Clinics in Medicine

ISSN: 2688-6731

Case Report

Harlequin Syndrome after Radiofrequency Ablation under Epidural Anesthesia: A Case Report

*Liangrong Wang, Baihui Chen and Xiyue Zhao**

Department of Anesthesiology, The First Affiliated Hospital of Wenzhou Medical University, Wenzhou, China

Abstract

We present a case of harlequin syndrome after thoracic epidural anesthesia following Radiofrequency Ablation (RFA) for liver tumor. The patient under combined epidural and general anesthesia developed reversible, well-demarcated, dextral facial erythema and sweating after surgery, which was accompanied by sudden changes in heart rate and blood pressure. This harlequin feature was due to sympathetic nervous system's thermoregulatory response by the erythema side to excessive heat produced during RFA amidst a left sided epidural block.

Keywords: Erythema; Radiofrequency ablation; Epidural anesthesia; Hyperthermia; Sympathetic nervous system

Introduction

Harlequin syndrome is characterized by sudden onset of asymmetrical facial flushing and sweating, sharply demarcated at the midline, in response to heat, emotion or exercise. The first case of harlequin syndrome was reported in 1988 [1], and since then more and more cases have been found in perioperative period. This report however is most likely to be the first case of hyperthermia response following RFA under epidural block.

Case Presentation

A 61-year-old, 40 kg, American Society of Anesthesiologists physical status II female was scheduled to undergo laparotomy and RFA treatment for primary liver tumors at the right hepatic lobe. She had past history of congenital Hepatitis B Virus (HBV) infection and 1 week history of cirrhosis. The CT scan of the abdomen demonstrated a large occupying lesion in the right liver (55 mm × 61 mm), cirrhosis, portal hypertension and splenauxe. Other preoperative exams revealed no apparent abnormality. After connection to the standard monitors, an epidural block was performed at the T7-T8 interspace, and a catheter was smoothly inserted 4 cm into the epidural space. The patient was then placed in supine position and given a test dose of 3 ml 2% lidocaine epidurally after no aspiration of CSF or blood was seen in epidural catheter. The patient had no signs of total spinal anesthesia and local anesthetic intoxication. General anesthesia was afterwards induced with propofol (60 mg), etomidate (16 mg), sulfentanyl (15 ug) and tracheal intubation was facilitated with cis-atracurium (8 mg). Anesthesia was maintained with hourly epidural bolus of 3 mL 0.375% ropivacaine, sevoflurane 0.5% to 1.5%, propofol 120 mg/h to 200 mg/h and remifentanil 200 μg/h to 400 μg/h. The patient was placed on the supine position with left lateral tilt, scrubbed with povidine and aseptically draped with disposable surgical drapes. A tumor size of about 6 cm in diameter was observed on liver surface with no metastasis or other abnormality noticed in the abdominal cavity. Subsequently, a monopolar needle electrode was introduced into the tumor and a radiofrequency sinusoidal current of 460 kHz was delivered. RFA therapy was conducted on three consecutive times with 15 min exposure per each time. The patient's vitals were stable through-out the whole procedure. At the end of surgery, a 2 ml h^{-1} infusion of 0.15% ropivacaine with a bolus injection of 0.5 ml was commenced for postoperative pain control. After extubation, the patient was finally transferred to PACU.

At recovery, the patient recorded a noninvasive blood pressure of 162/93 mmHg, heart rate of 125 beat per min (bpm), and pulse oxygen saturation of 97%. She also recorded slight increase in temperature (37.9°C) as compared to her initial temperature of 37.0°C before surgery. Sensory blockade was tested up to T4 level through skin prick. Her pupils were round, equal and reacted to light. Approximately 45 min later, the patient developed sudden well-demarcated right facial flushing and sweating with a clear line dividing the face into two regions (Figure 1), which was accompanied by a sharp decrease of her heart rate (30 bpm) and a drop of blood pressure to 140/70 mmHg but with no neurological abnormality. The harlequin feature however disappeared within one hour of discontinuing epidural infusion and continuous physical cooling. Her temperature also returned to normal and was therefore transferred from PACU to ward for further treatment.

Figure 1: The patient experienced right-sided facial flushing following radiofrequency ablation under epidural anesthesia.

Citation: Wang L, Chen B, Zhao X. Harlequin Syndrome after Radiofrequency Ablation under Epidural Anesthesia: A Case Report. Clin Med. 2019; 1(3): 1012.

Publisher Name: Medtext Publications LLC

Manuscript compiled: October 03rd, 2019

***Corresponding author:** Xiyue Zhao, Department of Anesthesiology, The First Affiliated Hospital of Wenzhou Medical University, Wenzhou, 325000, China, Tel: 8613587884540; Fax: 0086-57788069459; E-mail: 544649314@qq.com

Discussion

Harlequin syndrome was first described by Lance as the sudden onset of unilateral facial flushing and sweating with or without Horner's syndrome (ptosis, miosis, enophthalmos and anhydrosis). It occurs in response to excessive temperatures, emotions or strenuous exercises [1]. Harlequin syndrome is believed to result from one-sided blockade of the vasomotor sympathetic fibers originating from T2 to T4 which leads to the contralateral side compensating with excessive sweating and flushing for the impaired ipsilateral side; with anhydrosis and no facial flushing [2,3].

RFA is a thermal coagulation therapy, which uses a heating needle electrode to destroy cell membranes and intracellular proteins, and been widely accepted for treating liver tumors [4]. However, it has been reported that RFA causes systemic heating (the body temperature increased approximately by 1°C for every 3000 J/kg of total output energy from RFA) by the transfer of heat from the intrahepatic blood stream to the cardiopulmonary system and then to the whole body [4]. In addition, breakdown products of proteins and production of cytokines would act as pyrogens and affect the actions of hypothalamic thermoregulatory centers, whose response is somewhat impaired by volatile anesthetics [4].

In our case, temperature regulation seems to be a more convincing starting point to explain the harlequin face and the sudden changes in vital signs observed. We believe the patient went through different stages of temperature regulation. First and foremost, the hypothalamus down regulated the set-point of thermoregulatory system in response to cytokines release and protein degradation. This was followed by cutaneous vasodilatation, diaphoresis, and other heat loosing processes that occur when the thermoregulatory center is signaled [5,6]. The unexpected unilateral sympathetic block following thoracic epidural anesthesia serves as an indispensable co-factor in harlequin syndrome, due to blockade of vasomotor sympathetic fibers in the left face, heat loosing processes (Cutaneous Vasodilatation and Diaphoresis) were impaired, leading to compensation by the right side. However, sensory block was tested to be bilaterally equal in the patient, it perhaps attribute to the subjective judgment of pain when the patient was tested, and the favorite approach to testing pain perception through stimulating skin might be considered inaccuracy. The changes in heart rate and blood pressure observed in our case could be explained by drop in body temperature.

Conclusion

In conclusion, hyperthermia associated with RFA under thoracic epidural block can act as contributing factor to bring about harlequin syndrome. Furthermore, close monitoring of body temperature and preparations for body cooling are important for RFA of liver tumors under epidural block. We also suggest further studies into the harmful effect of hyperthermia in the flushing side.

References

1. Lance JW, Drummond PD, Gandevia SC, Morris JG. Harlequin syndrome: The sudden onset of unilateral flushing and sweating. J Neurol Neurosurg Psychiatry. 1988;51(5):635-42.

2. Burlacu CL, Buggy DJ. Coexisting harlequin and Horner syndromes after high thoracic paravertebral anaesthesia. Br J Anaesth. 2005;95(6):822-4.

3. Thomas JJ, Polaner D. Harlequin syndrome associated with thoracic epidural analgesia. Anesthesiology. 2015;123(5):1187.

4. Sawada M, Watanabe S, Tsuda H, Kano T. An increase in body temperature during radiofrequency ablation of liver tumors. Anesth Analg. 2002;94(6):1416-20.

5. Mekjavic IB, Eiken O. Contribution of thermal and nonthermal factors to the regulation of body temperature in humans. J Appl Physiol. 2006;100(6):2065-72.

6. Hemingway A, Price WM. The autonomic nervous system and regulation of body temperature. Anesthesiology. 1968;29(4):693-701.