


CASE REPORT

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# Complete atrio-ventricular block with coronary artery spasm due to direct laryngoscopy in a pediatric patient with laryngeal papillomatosis: a case report

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## Abstract

**Background:** Coronary artery spasm has rarely been reported in pediatric patients. Previous studies have reported comorbidities and risk factors for coronary artery spasms. We present the case of a complete atrio-ventricular (AV) block that occurred in the absence of other risk factors immediately after direct laryngoscopy.

**Case presentation:** A 2-year-old girl developed severe coronary artery spasm after direct laryngoscopy for elective laryngeal papillomatosis resection. Immediately after the initiation of laryngoscopy, complete AV block and ST elevation on lead II of the electrocardiogram were observed. These findings indicated that the complete AV block was caused by a right coronary artery spasm.

**Conclusion:** Coronary artery spasm resulting in lethal arrhythmia rarely occurs in healthy pediatric patients. To the best of our knowledge, this is the first pediatric case of a severe coronary artery spasm resulting in a complete AV block due to direct laryngoscopy in a healthy patient.

**Keywords:** Coronary artery spasm, Direct laryngoscopy, Atrio-ventricular block, Laryngeal Papillomatosis

## Background

Coronary artery spasm has rarely been reported in pediatric patients [1–5]. Patients with a history of coronary artery spasm have comorbidities such as Kawasaki disease [4], systemic lupus erythematosus [5], and heart disease [2, 3]. A previous review [6] revealed that perioperative coronary spasm occurred in men with associated risk factors; however, none of the patients were under 30 years of age. Vascular smooth muscle hyperactivity and endothelial dysfunction have been described as the main etiologies of coronary artery spasms. Furthermore, the role of an imbalance between parasympathetic and sympathetic tone in triggering coronary

artery spasm is well known [7]. Due to an immature autonomic nervous system, children often experience hypervagotonia. In the perioperative period, nasopharyngeal or esophageal stimulation, such as nasogastric or endotracheal tube placement, often stimulates the vagus nerve; however, bradycardia resulting from such stimulation is usually transient and rarely turns severe. Herein, we report a pediatric patient without comorbidities who developed a complete atrio-ventricular (AV) block with right coronary artery spasm induced by direct laryngoscopy. Written informed consent was obtained from the parents of the patient for the publication of this report. This manuscript adheres to the CARE guidelines.

## Case presentation

A 2-year-old girl (height, 91.3 cm; weight, 12.2 kg) presented to our hospital with hoarseness of voice, which

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began soon after the patient started speaking. Dyspnea, laryngeal stridor, and snoring were absent. Laryngeal papillomatosis was diagnosed, and the patient was scheduled for a resection of the lesion. The patient did not have any significant medical history, and preoperative examinations, including chest radiography and electrocardiogram (ECG), were normal (Fig. 1). The preoperative blood pressure was 102/58 mmHg, and the heart rate was 110 beats/minute.

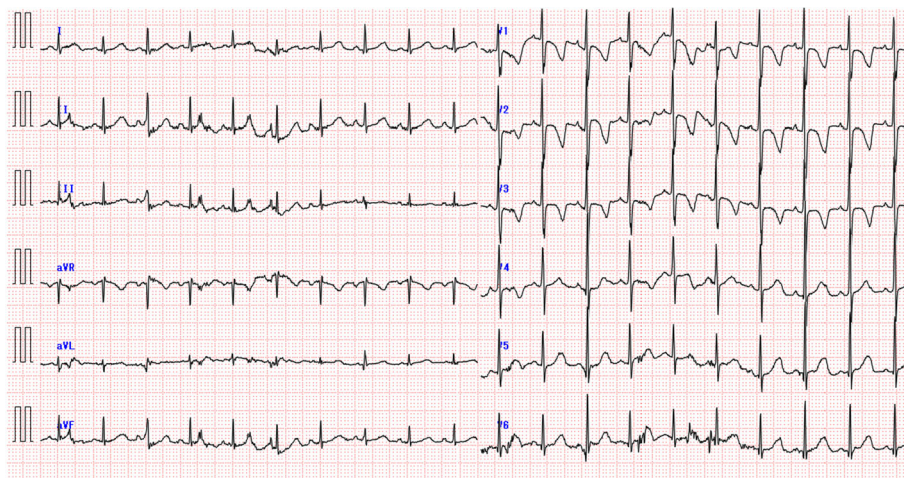
Inhalational anesthesia was induced with 5% sevoflurane and 50% nitrous oxide in oxygen. After the intravenous administration of suxamethonium, tracheal intubation was performed with an 8.0-Fr airway exchange catheter (AEC) for jet ventilation during surgery. Anesthesia was maintained with continuous infusion of remifentanyl ( $0.3 \mu\text{g kg}^{-1} \text{min}^{-1}$ ) and propofol ( $10 \text{ mg kg}^{-1} \text{h}^{-1}$ ). A bolus dose of rocuronium was administered as required. Arterial pressure and bispectral index (BIS®) monitors were used in addition to standard monitors. Jet ventilation via AEC was adjusted on the basis of the arterial blood gas readings. The rate of remifentanyl infusion was increased to  $0.5 \mu\text{g kg}^{-1} \text{min}^{-1}$  2 min before the surgery, and direct laryngoscopy for laryngeal papillomatosis resection was initiated by the surgeon. Immediately after the larynx was exposed with a laryngoscope, the patient's heart rate increased from 77 beats/minute to 101 beats/minute. After 1 min, a complete AV block with ST segment elevation on lead II of the ECG and reduction in the arterial blood pressure from 128/84 mmHg to 49/35 mmHg occurred. The minimum heart rate was 47 beats/minute (Fig. 2). Laryngoscopy was aborted, and remifentanyl infusion was stopped. Atropine (0.5 mg) was administered, and a continuous infusion of trinitroglycerin ( $0.5 \mu\text{g kg}^{-1} \text{min}^{-1}$ ) was initiated. The complete AV block and ST segment elevation were reversed 14 min later. Right coronary artery spasm due to

direct laryngoscopy was suspected; therefore, trinitroglycerin infusion was continued, and remifentanyl infusion was restarted at an increased rate ( $0.6 \mu\text{g kg}^{-1} \text{min}^{-1}$ ). Under these conditions, laryngoscopy was attempted a second time. No ECG or hemodynamic abnormalities were observed, and the surgery was completed without further problems. No abnormalities were observed during the postoperative echocardiographic examination, and the postoperative ECG was the same as the preoperative ECG. The patient was discharged 2 days after the surgery.

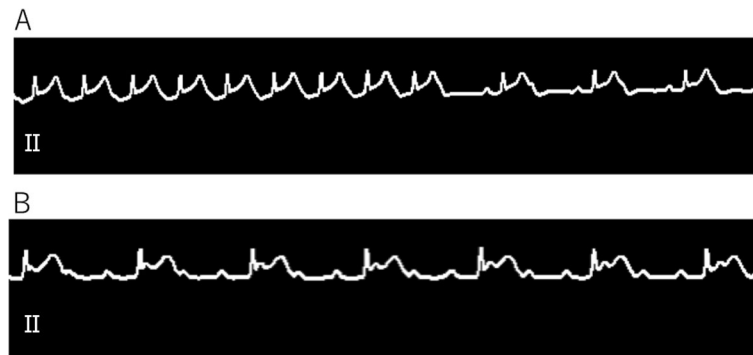
## Discussion

Coronary artery spasm is defined as intense vasoconstriction of the coronary arteries that causes total or subtotal vessel occlusion. It plays an important role in myocardial ischemic syndromes, including stable and unstable angina, acute myocardial infarction, and sudden cardiac death [8]. The characteristics of coronary artery spasm are that it occurs at rest and is not provoked by exertion, is associated with ST segment elevation on the ECG, and is often more prolonged than ischemic angina pectoris [9]. The mechanisms underlying coronary artery spasm remain unexplained; however, autonomic nervous system imbalance and endothelial dysfunction are known to be significant factors.

Coronary artery spasm is recognized as one of the causes of sudden death in adults; however, it is rarely observed in pediatric and adolescent patients in the absence of confounding risk factors [10]. Few cases of coronary artery spasm have been reported in pediatric patients. Subramanian et al. reported that a 15-month-old girl without coronary aneurysm developed acute myocardial infarction 3 months after treatment for Kawasaki disease [4]. Jasmin et al. reported that a 13-year-old girl with systemic lupus erythematosus



**Fig. 1** Pre-operative 12-lead electrocardiogram revealing normal sinus rhythm with normal QRS complexes and ST segments



**Fig. 2** Electrocardiogram monitor showing ST-segment elevation and bradycardia after initiation of laryngoscopy (a). Complete atrio-ventricular block with ST-segment elevation is observed (P waves with a regular P-to-P interval, QRS complexes with a regular R-to-R interval, and variable PR interval) on the electrocardiogram monitor (b)

experienced left-sided chest pain, ECG changes, and troponin elevation, which were suggestive of acute myocardial infarction, possibly due to coronary artery spasm. In these cases, the patients had comorbidities and risk factors related to endothelial dysfunction or small vessel disease, which may have led to coronary artery spasm. In our case, a complete AV block occurred in the absence of other risk factors after a direct laryngoscopy was performed by the surgeon. Simultaneously, ST elevation in lead II of the ECG was observed. These clinical findings suggest that vagal stimulation due to direct laryngoscopy resulted in right coronary artery spasm, which decreased the blood flow to the AV node and caused a complete AV block. The branches of the vagus nerve are distributed in the pharyngeal and laryngeal mucosa. Mechanical stimulation by the laryngoscope or tracheal tube stimulates the vagal cardiac branch through the medullary vagal nucleus, which induces bradycardia [11].

Moreover, propofol and remifentanyl induce bradycardia, which has an inhibitory effect on the sympathetic nervous system [12]. In the present case, vagal stimulation by direct laryngoscopy and the use of propofol and remifentanyl led to an imbalance between the parasympathetic and sympathetic nervous systems, which may have caused severe right coronary artery spasm and the complete AV block. We believe that the infusion rates of remifentanyl and propofol were sufficient for direct laryngoscopy; however, pediatric patients might need more profound anesthesia than adults, and we should have been more careful while performing direct laryngoscopy.

Pediatric life support algorithm suggests the consideration of atropine administration for bradycardia caused by an increased vagal tone [13]. However, we should have considered epinephrine administration or the use of an external pacemaker because atropine, which acts at the AV node, is rarely effective in patients with a complete AV block. Continuous infusion of

trinitroglycerin might have worked prophylactically against coronary artery spasm during the second laryngoscopy.

We searched the PubMed database on 27 March 2021 using the following keywords: <coronary artery spasm or coronary vasospasm> AND <child> or <coronary artery spasm or coronary vasospasm> AND <complete atrio-ventricular block>. A total of 109 papers and 38 papers were found respectively, but reports describing coronary artery spasms secondary to direct laryngoscopy in healthy pediatric patients were not found. We additionally checked the references cited in these papers and found no reports on pediatric patients suffering from a coronary artery spasm following a direct laryngoscopy. Therefore, to the best of our knowledge, this is the first report of a severe coronary artery spasm in a healthy pediatric patient at the time of direct laryngoscopy.

#### Abbreviations

AV: Atrio-ventricular; ECG: Electrocardiogram; AEC: Airway exchange catheter; BIS: Bispectral index

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#### Authors' contributions

KM conceptualized and designed the case report, and drafted and revised the manuscript. RA conceptualized and designed the case report and cared for the patient. TN assisted with the planning and execution of the anesthesia procedure. TK helped draft and revise the manuscript. KY helped draft and revise the manuscript. All authors read and approved the final manuscript.

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#### Availability of data and materials

Not applicable.

#### Declarations

#### Ethics approval and consent to participate

Not applicable.

**Consent for publication**

Written informed consent was obtained from the patient's parents for publication of this case report and accompanying images.

**Competing interests**

The authors declare that they have no competing interests.

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**References**

- Castellano-Martinez A, Rodriguez-Gonzalez M. Coronary artery spasm due to intravenous atropine infusion in a child: possible Kounis syndrome. *Cardiol Young*. 2018;28(4):616–8. <https://doi.org/10.1017/S1047951117002785>.
- Ferguson ME, Pearce FB, Hsu HH, Misra VK, Kirklín JK. Coronary artery spasm during angiography in a pediatric heart transplant recipient: subsequent prevention by intracoronary nitroglycerin administration. *Tex Heart Inst J*. 2010;37(4):469–71.
- Bansal N, Delius RE, Aggarwal S. Coronary artery spasm in a neonate with transposition of great arteries: a rare complication and reason for heart transplant. *Cardiol Young*. 2018;28(1):163–7. <https://doi.org/10.1017/S1047951117001652>.
- Subramanian S, Gaum WE. Acute myocardial infarction caused by transient coronary vasospasm in a child with Kawasaki disease and no coronary aneurysms. *Pediatr Cardiol*. 2010;31(6):875–7. <https://doi.org/10.1007/s00246-010-9712-y>.
- Jasmin R, Ng CT, Sockalingam S, Yahya F, Cheah TE, Sadiq MA. Myocardial infarction with normal coronaries: an unexpected finding in a 13-year-old girl with systemic lupus erythematosus. *Lupus*. 2013;22(14):1518–22. <https://doi.org/10.1177/0961203313503913>.
- Koshihara K, Hoka S. Clinical characteristics of perioperative coronary spasm: reviews of 115 case reports in Japan. *J Anesth*. 2001 Apr;15(2):93–9. <https://doi.org/10.1007/s005400170034>.
- Yasue H, Touyama M, Shimamoto M, Kato H, Tanaka S, Akiyama F. Role of autonomic nervous system in the pathogenesis of Prinzmetal's variant form of angina. *Circulation*. 1974 Sep;50(3):534–9. <https://doi.org/10.1161/01.CIR.50.3.534>.
- Hung MJ, Hu P, Hung MY. Coronary artery spasm: review and update. *Int J Med Sci*. 2014;11(11):1161–9. <https://doi.org/10.7150/ijms.9623>.
- Yasue H, Mizuno Y, Harada E. Coronary artery spasm—clinical features, pathogenesis and treatment. *Proc Jpn Acad Ser B Phys Biol Sci*. 2019;95(2):53–66, DOI: <https://doi.org/10.2183/pjab.95.005>.
- Lane JR, Ben-Shachar G. Myocardial infarction in healthy adolescents. *Pediatrics*. 2007 Oct 1;120(4):e938–43. <https://doi.org/10.1542/peds.2006-3123>.
- Mu L, Sanders I. Sensory nerve supply of the human oro- and laryngopharynx: A preliminary study. *Anat Rec*. 2000 Apr 1;258(4):406–420, DOI: [https://doi.org/10.1002/\(SICI\)1097-0185\(20000401\)258:4<406::AID-AR9>3.0.CO;2-5](https://doi.org/10.1002/(SICI)1097-0185(20000401)258:4<406::AID-AR9>3.0.CO;2-5)
- Deutschman CS, Harris AP, Fleisher LA. Changes in heart rate variability under propofol anesthesia: a possible explanation for propofol-induced bradycardia. *Anesth Analg*. 1994 Aug 1;79(2):373–7. <https://doi.org/10.1213/0000539-199408000-00031>.
- Ian K Maconochie, Richard Aickin, Mary Fran Hazinski. Pediatric Life Support: 2020 International Consensus on Cardiopulmonary Resuscitation and Emergency Cardiovascular Care Science With Treatment Recommendations. *Resuscitation*. 2020 Nov;156:A120–A155

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