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CLINICAL INFORMATION

''Loss of breath'' as a cause of postoperative hypoxia and bradycardia in children submitted to tonsillectomy

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Abstract
Background and objectives: the ''shortness of breath'' or ''breathing interruption'' crisis can be considered a cause of hypoxia in childhood. It is characterized by the presence of a triggering factor followed by weeping and apnea in expiration accompanied by cyanosis or pallor. The sequence of events may include bradycardia, loss of consciousness, abnormal postural tone and even asystole. A review of the literature revealed only two reports of postoperative apnea caused by ''shortness of breath''.
Case report: this article describes the case of a child with a history of ''shortness of breath'' undiagnosed before the adenotonsillectomy, but that represented the cause of episodes of hypoxemia and bradycardia in the postoperative period.
Conclusions: the ''shortness of breath'' crisis should be considered as a possible cause of perioperative hypoxia in children, especially when there is a history suggestive of this problem. As some events may be accompanied by bradycardia, loss of consciousness, abnormal postural tone and even asystole, observation in a hospital setting should be considered.

PALAVRAS-CHAVE
Apneia;
Perda de fôlego;
Criança;
Hipóxia;
Bradicardia;
Recuperação pós-anestésica

''Perda de fôlego'' como causa de hipóxia e bradicardia pós-operatória em criança submetida à amigdalectomia

Resumo
Justificativa e objetivos: A crise de ''perda de fôlego'' ou de ''interrupção respiratória'' pode ser considerada uma causa de hipóxia na infância. É caracteriza pela presença de um fator desencadeante seguido por choro e apneia em expiração acompanhada de cianose ou palidez-cutânea. A sequência de eventos pode incluir bradicardia, perda da consciência, alteração do tônus postural e até assistolia. Uma revisão da literatura evidenciou apenas dois relatos deapneia pós-operatória causada por ''perda de fôlego''.

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Relato do caso: O presente artigo descreve um caso de criançaa com antecedente de crises de ''perda de fôlego'' não diagnosticadas antes da feitura de adenoamigdalectomia, mas que representaram a causa de episódios de hipoxemia e bradicardia no período pós-operatório.
Conclusões: As crises de ''perda de fôlego'' devem ser consideradas como possível causa de hipóxia perioperatória em criançaas, principalmente quando há história prévia sugestiva. Como alguns eventos podem ser acompanhados de bradicardia, perda da consciência, alteração do tônus postural e até assistolia, a observação em ambiente hospitalar deve ser considerada.

Introduction

The ''shortness of breath'' or ''breathing interruption''' crisis can be considered a cause of hypoxia in childhood. It is characterized by the presence of a triggering factor such as anxiety, fear, pain or frustration, followed by weeping and apnea in expiration accompanied by cyanosis or pallor. The sequence of events may include bradycardia, loss of consciousness, abnormal postural tone and even asystole.[1,2] A review of the literature revealed only two reports of postoperative apnea caused by ''shortness of breath''.[3,4]

Case report

Male child, 1 year and 11 months of age, evaluated in preanesthetic consultation for an adenotonsillectomy, showed no abnormalities suggestive of systemic disease. Premedication with midazolam (0.5 mg kg^{-1}) PO 20 min before the procedure. In the operating room the patient was monitored with electrocardiogram (DII), pulse oximetry and noninvasive blood pressure. After administration of mixture of O_2 by inhalation and N_2O 60% and sevoflurane 6% by face mask, a catheter 24G was inserted by venoclysis after induction of anesthesia with remifentanil in continuous infusion (0.5 μg kg^{-1} min^{-1}), propofol 3 mg kg^{-1} and cisatracurium 0.1 mg kg^{-1}. Tracheal intubation and insertion of a tube of 4 mm with balloon were done uneventfully. The patient was maintained on mechanical ventilation in a closed system with reabsorption of CO_2 and then monitoring of expired carbon dioxide ($PetCO_2$) was added. The anesthesia was maintained with remifentanil (0.3 μg kg^{-1} min^{-1}) and sevoflurane 1–1.5%. The surgery lasted 40 min without complications. The child presented a calm awakening, about 10 min after stopping the infusion of anesthetic agents. Postoperative analgesia was introduced with morphine (0.1 mg kg^{-1}) and dipyrone (30 mg kg^{-1}). The child was transferred to the postanesthesia recovery room (PARR), where it stayed with O_2 face mask and monitoring with pulse oximetry. During the permanence in PARR and after the expected initial agitation for the first 30 min after the procedure, the patient had periods of calm and agitation, without evident bleeding and with a pain considered of mild to moderate intensity. Approximately 3 h after the transference to PARR, the child showed cyanosis and muscle hypertonia which resolved spontaneously after a few seconds. The episode, characterized by apnea during crying, cyanosis, SpO_2 70%, bradycardia (37 bpm) and loss of consciousness, recurred after 2 h. The patient, with no signs of airway obstruction, received naloxone 0.4 mg intravenously. However, the patient still experienced two episodes with the same characteristics, always during crying spells and after apnea during expiration. The child was then sent to the Intensive Care Unit, where he remained for 12 h. In this period he showed some sporadic bouts of crying accompanied by a decrease in SpO_2. During the hospitalization, the echocardiogram and electrocardiogram were normal. The child was discharged with a diagnosis of ''shortness of breath'' crisis. According to the mother, since three months of age the child had crises characterized by apnea during crying, accompanied by lip cyanosis. As the episodes were infrequent, she did not seek medical help. One week after surgery, the child experienced another crisis characterized by pallor and decreased postural tone.

Discussion

This article describes the case of a child with a history of ''shortness of breath'' crisis undiagnosed before the adenotonsillectomy, but that represented the cause of episodes of hypoxemia and bradycardia in the postoperative period. The diagnosis of ''shortness of breath'' crisis is based on the report of three or more episodes characterized by the presence of a triggering factor such as anxiety, fear, pain or frustration, followed by weeping and apnea in expiration accompanied by cyanosis or pallor.[1,2] The ''loss of breath'' crisis can be considered as paroxysmal, non-epileptic and involuntary events that may occur during childhood.[1] The incidence of children with a history of ''shortness of breath'' crisis ranges from 0.1% to 4.6%.[1] It has been shown that there is an autosomal dominant trait with reduced penetrance in a considerable proportion of patients.[5] According to Di Mario,[1] in a prospective study of 95 children with a history of ''shortness of breath'' crisis'', in 34% of diagnosed cases there had been a close relative with a history of similar crises, which was not observed in our paper. According to the mother of the child, the episodes were initially observed at three months of age, but as they were sporadic, triggered by situations of fear or anxiety with spontaneous regression, no referral for diagnostic investigation was made. Among the cases evaluated by Di Mario, 47 boys and 48 girls were identified with a history compatible with ''shortness of breath'' crisis; of those, 49 were characterized by cyanosis, 27 by pallor and 19 had both signals. Seizures precipitated by hypoxia were observed in 15 and syncope in 12 of 95 children. In all cases the electrocardiogram was normal, which excludes the possible differential diagnosis of QT prolongation. Most children presented the first crisis between six and 12 months of age, but in 12% of cases the first episode occurred before.

According to Di Mario study, in 5% of children the crises began when they were just newborns.

In our case, possible differential diagnoses, as the residual effect of opioids or edema in the surgical field, were discarded by the administration of naloxone and by direct visualization of the larynx. Although the frequency of crises is described as daily or weekly, in some children the interval between episodes may be greater than one month. The boys seem to have a more early peak frequency (13–18 months) compared to girls (19–24 months).[1] According to Bridge,[6] who followed for nine years 83 children with a history of ''shortness of breath'' crisis, approximately half of them showed the last episode at four years of age.

The pathophysiology of the cyanotic variant of ''shortness of breath'' crisis probably has multiple factors not yet fully known. These include hyperventilation (with consequent excessive reduction in $PaCO_2$), apnea and decreased venous return induced by the Valsalva maneuver.[7] Loss of consciousness seems to be the result of changes in autonomic regulation, because it has been shown that these children have the ocular-cardiac reflex exacerbated, which may be responsible for the asystole observed in 61% of cases of ''shortness of breath'' crisis accompanied by pallor (also called cardioinhibitory syncope) and in 25% of those characterized by cyanosis.[2,8] Although in our case the heart rate has reached values below 40 bpm, bradycardia associated with ''shortness of breath'' crises has been considered severe when the heart rate is <20 bpm or asystole occurs for more than 6 s.[2] When symptomatic, the bradycardia has been considered as an indication for pacemaker implantation.[2] There is no specific treatment for ''shortness of breath'' crisis. As a non-epileptic event, there is no favorable response after administration of anticonvulsants.[6] There is a need to properly educate the parents and, in the case of children undergoing anesthesia, these authors believe that its observation in the hospital for at least 12 h is important, because this is a period in which countless possible triggering factors for the crises are involved, and there is no way to predict whether there will be hemodynamic changes or loss of consciousness in the possible events that will occur postoperatively.

To conclude, although considered benign, the ''shortness of breath'' crisis should be considered as a possible cause of perioperative hypoxia in children, especially when there is a suggestive history. Considering that some events may be accompanied by bradycardia, loss of consciousness, abnormal postural tone and even asystole, observation in a hospital setting should be considered.

Conflicts of interest

The authors declare no conflicts of interest.

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