CASE OF THE MONTH

Lingual Thyroid Presenting as Life Threatening Haematemesis in Pregnancy

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Introduction

H ematemesis in pregnancy endangers two lives; that of the mother and the fetus. Apart from common causes like ulcers, erosions, and varices, pregnancy related conditions like hyperemesis, acute fatty liver (AFLP) and hemolysis, elevated liver enzymes and low platelet (HELLP) syndrome may be the etiologies.

Lingual thyroid is a rare embryological anomaly existing with a prevalence of 1: 100000 to 1:300000, occurring with female to male ratio of 7:1 to 4:1.1-3 The manifestations range from asymptomatic to voice change, dysphagia, bleeding and difficulty in intubation.1,3,4 Massive bleeding complicating a lingual thyroid in pregnancy is very rare.5 We report a case of massive hematemesis in a 36 week pregnant lady leading to hypovolemia, intrauterine fetal death, acute kidney injury and needing intensive care, the etiology of which was a previously asymptomatic undetected lingual thyroid.

Case Report and Discussion

A 28 years primigravida with 36 weeks of amenorrhea, presented to a private hospital with a one day history of 5-6 episodes of vomiting fresh blood and clots, each episode around 70-100 ml. There was no history of epigastric pain, gastroesophageal reflux, excessive retching, melaena, or similar episode in the past. There was no history of jaundice, epistaxis or pregnancy induced hypertension (PIH). On examination she was conscious, oriented, with a pulse rate of 102 beats per minute, blood pressure (BP) 90/64 mm Hg. She had pallor and abdominal examination revealed fundal height corresponding to the gestational age with a fetal heart rate of 146/min. Patient was referred to our tertiary care centre for further management. On arrival in casualty, she was pale, conscious, oriented, pulse rate 106 beats per minute, low volume, BP 90/60 mm Hg. There was no icterus, petechial rash or asterexis. Fetal heart sounds were absent; immediate ultrasonography confirmed foetal demise. There was nothing contributory in past and personal history. She had a low hemoglobin of 6.7 gram per deciliter with normal platelet counts and coagulogram. Table 1 outlines her investigations on admission.

Table 1: Investigations on admission

<table>
<thead>
<tr>
<th>Test</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hemoglobin</td>
<td>6.7 g/dl</td>
</tr>
<tr>
<td>WBC count</td>
<td>11300 cells/cc</td>
</tr>
<tr>
<td>DC</td>
<td>N65/L30/M3/E2</td>
</tr>
<tr>
<td>Platelet count</td>
<td>3.2 lak/ccc</td>
</tr>
<tr>
<td>S. urea</td>
<td>11 mg/dl</td>
</tr>
<tr>
<td>S. creatinine</td>
<td>1.1 mg/dl</td>
</tr>
<tr>
<td>Tot bili/ Direct bili</td>
<td>1.0/0.2 mg/dl</td>
</tr>
<tr>
<td>SGOT/SGPT</td>
<td>34/24 U/l</td>
</tr>
<tr>
<td>T. Prot</td>
<td>5.6 g/dl</td>
</tr>
<tr>
<td>Albumin</td>
<td>3.9 mg/dl</td>
</tr>
<tr>
<td>PT</td>
<td>12 sec (control 11 sec)</td>
</tr>
<tr>
<td>INR</td>
<td>1.09</td>
</tr>
<tr>
<td>APTT</td>
<td>30 sec (control 29.7 sec)</td>
</tr>
<tr>
<td>T. Prot</td>
<td>5.6 g/dl</td>
</tr>
<tr>
<td>USG Ab</td>
<td>intrauterine fetal death. No organomegaly. No ascites</td>
</tr>
</tbody>
</table>

She was stabilised with intravenous fluid resuscitation and blood transfusions in emergency ward. Upper GI scopy performed on day 2 of admission, revealed a visible vessel with small ulceration on lesser curvature of stomach, haemoclip was applied.

Hematemesis in pregnancy could be due to common causes like gastroduodenal ulcers, erosive esophagitis and esophageogastric varices. The increased intra abdominal pressure and relaxant effect of progesterone on smooth muscles increases the incidence of hiatus hernia and consequent hematemesis in pregnancy. The increased cardiac output at the end of the second trimester along with complex physiological vasodilatory responses may lead to dilatation and rupture of thin-walled angiodysplastic lesions in the GIT leading to hematemesis like in Osler Weber Rendu syndrome. The physiological changes in pregnancy may worsen portal
Hematemesis in Mallory Weiss tear and subsequent gravidarum predisposes to hematemesis. Hyperemesis in eclampsia and present complications with obstructive symptoms or hemorrhage. Hormonal changes in pregnancy can increase the size of hemangiomas predisposing to bleeding either spontaneously or following minor trauma.

A biopsy of lingual mass was done. A clinical differential of hemangioma versus lingual thyroid was considered. Dead fetus was delivered per vaginally, after induction of labour with oxytocin drip. Patient was shifted to MICU in an intubated state with persistent ooze from the back of the tongue. Lingual thyroid is most common ectopic presentation approaching 90 percent cases of ectopic thyroid. In 70% of the cases ectopic thyroid tissue is the only functioning thyroid in absence of eutopic thyroid. Hemangioma of the tongue is common but their occurrence at the back of the tongue is rare. Hemangiomas may be asymptomatic or present with obstructive symptoms or hemorrhage. Multifactorial; possible etiologies important in pregnancy. Our patient did not have any visible lesion in the oral or nasal cavity. The homogenously enhancing lesion at the base of the tongue could be a hemangioma or a very rare possibility of a lingual thyroid. The first upper GI scopy while revealing a visible vessel with small ulceration in the stomach, had not revealed any ooze from the back of the tongue. Vasospasm at the time of CT angiography may preclude the culprit lesion to be obvious. To resolve the diagnostic dilemma regarding the culprit lesion, on day 4, a repeat upper GI scopy, nasal endoscopy and direct laryngoscopy was planned under general anesthesia.

The patient was electively intubated in the operation theatre; subsequent upper GI scopy and nasal endoscopy were normal. Direct laryngoscopy revealed a lobular mass of around 2-3 centimetre at the base of the tongue with active ooze. Oropharyngeal packing was done. A clinical differential of hemangioma versus lingual thyroid was considered. Dead fetus was delivered per vaginally, after induction of labour with oxytocin drip. Patient was shifted to MICU in an intubated state with persistent ooze from the back of the tongue.
detected during periods of increased demand as in pregnancy, which stimulates the thyroid gland to increase production of thyroid hormones by as high as 40 to 100 percent to meet maternal and fetal needs.3,14 This increased demand culminates into thyroid gland enlargement by causing glandular hyperplasia and increased vascularity, human chorionic gonadotrophin (hCG) hormone being a contributory factor.14 This explains the presentation of lingual thyroid with bleeding during pregnancy; as evident in our case.

Our patient presented with foetal loss at full term due to life threatening haematemesis from an unusual source. While evaluating this diagnostic dilemma, we progressed to identify the source as per standard protocol, where we first ruled out common causes of haematemesis viz. nasal, oesophageal, gastric and duodenal. CT angiography gave a clue by demonstrating vascular mass at base of tongue which was confirmed on thyroid scintigraphy to be a lingual thyroid with absence of thyroid in neck. Such case reports are very rare; literature search revealed a case of lingual thyroid presenting as bleeding in a pregnant lady causing shock which was managed with embolization.5 Empirical embolization of a bleeding vascular lesion at the base of the tongue may prove hazardous if it turns out to be the only functional thyroid tissue in the form of lingual thyroid.

The development of thyroid gland starts around third week of intrauterine life.1 It arises from the pharyngeal endoderm, descends into the neck and reaches its eutopic position in the neck by 7th week of gestation.1,13 Any aberration during embryogenesis and/or migration process can lead to ectopic thyroid tissue, lingual position being commonest.13 The most common presentation of ectopic thyroid is clinical or subclinical hypothyroidism as was seen in our patient.2,13

Ectopic thyroid is usually scintigraphy with technetium 99m. This revealed focal uptake of tracer at base of tongue and absent uptake of tracer in the normal location of thyroid gland thus confirming lingual thyroid (Figure 3).

Patient gradually recovered and resumed oral feeds in the form of liquids. She was safely decannulated of tracheostomy on day 45 and discharged on day 53 with serum creatinine of 1.1 mg/dL. Her subsequent followup at 3 and 6 months revealed hemoglobin in the range of 9 to 10 g/dl with normal thyroid function test. She was advised to continue 50 μgm thyroxine daily and counseled regarding risk of recurrence of bleeding during subsequent pregnancies and asked to keep a regular followup.

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Ectopic thyroid is usually

**Fig. 3:** Thyroid scintigraphy with technetium 99m confirming lingual location of thyroid and absence of uptake at normal location of thyroid

**Conclusion**

The case has been presented to increase awareness regarding lingual thyroid, a rare anomaly, which can present as an atypical cause of haematemesis; especially in a pregnant patient and needs multispecialty co-ordination.

**References**

5. Chiu TT, Su CY, Hwang CF, Chien CY, Eng HL. Massive bleeding from an ectopic lingual thyroid follicular adenoma during...


