



Revista Colombiana de Anestesiología

Colombian Journal of Anesthesiology

www.revcolanest.com.co



Case report

Anesthesia for emergency tracheostomy due to bleeding hemangioma of the tongue in a pregnant patient[☆]

Marcelino Murillo^{a,*}, Merlys Zarza^b, Silvia Gómez^b, Juan Chica^c, Rubén Carrasquilla^d, Jairo de la Vega^c, Ramiro Alcalá^d, Diego Suescun^e

^a Anesthesiology Resident, Universidad de Cartagena, Colombia

^b Student, Universidad de Cartagena, Colombia

^c Anesthesiologist, Docente posgrado Anestesiología, Universidad de Cartagena, Colombia

^d Anesthesiologist, Clínica MEDIHELTH, Cartagena, Colombia

^e Otolaryngology Resident, Universidad de Cartagena, Colombia

ARTICLE INFO

Article history:

Received 3 February 2012

Accepted 19 May 2012

Available online 9 August 2012

Keywords:

Hemangioma

Pregnancy

Intubation

Vascular anomalies

ABSTRACT

We present the case of a 20-year-old pregnant patient diagnosed with cavernous hemangioma in her tongue, lips and neck with active bleeding. An emergency tracheostomy under general anesthesia was scheduled. Anesthesia was planned for conscious intubation with a fibrobronchoscope suited for the difficult airway management ASA algorithms. The airway was managed with a number 5.5 Fr. endotracheal tube under 0.02–0.05 $\mu\text{g/kg/min}$ remifentanyl infusion. The patient was sedated and given local anesthesia in the upper airway to reduce pain and gag reflex during spontaneous breathing. Intubation was successful. Having a total blood loss of 200 cc, the patient was taken to the postoperative care room hour after surgery. The airway was managed with a tracheostomy procedure using a number 7.5 Fr tube and without damaging the hemangioma. She was then scheduled for a vessel embolectomy and surgical removal. In this case, conscious intubation with remifentanyl was safe and adequate for the approach of the patient with bleeding airway hemangioma.

© 2012 Published by Elsevier España, S.L. on behalf of Sociedad Colombiana de Anestesiología y Reanimación.

Anestesia en embarazada para traqueostomía urgente por hemangioma sangrante en lengua

RESUMEN

Paciente embarazada de 20 años de edad con diagnóstico de hemangioma cavernoso en lengua, labios y cuello, con sangrado activo. Fue programada para traqueostomía de emergencia bajo anestesia general. La inducción anestésica fue planeada con intubación despierta con fibrobroncoscopio acorde a los algoritmos de vía aérea difícil de ASA, siendo la vía aérea manejada con tubo endotraqueal número 5.5 Fr. bajo infusión de remifentanil a 0.02–0.05 $\mu\text{g/kg/min}$ para sedación y se aplicó anestesia local en la vía aérea superior para

Palabras clave:

Hemangioma

Embarazo

intubación

Malformaciones Vasculares

[☆] Please cite this article as: Murillo M, et al. Anestesia en embarazada para traqueostomía urgente por hemangioma sangrante en lengua. Rev Colomb Anestesiología. 2012;40:313–7.

* Corresponding author at: Manzana K, lote 22, barrio los Corales, Cartagena, Colombia.

E-mail address: marcelmoro04@gmail.com (M. Murillo).

disminuir el dolor y el reflejo nauseoso, mientras respiraba espontáneamente. La intubación traqueal fue realizada exitosamente. Posterior a una hora de cirugía con 200 cc de pérdidas sanguíneas, la paciente fue trasladada a la sala de cuidados posoperatorios. Su vía aérea fue manejada con traqueostomía, tubo número 7.5 Fr, se realizó traqueostomía sin lesionar el hemangioma en cuello. Posteriormente fue programada para embolectomía de los vasos del hemangioma y luego será programará para escisión quirúrgica. En este caso la intubación en paciente despierto con fibrobroncoscopia bajo infusión con remifentanil fue segura y útil para el abordaje del paciente con hemangioma sangrante en vía aérea.

© 2012 Publicado por Elsevier España, S.L. en nombre de Sociedad Colombiana de Anestesiología y Reanimación.

Introduction

Hemangiomas are benign vascular pathological processes that result from endothelium cell proliferation¹; and for the most part are congenital. Such hemangiomas in the oral cavity are more frequent in the lips, with a low incidence in the tongue and more commonly found in females – 65% – more often than in males.² They may remain without growing and most show lesion involution, predominantly in teenage patients.¹ Histologically speaking, vascular proliferation in a connective tissue matrix covered by squamous epithelium is observed. Occasionally, it may show capillary endothelium with normal vessels. It differs from the cavernous hemangioma in its greater, dilated, tortuous and irregular vascular structures. Capillary vessels are predominant in the mucosa; and must be differentiated from other lesions such as Kaposi sarcoma, lymphangioma, mucocoele, among others.¹ Inside the oral cavity, they are more common in the dorsal side of the tongue, the lower lip and palate. These lesions are tuberous, flattened or cavernous. Their progress is slow, insidious and without pain. Hemangiomas in the tongue cause several problems, such as deformities, functional speech, deglutition and chewing impairment³ and can become large enough to compromise the patient's airway, which is why it must be secured.

Regarding hemangiomas, a close relation between bleeding frequency and pregnancy has been established. It is believed that hormonal, vascular and hemodynamic changes that occur in pregnancy increase the chance of bleeding by raising the circulating effective volumen from 30 to 50%, as well as increased venous distensibility caused by progesterone and endothelial growth by oestrogens. All of these result in growth of a present hemangioma.^{4,5} Fibrobronchoscopy on a conscious patient is an alternative to secure the airway before the possibility or certainty of a difficult airway (VAD).⁶ The most important indication for conscious fibrobronchoscopy is a patient with VAD and risk of bronchoaspiration. In our case, the risk was blood aspiration.⁷⁻⁹

A proper conscious fibrobronchoscopy requires the patient's cooperation, which is why an exhaustive explanation is very important. The procedure can be done using a nasal or oral pathway. The nasal pathway provides a more direct approach to the glottis, and it requires preparation with vasoactive drugs and local anesthesia to prevent pain and bleeding. Light sedation is recommended to avoid respiratory depression and the view can be improved by reducing secretions with antisialogogues, thus enhancing the anesthetic effect by allowing direct contact with the mucosa. If

the patient is at risk for bronchoaspiration, prophylaxis is carried out with H2 blockers, proton pump inhibitors and dopaminergic drugs to reduce gastric acid secretion and inhibit upper gastroesophageal tract motility, thus increasing lower esophageal sphincter tone.⁹ The patient must be monitored during the entire procedure. Providing oxygen through a cannula or the FBO is recommended. This method not only provides oxygen for the patient but also removes secretions and prevents fogging of the lens. No more than 5 l must be passed, cases of gastric distension and rupture have been reported.⁹

Case report

Patient, age 20, Gestations: 1 Deliveries: 0, 12 weeks at the time of admission, Date of last period: 02/25/2011; with a history of congenital hemangioma in tongue, lower lip and neck (Fig. 1). The patient had an episode of bleeding in July 2009, initially mild and occasional. As of April 2011 it has increased its frequency dramatically and has peaked in the last 10 days, with greater blood loss but has not required blood transfusions yet. She was referred to the Hospital Universitario de Caribe for integral management through the Otolaryngology service.

On physical examination (at sea level) findings were: female young adult, conscious, alert, calmed, pale, obedient, cooperative, communicated by means of writing. Vital signs: Heart rate: 108 per minute, BMI: 25 kg/m², Height: 167 cm, Weight: 70 kg. Symmetrical, mobile neck with no hyperextension movement limitation (Fig. 2), 2 cm × 2 cm scar in



Fig. 1 – Congenital hemangioma of the tongue.



Fig. 2 – Scar by involuted hemangioma – distance tiromentoniana (11 cm).

neck zone II 2 cm right of the midline due to mildly involved hemangioma. A fleshy purple mass with diffuse ulcerated areas is found in the tongue and extended all along the visible surface of the tongue, predominantly on the left side (Figs. 3 and 4) and protruding, making modified Mallampati scoring difficult, DTM 11 cm (Fig. 2), DEM 19 cm, cervical perimeter at 33 cm (Fig. 5), no micrognathia, lower lip with eutrophic scar. Cardiac examination showed no murmurs, rubbing or added sounds; pulmonary examination showed good ventilation. Abdominal examination: gravid uterus 12 cm long, intestinal sounds were present; eutrophic limbs, capillary refill greater than 2 s, paleness, symmetrical pulses. At the time of admission, the patient provided paraclinical reports: Hematocrit 25.5%, Hemoglobin 8.3 g/dl, Blood type A Rh negative; oropharyngeal contrasted angioresonance results: global volume increase caused by enlarged arterial, capillary and venous structures due to a hemangioma in the left half of the tongue.

During assessment at the Otolaryngology service the patient had profuse active bleeding which worsened while she chewed her food. She was then taken to the operating room for



Fig. 4 – Tongue of purple color, with diffuse ulcerative areas.

tracheostomy and securing the airway. Pre-anesthetic assessment classified the patient as ASA II/VI-E.

Anesthetic procedure description

The patient was admitted into the operating room with active bleeding of a giant hemangioma involving her tongue and lip. Non-invasive monitoring was carried out with (heart rate, respiratory rate, blood pressure, O₂ saturation). Local anesthesia was carried out in right nasal fossa with local nasal oximetazoline and remifentanyl at 0.02 mcg/kg/min. We then introduced the fibrobronchoscope through the right nasal fossa (Fig. 6), out the choana and through the glottis. A No. 5.5 Fr. Nasotracheal tube was introduced. Tube placement in the airway is confirmed through a capnography and symmetrical ventilation. The airway blocking balloon was inflated with 5 cc of air until leaking. Anesthesia was carried out with tiopental 250 mg, Rocuronium/mg: 10–20. Balanced anesthesia continued with Sevoflurane at half MAC and Remifentanyl at 0.02–0.05 µg/kg/min. The tracheostomy was carried out with a 7.5 Fr cannula. The procedure carried on and ended with no technical complications.



Fig. 3 – Mass in tongue, fleshy, extended over the entire visible surface of the tongue, left dominance.



Fig. 5 – Cervical perimeter (34 cm).



Fig. 6 – Introduction of the fibrobronchoscope through the right nostril.

Discussion

Congenital hemangiomas appear in neck and head in un 56–60% of cases, and those that appear in the oral cavity are generally located in the tongue (affecting it partially or totally), lips, under the tongue or the palate.^{1,10,11} We have considered a few cases published in literature on bleeding hemangiomas in pregnant women, but more often in the vertebrae,^{4,5,12,13} some in the nose¹⁴ and other gastrointestinal hemangiomas.¹⁵ However, we have not identified a case of a bleeding hemangioma of the tongue with a difficult airway in a pregnant patient. During pregnancy, recurrent bleeding occurs with a higher frequency during the second and third trimesters, reaching its peak at 32 weeks. In this case, however, bleeding began in the first trimester.⁵

Congenital hemangiomas are more common in female patients. If they are not treated properly, as in the case of our patient, there may be significant growth during pregnancy, increasing the risk of non-obstetric surgery and with it the use of anesthetic drugs as needed in our case due to its location. No anesthetic drugs have been proven to have teratogenic or abortive effects in humans.¹⁶ In the case of our patient, remifentanyl was used as part of balanced anesthesia. There is evidence in several trials that support the use of remifentanyl in pregnant patients because of its safety.^{11,16–18} Also, remifentanyl does not increase histamine release, making it safer for use in pregnancy due to the hemodynamic changes it that pregnancy causes. Several studies and some case reports have shown that remifentanyl is safe for both the mother and the fetus.¹⁹ A case report by Fuentes et al.¹⁶ used remifentanyl, isoflurane and fentanyl in oncologic surgery in a pregnant patient at 18 weeks. Even though the literature mentions risk of low birth weight after the use of anesthetics, this patient's follow-up did not report any alteration of the fetus. The infant was delivered after 41 weeks of pregnancy, weight was normal and without evidence of malformation. The report concluded that Remifentanyl allowed hemodynamic stability and quicker reawakening. Our patient was assessed by the gynecology-obstetrics service for integral management four days later.

Transvaginal ultrasound was carried out and no abnormalities were reported, the patient will continue under surveillance.

On the other hand, fibrobronchoscopy is clearly useful for difficult airway management. There are reported cases of giant tongue hemangiomas in adults.^{20,21} Securing the airway was necessary and nasotracheal intubation by fibrobronchoscopy. Among difficult airway predictors there is: (short neck, thick or muscled, retracted jaw, arched palate, long or ogival; facial or cervical scars, dental conditions and tongue size.^{14,22} Another issue is that manipulation of the hemangioma in the oral cavity at the time of orotracheal intubation may increase the risk of bleeding. Our patient was admitted with profuse bleeding and was classified as Mallampati IV (with laryngoscope placing difficulty) and was pregnant as well, which implies hormonal and physiologic changes involved in difficult airway. Because of this, nasotracheal intubation with fibrobronchoscope was highly indicated.

An alternative would have been to carry out the tracheostomy using local anesthesia. However, this was not an adequate decision considering the patient had a lightly involved hemangioma in the neck and its true extension was unknown. Therefore, the risk of bleeding during the tracheostomy was unknown as well and the airway had to be secured. Also, transtracheal local anesthesia may inhibit the airway's protective reflexes.

Conclusion

Pregnancy is associated to congenital hemangioma growth as well as increased risk of bleeding. For that reason, congenital hemangiomas in female patients must be managed aggressively and early. Hemangiomas of the tongue are important because of its close relation to the airway and the possibility of affecting it.

The use of remifentanyl for balanced anesthesia in non-obstetric surgery in pregnancy can be used because of its evidence based safety and its pharmacokinetic properties. It can also be used as a complimentary drug to reduce the MAC of inhaled anesthetics such as Sevoflurane.

Conscious intubation with fibrobronchoscope is indicated in patients with a difficult airway who require surgical approach.

Consent

Informed consent was obtained from the patient's family for publication of this case, as well as complementary images. A copy of the document is available for revision for whom it may concern.

Funding

Authors' own resources.

Conflict of interest

Authors declare there are no relevant interests in this report.

Contributions

All data analysis and interpretation were assessed by the authors. The final draft of this case report was reviewed and approved by the authors.

Acknowledgement

The patient's family, to whom we offer our full support.

REFERENCES

1. Domínguez J, Torres D, Gutiérrez J. Extirpación de hemangioma bucal en mucosa yugal mediante el laser de. SECIB. 2011;2:1-9 [on line], Available from: http://www.secibonline.com/web/pdf/vol2.2009.articulo_actualizacion1.pdf
2. Bonet C, Mínguez I, Palma C, Galán S, Peñarrocha M, Mínguez J. Clinical characteristics, treatment and outcome of 28 oral haemangiomas in pediatric patients. *Med Oral Patol Oral Cir Bucal*. 2011;16:19-22.
3. Qureshi SS, Chaukar DA, Pathak KA, Sanghvi VD, Sheth T, Merchant NH, et al. Hemangioma of base of tongue. *Indian J Cancer*. 2004;41:181-3.
4. Vijay K, Shetty A, Rajasekaran S. Symptomatic vertebral hemangioma in pregnancy treated antepartum. A case report with review of literature. *Eur Spine J*. 2008;17 Suppl. 2:S299-303.
5. Tekkök IH, Açıkgöz B, Sağlam S, Onol B. Vertebral hemangioma symptomatic during pregnancy—report of a case and review of the literature. *Neurosurgery*. 1993;32:302-6.
6. Melendez H, Leal D, Ramirez D. Agreement between the objective and subjective evaluation for predicting and finding a difficult airway. *Rev Colomb Anesthesiol*. 2010;38:34-49.
7. Pandit JJ. Orotracheal fiberoptic intubation for rapid sequence induction of anesthesia. *Anesthesia*. 2002;57:123-7.
8. Mariscal M, Pindado M, Ortigosa E. Fibrobroncoscopio flexible. In: Pindado M, Mariscal M, editors. *Vía Aérea difícil*. Madrid: Ergon; 2000.
9. Riveros E. Validation of the clinical usefulness of an articulated orotracheal catheter in the operating room and intensive care. *Rev Colomb Anesthesiol*. 2011;39:330-9.
10. Pérez M. Tratamiento de hemangiomas y malformaciones vasculares congénitas de región bucal y peribucal con criocirugía con la técnica de microhisopo utilizando nitrógeno líquido [Tesis en Licenciatura]. Universidad Francisco Marroquín Facultad de Odontología: Guatemala; 1993. p. 4.
11. Novoa L, Navarro M, Vieito M, Hernandez J, Arxer A, Villalonga A. Analgesia y anestesia obstétrica con remifentanilo a una paciente con enfermedad de von Willebrand. *Rev Esp Anesthesiol Reanim*. 2003;50:242-4.
12. Wang MT, Shue HN, Ho KY, Yu HS, Juan KH, Fang KT. Nasal septal hemangioma of pregnancy. *Gaoxiong Yi Xue Ke Xue Za Zhi*. 1988;4:426-30.
13. Kiroglu Y, Benek B, Yagci B, Cirak B, Tahta K. Spinal cord compression caused by vertebral hemangioma being symptomatic during pregnancy. *Surg Neurol*. 2009;71:487-92.
14. Noorizan Y, Salina H. Nasal septal haemangioma in pregnancy. *Med J Malaysia*. 2010;65:70-1.
15. Gottlieb K, Coff P, Preiksaitis H, Juvelier A, Fern P. Massive hemorrhage in pregnancy caused by a diffuse cavernous hemangioma of the rectum—EUS as imaging modality of choice. *Medscape J Med*. 2008;10:206.
16. Fuentes R, Sebastianes M, Martínez E, Guerrero M, Torres L. Remifentanilo y cirugía no obstétrica durante el embarazo. *Dol Clin Ter*. 2005;3:11-2.
17. Bocanegra J, Rios A, Aquirre O. Rigid fiberscope Intubation of a patient awake with remifentanil sedation. *Rev Colomb Anesthesiol*. 2010;38:395-401.
18. Costa J, Carvalho D, De Oliveira J, Barrozo A, Santos G. Anestesia Venosa Total para Laringectomia Parcial em Paciente na 28ª Semana de Gestação. Relato de Caso. *Rev Bras Anesthesiol*. 2005;55:217-23.
19. Hill D. Remifentanil in obstetrics. *Curr Opin Anaesthesiol*. 2008;21:270-4.
20. Tasker J, Geoghegan J. Giant cavernous haemangioma of the tongue. *Anaesthesia*. 2005;60:1043.
21. Montero FI, Giménez I, Valentí P, Aguilar JL. Vía aérea difícil en paciente con hemangioma gigante de lengua. *Rev Esp Anesthesiol Reanim*. 2008;55:131.
22. Mariscal ML, Pindado ML, Rey F, Arrasola B, Engel W. Valoración y predicción de la vía aérea difícil; 2012. Available from: <http://www.arydol.es/valoracion-prediccion-via-aerea-dificil.php>