Spontaneous sublingual hematoma, Anticoagulation, Difficult airway management

INTRODUCTION

Spontaneous sublingual hematoma (SSH) can be caused by anticoagulation or thrombolytic therapy (heparin, warfarin, streptokinase, etc.). In severe cases it may compromise the patient’s airway [1-4]. Treatment protocols for SSH are not evidence-based, so the decision as to whether to opt for surgical or supportive care is based on clinical judgment and experience. In this paper we present two cases of SSH that were managed with different therapeutic approaches and had different outcomes with a review of the relevant medical literature.

CASE REPORT

Case 1: A 77-year-old woman was admitted to the intensive care unit (ICU) of our hospital with a history of marked swelling of the tongue and the submental region. Past medical history was remarkable for open heart surgery due to coronary artery disease; a bypass graft procedure and MVR in 1992. Since then she was on acenocoumarol (sintrom) - 4 mg/day. Her INR level was measured weekly with a target of 2-3. There was no history of recent trauma.
On physical examination the patient appeared to be well oxygenated and ventilated. The blood pressure was 160/60 mmHg, the pulse rate 79/min, and the respiratory rate was 12/min. Rectal temperature was 37.9 ºC. Laboratory findings included Hb - 10.3 g/dl, WBC count – 8x10³/mm³, platelets – 130x10³/mm³ and admission INR was 2.3. The patient’s oral cavity and sublingual space were extremely swollen with involvement of the floor of the mouth (figures 1, 2).

The patient was hoarse and had difficulty opening her mouth.

After initial observation she was transferred to the operating room where the hematoma was evacuated and a tracheostomy was performed. No other source of bleeding was found.

However, during the first three post-operative days, the swelling of the tongue and the edema of the floor of mouth has not resolved. Only supportive therapy was provided, including fluid management, mechanical ventilation, parenteral nutrition, stress ulcer prevention and sedation. Four days after surgery the patient developed septic shock caused by staphylococcus aureus and streptococcus oralis. There was no evidence of line sepsis, bacterial endocarditis or pneumonia. The clinical manifestations of septic shock were ARDS, hypotension and acute renal failure. The patient died approximately four weeks after admission. A consent for publishing this case was obtained from the patient’s family.

Case 2: A 76-year-old woman was admitted to the ICU with progressive respiratory failure caused by sudden-onset oral swelling. She had a history of severe peripheral vascular disease with superior femoral artery and left renal artery stenosis and was treated with warfarin.

On admission she was well oxygenated and ventilated but had a grossly swollen and congested tongue making it difficult for her to open her mouth (figure 3 A, B).

Examination of the oral cavity showed a huge sublingual hematoma. The blood pressure was 176/90 mmHg, the pulse rate was 68 min, and the respiratory rate was 12 min. The rectal temperature was 36.6 ºC. Laboratory data on admission included Hb - 11.1 g/dl, WBC count – 7x10³/mm³, platelets count – 369x10³/mm³, PTT-93 sec, PT immeasurable.

Two hours after admission the patient complained of progressive swelling in her mouth. She was dyspnoic and had difficulty speaking. Given the potential risk of airway compromise an emergency intubation was performed, following which the patient’s condition stabilized with good oxygenation. Vitamin K and fresh frozen plasma (four units) were administered. Four days later the sublingual hematoma was resolved and the swelling subsided without surgical intervention. The patient was successfully extubated and discharged from the ICU five days after admission. Consent for publishing was obtained from the patient.
DISCUSSION

SSH can cause severe airway obstruction and present as a life-threatening condition. Any disease state that requires oral anticoagulants (warfarin, acenocoumarol, etc.) can cause spontaneous bleeds [5] since anticoagulants can antagonize the effect of vitamin K. The complications stem from retropharyngeal, sublingual and parapharyngeal bleeding, which is usually identified by direct visualization of the oral cavity.

The main management priority is to secure and control the patient’s airway. If intubation is impossible because of the size of hematoma, or not safe because of progressive swelling of the oral cavity, emergency tracheostomy may be indicated. In addition, the clinical management of sublingual hematomas includes control of spontaneous bleeding by correction of anticoagulation with or without surgical drainage.

As demonstrated in case 2, the non-operative management of SSH consists of cessation of oral anticoagulation and administration of FFP and/or vitamin K [6, 7].

There have been sporadic reports that the use of leeches to break down the hematoma can yield good results [8]. In general, spontaneous resolution occurs within a few days of cessation of anticoagulation therapy.

Surgical drainage of SSH has been reported in some cases [9]. However, in many cases surgery was performed at a late stage in the absence of spontaneous reabsorption of the hematoma. Surgical management can cause complications including increased swelling with complete airway obstruction and postoperative rebleeding. In our first case, surgical drainage was performed early after admission and was complicated by generalized infection. The nature of the microorganisms identified suggests that they probably originated in the oral cavity.

Since there is a paucity of case reports on the immediate management and follow-up of SSH, we cannot make recommendations as to the optimal treatment strategy, i.e., non-operative clinical management or surgical drainage.

CONCLUSION

We do not recommend surgical drainage because of the risk of serious, even lethal complications such as re-bleeding or sepsis (as in case 1). Moreover, there is no definitive evidence that surgical drainage decreases swelling, reduces ventilator-free days, or improves patient outcome. We believe that optimal management of SSH should include rapid airway assessment and protection, and immediate reversal of anticoagulation therapy.

**********

Author Contributions

Evgeni Brotfain – Substantial contributions to conception and design, acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published
Leonid Koyfman – Substantial contributions to conception and design, acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published
Schwartz Andrey – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Efim Rusabrov – Substantial contributions to conception and design, acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published
Amit Frenkel – Substantial contributions to conception and design, acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published
Alexander Zlotnik – Substantial contributions to conception and design, acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published
Moti Klein – Substantial contributions to conception and design, acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© Evgeni Brotfain et al. 2012; This article is distributed under the terms of Creative Commons attribution 3.0 License which permits unrestricted use, distribution and reproduction in any means provided the original authors and original publisher are properly credited. (Please see www.iijcasereportsandimages.com /copyright-policy.php for more information.)

REFERENCES

1. Lepore ML. Upper airway obstruction induced by warfarin sodium. Arch Otolaryngol 1976;102:505-507.
2. Rosenbaum L, Thurman P, Krantz SB. Upper airway obstruction as a complication of oral anticoagulation therapy. Report of three cases. Arch Intern Med 1979;139:1151-1154.
3. Boster SR, Bergin JJ. Upper airway obstruction complicating warfarin therapy— with a note on
reversal of warfarin toxicity. Ann Emerg Med 1983;12:711-714.

4. Genovesi MG, Simmons DH. Airway obstruction due to spontaneous retropharyngeal hemorrhage. Chest 1975;68:840-842.

5. Gonzalez-Garcia R, Schoendorff G, Munoz-Guerra MF, Rodriguez-Campo FJ, Naval-Gias L, Sastre-Perez J. Upper airway obstruction by sublingual hematoma: a complication of anticoagulation therapy with acenocoumarol. Am J Otolaryngol 2006;27:129-131.

6. Gooder P, Henry R. Impending asphyxia induced by anticoagulant therapy. J Laryngol Otol 1980;94:347-350.

7. Cohen AF, Warman SP Upper airway obstruction secondary to warfarin-induced sublingual hematoma. Arch Otolaryngol Head Neck Surg 1989;115:718-722.

8. Lee NJ, Peckitt NS. Treatment of a sublingual hematoma with medicinal leeches: report of case. J Oral Maxillofac Surg 1996;54:101-103.

9. Scully C, Wolff A. Oral surgery in patients on anticoagulant therapy. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2002;94:57-59.