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## ■ Case Report

# Traumatic Macroglossia Complicating Eclampsia in a Low Resource-setting

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## ABSTRACT

Orofacial injuries are well documented complications of eclampsia and other convulsive episodes. Tongue laceration and its sequel tongue oedema and airway obstruction in eclampsia patients is a serious but highly preventable complication. It increases the morbidity and mortality from eclampsia; and some of the effects such as slurred speech, speech defects persist even after complete resolution of eclampsia and other associated complications of severe pre-eclampsia. We describe three cases of women with tongue laceration complicated by oedema following eclamptic fits. The women had varying outcomes and associated morbidities which persisted after resolution of the precursor eclampsia. Tongue laceration and oedema must be prevented. Eclamptic patients must be assessed for tongue laceration at presentation at the healthcare facility for the prevention, early identification, prompt management; and prevention of oedema and attending complications.

**Keywords:** Traumatic macroglossia, Tongue laceration eclampsia, tongue oedema eclampsia, orofacial injury eclampsia, Eclampsia

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## Introduction

Eclampsia is a complication of severe pre-eclampsia which is a hypertensive disorder of pregnancy characterized by elevated blood pressure, proteinuria and progressive multi-organ systems impairment/disease. Eclampsia manifests as tonic-clonic seizures. Seizure-induced complications include orofacial injuries, head trauma, broken bones, aspiration pneumonia and loss of consciousness.<sup>1,2</sup>

Orofacial injuries are known to complicate convulsive episodes including eclamptic episodes.<sup>3,5</sup> These injuries may include tongue or lip abrasions and lacerations, tooth avulsion, tooth dislocation, temporomandibular joint (TMJ) dislocation and facial bone fractures.<sup>4</sup>

Tongue laceration in eclampsia may be complicated by tongue oedema and airway obstruction which occurs as a life-threatening but preventable complication. Tongue laceration

results from trauma to the tongue from rigorous jaw movement during eclamptic fits and not a direct consequence of the multi-system disease manifestation of pre-eclampsia and eclampsia;<sup>4,6</sup> thus it is less emphasized in the literature as one of the complications of eclampsia. It increases the morbidity and mortality associated with the condition; and the need for advanced care. It requires prevention, early identification and prompt management.

We describe clinical presentation, management and outcome of cases of tongue laceration complicated by oedema at a tertiary health institution.

### Case Report

- **Case 1:** A 27-year-old P 5<sup>+0</sup> (4 alive) unbooked woman presented to the Obstetrics and Gynaecology emergency unit on the first day postpartum with a history of bleeding from the mouth of 4 hours duration following generalized tonic-clonic seizure and unassisted spontaneous vaginal delivery at home. She was delivered of a live baby. Each episode of convulsion lasted about 2 minutes, self-aborted and associated with transient loss of consciousness. There was no history of antenatal care.

At presentation, she was conscious but confused, the Glasgow Coma Score (GCS) was 9/15, restless and in respiratory distress. Patient was afebrile (temperature 36.60C), severely pale, not cyanosed and had bilateral pitting pedal oedema. The pulse rate was 104 beats/minute, blood pressure of 144/95 mmHg, respiratory rate was 40 cycles/minute and oxygen saturation was 96%. The oral examination revealed a massive tongue swelling (oedematous), the tongue was protruding out of the mouth with multiple active bleeding sites. (Figure 1) There were lacerations on the posterior 3rd of the ventral and dorsal surface of the tongue measuring about 4cm and 3.6cm respectively. The Packed Cell Volume (PCV) was 18%, Urinalysis showed 3+ protein, serum electrolytes and urea, creatinine and uric acid showed elevated urea (56mg/dl) and uric acid (9.7mg/dl). A diagnosis of post-partum eclampsia with extensive tongue laceration and severe anaemia were made. She was managed in the intensive care unit by a multi-disciplinary team. She was transfused with 4 units of blood. Tongue laceration was managed with oral cleansing and daily dressing. Patient recovered and was discharged against medical advice on the 4th day on admission.



Figure 1A Before Dressing



Figure 1B After dressing

**Figure 1: Case 1 -A 27-year-old P5<sup>+0</sup> (4 alive) unbooked woman with postpartum eclampsia, tongue laceration and oedema.**

- **Case 2:** A 38-year old P5<sup>+0</sup> (5 alive) unbooked woman presented with a history of seizures and labour pains of 5-hours duration at a gestational age of 26 weeks 6 days. The seizure was initially mainly facial; lasted for about 20 minutes and self-aborted. Seizures progressed to generalized seizures lasting an average of 20-30 seconds at presentation; the patient had about 3 episodes over a period of 1 hour and each aborted with IV midazolam. She had a spontaneous expulsion of a macerated fetus per vaginum; with a residual fetal weight of 1.3kg. The estimated blood loss was about 120mls. A maintenance dose midazolam was administered as a continuous intravenous infusion at 1.5g per hour.

On examination at presentation; she had altered consciousness; and a GCS of 13/15 which deteriorated to 3/15 with recurrent episodes of seizures. She was not pale, anicteric, acyanosed afebrile (temperature 37.20C) but had bilateral pitting pedal oedema. The pulse rate was 92 beats/minute, blood pressure was 141/88mmHg, respiratory rate was 24 cycles/minutes and oxygen saturation 88-91 %. The oral examination revealed bleeding from oral cavity, multiple lacerations

about 4.0cm long and 4.5cm wide collectively. A diagnosis of eclampsia with tongue laceration was made. She was immediately intubated. A size 6.5mm cuffed endotracheal tube aided by fiberoptic intubation was inserted nasally and mechanically ventilated via intermittent positive pressure ventilation (IPPV mode). Haemostasis was secured; the tongue laceration and oropharynx were packed with wet gauze. She was managed using a multi-disciplinary team approach. The patient had debridement of the tongue laceration and necrotic areas were excised in addition to oral cleansing and daily dressing of the lacerated tissues; the tongue swelling progressively subsided. Urinalysis showed proteinuria; PCV was 31%. The tongue lacerations had healed and swelling subsided by the 5th day on admission in the ICU. As a result of initial repeated seizures she suffered hypoxic ischaemic brain injury leading to persistent vegetative state. She had tracheostomy tube inserted by otorhinolaryngology team and was discharged after 26 days with the GCS of 10/15. She spent another 28 days on the ward before finally discharged for home care.



**Figure 2: Case 2 - A 38-year old P5<sup>+0</sup> (5 alive) unbooked woman with a history of seizures and labour pains and tongue swelling.**

- **Case 3:** A 35-year-old G4 P3<sup>+</sup> (3 Alive) unbooked woman, presented to the obstetric and gynaecologic emergency unit with a history of 4 episodes of generalized tonic-clonic convulsions of 4 hours duration; tongue swelling and bleeding of 2 hours duration at a gestational age of 35 weeks and 6 days. She had bitten her tongue during the recurrent episodes of seizures and sustained laceration of the tongue. She had spontaneous vaginal delivery of a fresh stillbirth fetus 3 hours after the first convulsion.

On examination, the patient was unconscious, GCS was 8/15 at presentation but heavily sedated (via intravenous diazepam administered before referral), not in respiratory distress, not pale,

anicteric, afebrile (Temperature 36.9°C) and bilateral pitting oedema. The pulse rate was 98 beats/minute, blood pressure was 198/108 mmHg, and SpO<sub>2</sub> was 98%. Oral examination revealed a swollen tongue with several bleeding points (punctate); blood clots on the ventral surface of the tongue and the haematocrit was 40%. Urinalysis showed proteinuria. A diagnosis of eclampsia with tongue oedema following laceration was made. She was managed in the Intensive Care Unit (ICU). The GCS improved to 15/15. Tongue laceration was managed with oral cleansing and oedema resolved. The patient recovered and was discharged from the ICU to the postnatal ward.



**Figure 3: Case 3 - A 35-year-old G4 P3<sup>+</sup> (3 alive) eclampsia complicated by tongue laceration and oedema at a gestational age of 35 weeks and 6 days.**

All the patients were co-managed with the intensivist (anaesthetist), oral and maxillofacial surgery and otolaryngology surgery teams. The patients had Intravenous Magnesium sulphate 4g loading dose was injected slowly over 15 minutes and maintenance dose of 5g 5 hourly for 24 hours to control the eclamptic fits. The blood pressure

was controlled with anti-hypertensive drugs- intravenous and oral Labetalol and oral Nifedipine in the three patients. The patients had IV dexamethasone 8mg, 8 hourly for 3 days and prophylactic antibiotic IV Amoxicillin-clavulanic acid 600mg 8 hourly for 5 days and IV Metronidazole 500mg 8 hourly for 5 days.

## Discussion

This study highlights three cases of eclampsia with laceration of the tongue complicated by oedema and with different outcomes of management.

Severe pre-eclampsia and eclampsia are complications of hypertensive disease of pregnancy and constitute obstetric emergencies which require prompt attention in order to prevent maternal and perinatal morbidity and mortality.<sup>2</sup>

Orofacial injuries are known complications of convulsive episodes including eclampsia and sometimes overlooked by the obstetrician, midwives and patients' relatives in the face obstetric emergency with ongoing seizure episode or loss of consciousness. Tongue laceration and other orofacial injuries have also been observed in children with febrile convulsions, individuals with epilepsy and during electroconvulsive therapy.<sup>7,8</sup>

The spectrum of injuries includes soft tissue and hard tissue injuries. Injuries include abrasion or laceration of the tongue and lips, cheek injury, injury to the gingiva, tooth crack, tooth fracture, tooth avulsion or dislocation, bone fracture, joint dislocation.<sup>4,6,9</sup> The mechanism of the injury involves biting of the soft tissues of the oral cavity, soft tissue trauma due to forceful insertion of hard objects or metals into the mouth by relatives during the seizure episodes.<sup>4,6</sup> Many times, these patients have had several episodes of eclamptic fits prior to presentation in the hospital; and the pre-hospital care of the eclamptic patient is a predisposing factor. The details of the predisposing (or antecedent) events or pre-hospital care during the seizure episodes or mechanism of injury is usually not emphasized.

In this case series, the tongue lacerations occurred due to a combination of tongue biting and forceful insertion of objects into the mouth during the recurrent convulsive episodes. The incidence of orofacial injuries among eclampsia patients was reported as 12.1% by Ndukwe et al;<sup>5</sup> while Adeyemo et al reported 42%.<sup>4</sup> In another survey, the prevalence of orodental trauma during seizures was as high as 52.4%.<sup>9</sup>

Biting and forceful insertion of objects into the mouth accounted for 35% of the injuries each and

91% were soft tissue(4); this is consistent with the features seen in this case series. Tongue lacerations occur commonly on the anterior dorsum of the tongue; followed by mid-dorsum and anterior ventral surfaces to a lesser extent.<sup>10</sup>

In this study, the tongue laceration was complicated by oedema and airway obstruction in the unconscious patients. This phenomenon is known as traumatic macroglossia and potentially life-threatening. The mechanism of traumatic macroglossia involves haemorrhage into the tongue, obstruction of lymphatic vessels and inflammation.<sup>11</sup>

Laceration of the tongue in eclampsia and its complications presents an additional burden to the obstetrician, anaesthetist/intensivist, maxillofacial and otolaryngology teams. It constitutes a challenge in the management of the patient and rate of recovery of the patients. A delay in identification and management of tongue laceration increases the morbidity and associated complications such as respiratory problems and thus; a medical emergency. Tracheal intubation in these cases is a challenge.<sup>(12)</sup> In case 2, patient was intubated trans-nasally via fibre-optic intubation with a size 6.5mm endotracheal tube due to a reduction in the oral cavity and obstruction caused by the tongue enlargement. Other complications of tongue laceration include bleeding/anaemia, oedema, airway obstruction, aspiration pneumonia and impaired speech.<sup>10</sup> This case series reveals that tongue laceration and associated traumatic macroglossia increases the need for advanced care as demonstrated by the need for intensive care admission, trans-nasal fibre-optic endotracheal intubation, mechanical ventilation, oral wound debridement, multi-disciplinary team management among others. These are usually not available in the primary and secondary care facilities in low resource settings; and when available the facility is sparsely distributed. Consequently, it is better to prevent and promptly treat tongue laceration in its early phase before oedema sets in.

The principle of management of eclampsia includes management of the unconscious ensuring

a patent airway, breathing, circulation, control convulsions and prevent further convulsions; control the blood pressure; and to evaluate for associated complications.<sup>2</sup>The treatment of tongue laceration with oedema, includes muscle relaxant and bite riser or bite guard together with earlier manual replacement into the oral cavity are advised in order to prevent additional trauma and arrest the cycle of obstruction and congestion that leads to further edema and swelling.<sup>11,13,14</sup> Oropharyngeal airway device is a simple device that will prevent injury to the orodental tissues during seizures; and must be available in the emergency trays in all hospitals. Furthermore, orodental trauma during seizures can be averted by the use of custom-made mouth guards.<sup>9</sup>

The goal of repair of tongue laceration includes wound closure or apposition, haemostasis, preserve tissue integrity, mobility and function in articulation and deglutition.<sup>15</sup> The decision to suture a laceration depends on the size, extent of laceration, gaping of the wound and delay in presentation<sup>10</sup> and expertise. Healing by secondary intention is the alternative when the wound is potentially contaminated or devitalized; while ensuring oral cleansing and dressings.

The number of eclamptic fits was also associated with the outcome of eclampsia in this case series. Case 2 had 4 episodes of eclamptic fits and sustained more extensive tongue laceration, necrosis, tissue loss and hypoxic brain injury. Adeyemo et al documented a correlation between recurrent eclamptic fits and orofacial injuries.<sup>4</sup>

All patients in this case series were given antibiotics and steroid. The criteria for the use of prophylactic antibiotics include heavily contaminated wounds, immuno-compromised status of the patient and delayed wound closure.<sup>10</sup> The choice of antibiotic should give consideration to coverage of both gram-positive and anaerobic organisms.<sup>16</sup> Moreover, these patients were postpartum women with the risk of puerperal infection and the premorbid oral hygiene status may predispose them to wound infection. The use of prophylactic antibiotics for tongue injury is of benefit and controversial.<sup>16,18</sup>

The obstetricians and midwives should be aware and alert to the possibility of a tongue laceration and other orofacial injuries in eclampsia; and must be prompt in evaluating the patient for orofacial injuries at presentation during the primary survey of the eclamptic patient whether she is conscious or unconscious. The obstetricians and midwives are central and key players in the management of the eclampsia with tongue.

There is a need to educate the pregnant women, relatives and the communities on the hazards of using unpadded objects and forceful insertion of these into the oral cavity during eclamptic fits. The individuals and communities must be well-equipped with this information, alternative positive behaviours and interventions that will be beneficial to the patients during seizure episodes.

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