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Lingual hematoma causing upper airway obstruction: an unusual manifestation of dengue fever

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ABSTRACT

The spectrum of dengue infection varies from dengue fever to more severe forms such as dengue hemorrhagic fever (DHF) and dengue shock syndrome (DSS). In DHF bleeding may be overt (external) or internal. At time bleeding can occur at unusual sites. We report one such case of DHF in which bleeding occurred within the tongue manifesting as an intra-lingual hematoma, causing significant upper airway obstruction.

1. Introduction

Dengue presents as an acute febrile illness with headache, skin rash, itchy skin, profuse sweating, myalgia, chills, retro orbital pain, diarrhea and hemorrhagic manifestations^[1–2]. Hemorrhagic manifestations, which are invariably present in DHF, are usually mild and most commonly found as scattered tiny petechiae in the skin and occasionally submucosa. A positive tourniquet test, which indicates increased capillary fragility, is the most common finding that appears early^[3]. Major hemorrhage is unusual except when in association with profound or prolonged shock^[4]. Gastrointestinal bleeding in the form of hematemesis and/or melena are the most common severe bleeding symptoms. In those who die after prolonged shock, bleeding has been observed in various organs, such as the gastrointestinal tract, heart, lungs, liver, and brain. We present a case of dengue with an unusual site of bleeding *i.e.* tongue, leading to upper airway obstruction.

2. Case report

A young man presented to emergency medicine with fever, headache and body ache of one week duration, one episode of bleeding from nose and vomiting for one day. There was no rash, joint pain, cough, breathlessness, or bleeding from any other site. Past, family and personal history were non-contributory. Sensorium was normal and other systemic examination was unremarkable.

Investigations revealed hemoglobin of 12.4 g/dL, total leucocyte count of 5 600 cells/dL, with 54% neutrophils and 42% lymphocytes. However, platelet count was only 20 000 cells/dL. Peripheral smear was negative for malarial parasite. Urine routine, kidney and liver function tests and coagulation profile were normal. Dengue serology (IgM) was positive. Chest X-ray and ultrasonography of abdomen showed no abnormality.

Four hours after admission he complained of breathlessness and difficulty in speaking. However, he was conscious, oriented and had no any neurological deficit. He could not protrude his tongue and was unable to speak, however he could express himself by writing without any problem. At this time his chest was clear and oxygen saturation was 98%. A possibility of metoclopramide induced dystonia was considered and treated. But by next day the tongue was found to be enlarged and swollen (Figure 1), it was touching the hard palate and obstructing the entire oral cavity resulting in respiratory distress. Chest examination

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was normal, but oxygen saturation by pulse oximetry was 90%. Arterial blood gas analysis confirmed presence of hypoxemia; that was corrected by administering oxygen by nasal prongs at 4 L/min. An urgent tracheostomy was requested but not considered due to thrombocytopenia. A diagnosis of dengue hemorrhagic fever with intra-lingual hematoma was made and patient treated conservatively. Platelets were transfused; adequate oxygenation and fluid balance were maintained. Subsequently with improving platelet counts, hematoma of the tongue started to reduce (Figure 2). At no point of time, there was overt bleeding from the tongue. By the 7th day of hospitalization, the patient recovered with sufficiently clear spoken speech and was discharged.



Figure 1. Lingual hematoma.



Figure 2. Resolving lingual hematoma.

3. Discussion

Trauma and anticoagulation/thrombolysis with heparin, warfarin, streptokinase, and tissue plasminogen activator are recognized as the most common causes of lingual hematoma^[5–13]. In the present case the cause was thrombocytopenia due to dengue infection. However, the decrease in saturation was not completely explained by the hematoma. Micro-hemorrhages within the lung are a distinct probability and were possibly responsible for the hypoxemia.

Other causes of inability to protrude tongue are tongue-tie, carcinoma of tongue, oral submucous fibrosis (OSMF), herpes zoster, partial seizure, hypoglossal nerve palsy (intracranial

lesions, neuroma or schwannoma of the hypoglossal nerve, vascular anomaly of the vertebral or basilar artery, infected impacted tooth or head and neck injury) etc.

Patients with lingual hematoma can present with greatly varying degrees of airway compromise, accounting for the wide variation in airway management approaches. Indeed, the potential for lingual hematoma to completely obstruct the upper airway must not be overlooked. In acute upper airway obstruction, nasotracheal intubation, tracheostomy or cricothyrotomy may be required. But in the present case, no such intervention was required because dengue being self-limiting, platelet counts started to rise soon. Thus successful management without surgery indicates that observation, close monitoring and reversal of thrombocytopenia by transfusing platelets is a reasonable management option in such cases.

Conflict of interest statement

We declare that we have no conflict of interest.

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