

Sublingual hematoma as a complication of mandibular midline osteotomy: a case report and literature review

Meghana Anishetty, Radhika Menon, Elavenil Panneerselvam, Shri Krishna Prasanth B, Krishnakumar V. B. Raja

Department of Oral and Maxillofacial Surgery, SRM Dental College, Chennai, India

Sublingual hematoma is a serious complication associated with anterior mandible surgery. It presents considerable risk due to the potential for airway obstruction, which can be life-threatening and necessitates prompt diagnosis and intervention. This case report describes the occurrence of sublingual hematoma following mandibular midline osteotomy and the subsequent management of this condition. Mandibular midline osteotomy is performed to correct skeletal or dental discrepancies in the transverse plane. Sublingual hematoma as a complication of a midline osteotomy has not been previously documented in the literature. This article provides a comprehensive review of the various etiologies associated with sublingual hematoma, along with its management modalities.

Keywords: Airway obstruction / Case reports / Fractures / Hematoma / Mandibular osteotomy / Mouth floor

INTRODUCTION

Sublingual hematoma is characterized by the accumulation of blood within the sublingual and submandibular spaces, resulting in the elevation of the tongue and/or the floor of the oral cavity. This condition can arise from local or systemic factors. Local causes include trauma to the mandible [1,2] or tongue [3,4], as well as surgical procedures [5-7], while systemic causes encompass severe hypertension [8] and the use of anticoagulant medications [9]. Sublingual hematoma is sometimes referred to as “pseudo-Ludwig angina,” a term that reflects the potential for life-threatening airway obstruction associated with the condition [10]. Most documented cases of sublingual hematoma in

the literature are related to dental implant placement [5,6]. The prevention of periosteal perforation is a key step in avoiding such complications [11]. However, this may not be feasible during procedures like osteotomy, particularly midline osteotomy, which necessitates breaching the periosteal envelope to permit the movement of bone segments. Although most case reports have attributed sublingual hematoma to implant placement [12], and preventive strategies have been suggested, no instances have been reported of sublingual hematoma associated with midline osteotomy of the mandible. The aim of this case report is to raise awareness about the potential for this complication in the context of osteotomy and to discuss the preventive measures and management options available.

CASE REPORT

A 43-year-old man presented with a malunited mandibular fracture that necessitated midline osteotomy to correct the mandibular shape and occlusion. The procedure was performed under general anesthesia. A vestibular incision was made to expose the symphysis region, followed by the elevation

Correspondence: Elavenil Panneerselvam

Department of Oral and Maxillofacial Surgery, SRM Dental College, Bharathi Salai, Chennai 600089, India
E-mail: elavenilomfs@gmail.com

How to cite this article:

Anishetty M, Menon R, Panneerselvam E, B SKP, Raja KVB. Sublingual hematoma as a complication of mandibular midline osteotomy: a case report and literature review. Arch Craniofac Surg 2024;25(6):303-308. <https://doi.org/10.7181/acfs.2024.00402>

Received July 10, 2024 / Revised August 17, 2024 / Accepted October 4, 2024

of a mucoperiosteal flap. A vertical, linear osteotomy line was marked, extending downward from the upper alveolar margin between the central incisors to the inferior border of the mandible. The osteotomy was performed using a surgical handpiece equipped with a #702 surgical cutting bur, which cut through the buccal cortex and into the cancellous bone while sparing the lingual cortex. The osteotomy was completed using an osteotome, and the two halves of the mandible were separated using a Smith spreader. At this point, a brisk bleed was observed in the sublingual region, resulting in rapid elevation of the floor of the mouth and the tongue (Fig. 1). The sublingual region area felt tense and firm. Steady downward intraoral compression of the hematoma, along with extraoral pressure directed superiorly at the chin region, was delivered using a bimanual maneuver for 5 minutes to prevent the hematoma from expanding and to facilitate drainage. The hematoma shrank con-

siderably within minutes. Concurrently, the osteotomized fragments were repositioned and secured with two 2.0-mm mini-plates: one at the superior border to avoid injury to the tooth roots and the other at the inferior border. This re-approximation of the hemimandibles prevented further bleeding and growth of the hematoma (Fig. 2). Dental occlusion was restored, and the surgical wound was closed in layers using 3-0 Vicryl suture material. The patient was then extubated without complications. In the immediate postoperative period, the patient's vitals remained stable. However, the tongue and floor of the mouth turned deep purple, and tongue movements appeared restricted (Fig. 3). The patient was monitored daily with careful attention to vitals and the extent of the swelling (Figs. 4, 5). By the seventh postoperative day, the clinical signs had completely resolved (Fig. 6).



Fig. 1. Intraoperative image demonstrating midline osteotomy and elevation of the floor of the mouth and tongue.



Fig. 3. Immediate postoperative image (taken 5 hours after surgery) displaying a hematoma on the floor of the mouth, with discoloration visible.

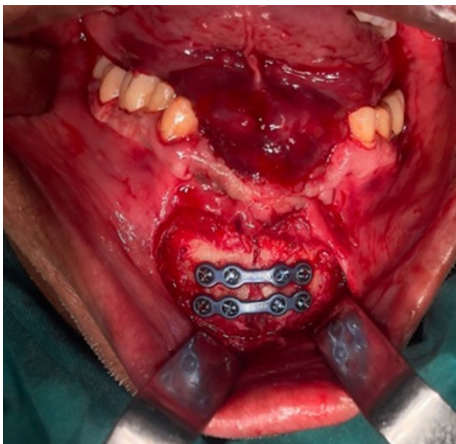


Fig. 2. Intraoperative image demonstrating reduction and fixation of the osteotomized segments.

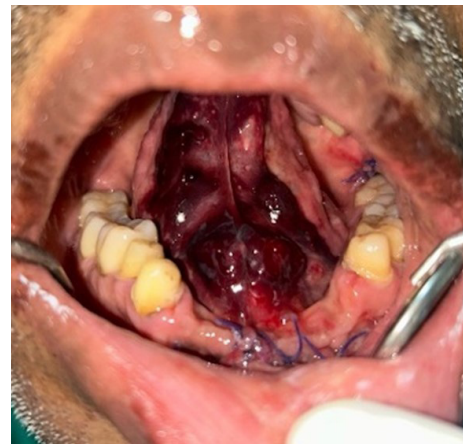


Fig. 4. Image from postoperative day 1 demonstrating meaningfully reduced swelling and a change in the color of the hematoma.



Fig. 5. Image from postoperative day 4 showing further reductions in swelling and discoloration from the hematoma.



Fig. 6. Image from postoperative day 7, demonstrating complete resolution of the hematoma.

LITERATURE REVIEW

In cases of sublingual hematoma, the hemorrhage is typically attributed to the arterial triad comprising the sublingual, submental, and incisive arteries [13]. Flanagan [14] reported that injury to the sublingual or submental artery can result in the loss of up to 450 mL of blood within 30 minutes. The flow rate, coupled with the anatomy of the oral cavity floor, facilitates the spread of the hematoma into the posterior and inferior sublingual spaces, potentially compromising the airway. Less severe complications associated with sublingual hematoma include drooling, voice changes, sore throat, dysphagia, trismus, neck ecchymosis, tachypnea, and stridor. However, the course of these blood vessels and the consequences of their injury are comparatively poorly understood.

Bleeding source and implications

The submental and sublingual vessels enter the mandible through the lingual foramina, which can be either median or lateral in location. The median lingual foramen is located precisely at the mandibular midline, near the genial tubercles. This anatomical feature is present in every individual, as demonstrated by Tepper et al. [15] and Tagaya et al. [16]. To minimize bleeding, many surgeons recommend avoiding perforation of the lingual cortex. Clinically, sublingual hematoma can be classified as either stable or expanding. Furthermore, based on the timing of occurrence, it is categorized as either intraoperative or postoperative [6].

Intervention is determined based on the clinical classification described above. A stable hematoma is considered self-limiting when it ceases to expand after a certain period, often due to the tamponading effect of the surrounding muscles and accumulated blood. In contrast, progressive or dissecting hematomas

continue to grow, potentially causing complications such as damage to vital structures or airway obstruction. These complications can lead to more severe outcomes, including anoxic brain injury and pulmonary edema.

Management modalities

While stable sublingual hematoma can be managed conservatively, progressive cases require surgical intervention. Management is relatively simple in cases of intraoperative hematoma with the patient intubated. In contrast, a hematoma that exhibits growth in the postoperative period, particularly in a non-intubated patient, presents a more critical situation. The most important step is to ensure a patent and secure airway. As the primary method of management, recommendations support establishing a definitive airway using fiber-optically guided nasotracheal intubation. However, in cases where nasotracheal intubation is not feasible, orotracheal intubation is contraindicated, and mask ventilation is not possible, alternatives such as cricothyrotomy or awake tracheotomy may be considered [17]. If the size of the hematoma interferes with the intubation technique, emergency tracheostomy may be necessary [18].

Digital compression and the application of pressure with a gauze pack constitute the primary measures to limit the progression of a hematoma. The bimanual compression maneuver, as described in the case report section of this article, is considered the most effective [13].

Sublingual hematoma may be managed medically with steroids to reduce edema [19], tranexamic acid to promote hemostasis, and antibiotics to prevent infection or septic shock. For cases in which anticoagulants have precipitated or exacerbated the situation, the administration of vitamin K, fresh frozen plasma, prothrombin complex concentrate, and blood transfusions are key components of medical management [20]. Addi-

tionally, local hemostatic agents like gel foam and fibrin glue, as well as the cauterization of bleeding vessels, can aid in achieving hemostasis [19].

Opinions regarding the aspiration or drainage of sublingual hematoma as a treatment modality remain polarized. While some surgeons advocate for the evacuation of the hematoma to quickly alleviate symptoms [19], others advise against it due to concerns about (1) exacerbating bleeding; (2) the potential risk of aspiration into the airway; and (3) the probable ineffectiveness of the procedure when the blood has accumulated in the intrinsic muscles of the tongue, which prevents adequate drainage [21]. As an alternative, releasing sutures can aid in the immediate evacuation of a hematoma. Furthermore, hirudotherapy [22], has been utilized under sedation, in the management of sublingual hematoma. Enzymes released by the leeches facilitate the gradual dispersal of blood (about 20 mL in 2 hours) collected in the sublingual space. When these methods fail, ligation and embolization procedures remain the last resort for management. Ligation of the lingual artery is the most effective approach to stop bleeding; furthermore, Bavitz et al. [23] recommend ligation of the submental artery or potentially the facial artery. Intraoral ligation of bleeding vessels is considered ideal. However, this may be challenging due to technical difficulties in identifying the vessels (due to local edema or hematoma altering the regional tissue anatomy), retraction of the severed vessel, and restricted access [11]. In such cases, extraoral ligation of the lingual, facial, or external carotid artery is performed. For severe cases, transcatheter arterial embolization is recommended following the identification of the severed vessel using advanced imaging modalities such as magnetic resonance angiography, contrast-enhanced computed tomography angiography, and digital subtraction angiography [7,24].

DISCUSSION

Many surgeons recommend avoiding perforation of the lingual cortex to prevent bleeding and related complications [6]. However, in midline osteotomy, breaching the lingual periosteum is typically inevitable, particularly for procedures requiring transverse expansion of the dental arches. Therefore, the risk of sublingual hematoma must be considered during the planning stage. Mandibular midline osteotomy is indicated for the correction of various clinical conditions, including transverse skeletal discrepancies of the face, long-standing temporomandibular joint dislocation, and malunited mandibular fracture. These procedures require an osteotomy cut that extends from the buccal cortex through to the lingual cortex, enabling the complete separation and mobilization of the hemimandibles to the

desired position.

Any violation of the periosteal boundary on the lingual aspect can lead to hemorrhage, with blood flowing unhindered into the sublingual plane due to the lack of a limiting barrier in the form of bone or muscular attachments. To prevent this complication, placing the osteotomy in a paramedian position, which avoids the lingual foramen, has been suggested [25]. In paramedian stepped mandibular osteotomy [26], the upper vertical section of the osteotomy is situated between the ipsilateral central incisor and the lateral incisor or between the lateral incisor and the canine. The middle horizontal section lies between the superior and inferior genial tubercles, where the genioglossus and geniohyoid muscles attach. Above and below these tubercles are the superior and inferior midline lingual foramina [27]. The lower vertical section is positioned between the posterior margin of the mandibular attachment of the digastric muscle anterior belly and the anterior margin of the mental foramen. The osteotomy line is meticulously planned to avoid both the tooth roots and the mental neurovascular bundle. By preserving the midline lingual foramina and their contents, this osteotomy technique helps prevent bleeding. In the present case report, hematoma progression was prevented by achieving hemostasis through the reduction and fixation of fracture fragments. This result stems from the occlusion of any bleeding vessels and the compressive forces exerted by the interfragmentary apposition of the osteotomized fragments. However, a similar approach to hematoma management following genioplasty provided only transient control; the hematoma reappeared in the immediate postoperative period, necessitating intubation [7].

In conclusion, sublingual hematoma requires prompt identification and immediate intervention to prevent complications. Adequate preoperative planning is essential and can be facilitated by the use of virtual surgical models, surgical guides, and computed tomography scans to determine the osteotomy lines before surgery and avoid damaging vital structures near the lingual foramina. Intraoperative navigation may also be employed to assist in guiding the osteotomy procedure. Controlled use of an oscillating saw and osteotome at the lingual cortex can help prevent sublingual hematoma [28]. Management of intraoperative bleeding should adhere to a standardized stepwise protocol, ranging from conservative measures such as the application of pressure to procedures like embolization [29].

NOTES

Conflict of interest

No potential conflict of interest relevant to this article was reported.

Funding

None.

Ethical approval

The study has been approved by Institutional Review Board of SRM Dental College (SRMU/M&HS/SRMDC/2024/PG/007).

Patient consent

The patient provided written informed consent for the publication and use of his images.

ORCID

Meghana Anishetty <https://orcid.org/0009-0008-9739-5606>

Radhika Menon <https://orcid.org/0009-0004-0312-3679>

Elavenil Panneerselvam <https://orcid.org/0000-0002-9930-3292>

Shri Krishna Prasanth B
<https://orcid.org/0000-0003-4584-6483>

Krishnakumar V. B. Raja
<https://orcid.org/0000-0002-1220-4081>

Author contributions

Conceptualization: Elavenil Panneerselvam. Data curation: Meghana Anishetty, Radhika Menon, Elavenil Panneerselvam. Methodology: Elavenil Panneerselvam. Writing - original draft: Meghana Anishetty, Radhika Menon, Elavenil Panneerselvam. Writing - review & editing: Meghana Anishetty, Radhika Menon, Elavenil Panneerselvam, Shri Krishna Prasanth B, Krishnakumar V. B. Raja. Supervision: Elavenil Panneerselvam.

REFERENCES

- Shetty P, Nandan A, Shetty M, Shetty S. Coleman sign: a hallmark for mandibular fracture? A rare case exception. *Eur J Mol Clin Med* 2020;7:1064-70.
- Mouzourakis MM, Seo SS, Oral R. Case of accidental trauma resulting in Condylar fractures in 3-month-old. *Case Rep Dent* 2023;2023:7611475.
- Kyriaki V, Fani S, Jiannis H. Iatrogenic massive hemorrhagic edema of the tongue: a case report. *Egypt J Otolaryngol* 2022; 38:128.
- Bhandary R, Poojitha S. Lingual hematoma: a rare presentation. *Austin J Otolaryngol* 2021;8:1121.
- Woo BM, Al-Bustani S, Ueek BA. Floor of mouth haemorrhage and life-threatening airway obstruction during immediate implant placement in the anterior mandible. *Int J Oral Maxillofac Surg* 2006;35:961-4.
- Barrientos-Lezcano FJ, Corchero-Martin G, Gonzalez-Nunez AB, Soler-Presas F. Life-threatening sublingual hematoma after mandibular implant placement: a case report. *Ann Maxillofac Surg* 2021;11:169-72.
- Vargas N, Donado D, Sifuentes-Cervantes JS, Castro-Nunez J, Guerrero LM, Ferrer-Nuin L. Life-threatening hemorrhage from the lingual artery after a genioplasty: case report and review of possible complications associated with orthognathic surgeries. *Oral* 2023;3:92-100.
- Satpathy S, Guha R, Satpathy A, Guha P. Spontaneous sublingual space hematoma secondary to hypertension: a case report and review of literature. *Natl J Maxillofac Surg* 2015;6:96-8.
- Adhikari A, Sharma S, Ghimire N, Mainali G, Yadav SK, Rajkarnikar R. Spontaneous sublingual hematoma secondary to warfarin therapy: a rare complication. *Clin Case Rep* 2022;10:e6739.
- Orhun N, Ekin U, Sanchez J, Azzam MH, Manickam R. Pseudo-Ludwig's angina: spontaneous sublingual haematoma elicited by warfarin. *Eur J Case Rep Intern Med* 2024;11:004299.
- Kalpidis CD, Konstantinidis AB. Critical hemorrhage in the floor of the mouth during implant placement in the first mandibular premolar position: a case report. *Implant Dent* 2005;14:117-24.
- Carreira-Nestares B, Urquiza-Fornovi I, Carreira-Delgado MC, Gutierrez-Diaz R, Sanchez-Aniceto G. Clinical case and literature review of a potentially life-threatening complication derived from mouth floor hematoma after implant surgery. *Eur Dent Res Biomater J* 2023;4:13-24.
- Limongelli L, Tempesta A, Crincoli V, Favia G. Massive lingual and sublingual haematoma following postextractive flapless implant placement in the anterior mandible. *Case Rep Dent* 2015;2015:839098.
- Flanagan D. Important arterial supply of the mandible, control of an arterial hemorrhage, and report of a hemorrhagic incident. *J Oral Implantol* 2003;29:165-73.
- Tepper G, Hofschneider UB, Gahleitner A, Ulm C. Computed tomographic diagnosis and localization of bone canals in the mandibular interforaminal region for prevention of bleeding complications during implant surgery. *Int J Oral Maxillofac Implants* 2001;16:68-72.
- Tagaya A, Matsuda Y, Nakajima K, Seki K, Okano T. Assessment of the blood supply to the lingual surface of the mandible for reduction of bleeding during implant surgery. *Clin Oral Implants Res* 2009;20:351-5.
- Huang D, Wanchu R, Walker J, Ganti L. Life-threatening sublingual hematoma in the setting of anticoagulation and neck trauma. *Cureus* 2022;14:e24974.
- Brotfain E, Koyfman L, Andrey S, Rusabrov E, Frenkel A, Zlotnik A, et al. Spontaneous sublingual hematoma: surgical or nonsurgical management? *Int J Case Rep Images* 2012;3:1-4.

19. Tsai CM, Susarla SM, Pittaway AJ, Egbert MA. Sublingual hematoma after autogenous block grafting to the anterior mandible in a patient with Noonan syndrome. *Oral Maxillofac Surg Cases* 2019;5:100106.
20. Karmacharya P, Pathak R, Ghimire S, Shrestha P, Ghimire S, Poudel DR, et al. Upper airway hematoma secondary to warfarin therapy: a systematic review of reported cases. *N Am J Med Sci* 2015;7:494-502.
21. Marin E, Fauconnier S, Bauters W, Huvenne W. Idiopathic spontaneous sublingual hematoma: a case report and literature review. *Oral Maxillofac Surg* 2020;24:117-20.
22. Jha K, Garg A, Narang R, Das S. Hirudotherapy in medicine and dentistry. *J Clin Diagn Res* 2015;9:ZE05-7.
23. Bavitz JB, Harn SD, Homze EJ. Arterial supply to the floor of the mouth and lingual gingiva. *Oral Surg Oral Med Oral Pathol* 1994;77:232-5.
24. Yu J, Choi JW, Whang K, Oh J. The role of angiographic interventions in life-threatening traumatic maxillofacial bleeding. *J Neurointensive Care* 2023;6:35-41.
25. Rattan V, Rai S, Sethi A. Midline mandibulotomy for reduction of long-standing temporomandibular joint dislocation. *Cranio-maxillofac Trauma Reconstr* 2013;6:127-32.
26. Zhao S, Han Z, Qu Y. Stair-stepped mandibulotomy: clinical indication, surgery program design, and operation points. *J Stomatol Oral Maxillofac Surg* 2024;125(3S):101866.
27. von Arx T, Lozanoff S. Lingual foramina and canals. In: von Arx T, Lozanoff S, editors. *Clinical oral anatomy*. Springer; 2017. p. 463-87.
28. Na HY, Choi EJ, Choi EC, Kim HJ, Cha IH, Nam W. Modified mandibulotomy technique to reduce postoperative complications: 5-year results. *Yonsei Med J* 2013;54:1248-52.
29. Sohn JB, Lee H, Han YS, Jung DU, Sim HY, Kim HS, et al. When do we need more than local compression to control intraoral haemorrhage? *J Korean Assoc Oral Maxillofac Surg* 2019;45:343-50.