The severe adverse event in a locally anesthetized circumcision: A case report of a breath-holding spell

Syifa Fauziah Fadhly*, Irfan Wahyudi, Gerhard Reinaldi Situmorang, Arry Rodjani

ABSTRACT

Introduction: Circumcision in children can be done with local anesthesia. Indifferent from other surgeries, circumcision certainly has its own risks dan complications.

Case description: A 5-month-old baby who underwent circumcision with local anesthesia was reportedly losing consciousness intraoperatively, followed by a generalized seizure. No previous medical abnormalities were known. It was thought that a breath-holding spell (BHS) was a possible etiology of this event. The patient regains consciousness after a few hours. Electroencephalogram showed normal results. BHS is a harmless condition, where it is believed that crying may cause children to be hypoxic due to prolonged breath-holding and may lead to loss of consciousness or even seizure. Patients with BHS usually recover and regain consciousness after several minutes and should not have any long-term effects after the episode.

Conclusion: Although considered a benign condition, BHS is still a frightening situation for parents and even the surgeon; thus, good communication with the patient’s parents is necessary.

Keywords: breath-holding spell; circumcision; children


INTRODUCTION

Circumcision has been considered part of the Muslim tradition, which makes up a large percentage of Indonesia’s population. It generally can be performed on patients at any age; however, most are done at a young age. There are types of anesthesia options for circumcision: local, regional, or general anesthesia. In local anesthesia, the anesthetic agent is injected to block the dorsal penile nerve at the base of the penis to numb the surgical area. The patient is fully conscious thus may revolt during the procedure, making general anesthesia a more comfortable choice for a surgeon in doing circumcision. Nevertheless, infants should age at least over six months old, which is thought to have adequate development of body organs already to undergo general anesthesia safely.

Either way, any medical procedure has its risks and benefits, and no exception to circumcision.

CASE DESCRIPTION

A circumcision procedure was performed on a five-month-old baby with local anesthesia. It was requested by the patient’s parents beforehand to do local anesthesia to avoid the use of intubation for their baby. The patient was known to have no heart, lungs, or any other abnormalities, with all supportive examinations being unremarkable.

The circumcision was done in an operating theatre with an experienced surgeon. Penile block anesthesia was done using a lidocaine injection. The procedure was without a problem at the beginning. However, as expected, the patient cried and was fussy during the process. Near the end of the procedure, during the suturing of penile skin to the mucosa, the patient suddenly lost consciousness and experienced a generalized seizure for about 5-10 minutes. The procedure was stopped, and the anesthesiologist was called for help. The patient was then intubated and stabilized.

The anesthesiologist thought that a breath-holding spell was the etiology of this event. After a few hours, the patient started to wake up and cry. With the evaluation from the anesthesia team that spontaneous breathing had recovered, the patient was extubated. However, it was suggested that the patient still be monitored in a pediatric intensive care unit (PICU) at least for the next 24 hours.

During the hospitalization, the patient was stable, with no episodes of seizure or unconsciousness. The patient underwent an electroencephalogram (EEG) to evaluate brain electrical activity and detect any abnormalities. Fortunately, EEG showed normal results. The patient was discharged on the second day after surgery.

After a few days of follow-up, the patient’s parents still felt traumatized and refused on do further investigations on their baby.
CASE REPORT

DISCUSSION
A breath-holding spell may occur mainly in children aged less than 12 months.\textsuperscript{2} It is a harmless defense mechanism that mostly happens to children, where they lose consciousness that is often triggered by anger, frustration, or pain. It often causes psychological trauma, especially to the parents.\textsuperscript{3}

Breath-holding spell clinical manifestations are usually classified as cyanotic or pallid, based on the patient's face/skin coloration. Cyanotic is the most common BHS consisting of 72\% of BHS cases.\textsuperscript{4} The pathogenesis of BHS is not fully understood and is thought to be multifactorial, ranging from autonomic dysregulation, vagally-mediated cardiac inhibition, or delayed myelination of the brain stem.\textsuperscript{3} Young children showed their emotional responses as frustration and anger, mostly in the form of crying. It is thought that, at times, the crying may cause children to be hypoxic due to prolonged breath-holding due to forced expiration and may lead to loss of consciousness or even seizure.\textsuperscript{3}

BHS typically lasts for less than one minute, but patients may experience a prolonged seizure in some cases. Patients with prolonged seizures should be evaluated for other differential diagnoses. In this patient, it is thought that it might not be due to epilepsy as there was no history of previous seizures, and the EEG showed the normal result. Another differential diagnosis that should be considered is arrhythmia, such as prolonged QT syndrome. Prolonged QT syndrome may be manifested as an anoxic seizure. ECG should be obtained to exclude this condition.\textsuperscript{3}

Patients with BHS usually recover and regain consciousness after several minutes, just like the patient discussed in this report, and should not have any long-term effects after the episode.\textsuperscript{4} EEG that was done one day after the event showed normal results. Even though the patient's parents refused to do further investigation on their baby, the patient is currently healthy and does not show any delayed development in his milestone.

Lidocaine allergy was also considered a differential diagnosis of this event. Yet, it seemed more unlikely that allergy or hypersensitivity due to lidocaine is extremely rare and usually presents as urticaria, angioedema, or allergic dermatitis.\textsuperscript{3} Symptoms should appear soon after the injection. However, there were no signs of rash, erythema, or angioedema seen in this patient.

CONCLUSION
BHS should be considered a differential diagnosis if loss of consciousness or seizure occurs in children, preceded by any emotional stimuli such as anger, frustration, or pain. If the seizure lasted more than one-minute, other diagnoses should be excluded; thus, further evaluations are needed.

From this case, we learn that it is important to be aware of the presence of BHS as a possible risk in any surgery with local anesthesia in children; thus, health care workers can deal with this kind of situation more calmly. Although considered a benign condition, it is still a frightening situation for parents and even the surgeon. Good communication with the patient's parents is necessary.

ACKNOWLEDGEMENT
All authors contributed to the creation of the manuscripts.

ETHICAL CLEARANCE
Patient approval has obtained in this study and fulfilled ethics approval from International Committee of Medical Journal Editors (ICMJE).

CONFLICT OF INTEREST
We declare that there were no conflicts of interest in this study.

FUNDING
The authors are responsible for the study funding without the grant, scholarship, or other funding resources.

AUTHOR CONTRIBUTION
All of the authors are equally contributed to the study.

REFERENCES