Cardiac strangulation following epicardial pacemaker implantation: A rare pediatric complication

Erick M. Carreras,[a,b] Walter J. Duncan, MD, FRCP(C), FACC,[c,d] Ognjenka Djurdjev, BAH, MSc,[e] and Andrew I. M. Campbell, BSc, MD, FRCS(C)[a,b]

Objectives: The aim of our study was 2-fold: to determine the incidence of cardiac strangulation (CS) and to develop a clinical pathway to aid in the diagnosis and prognosis of CS. In <2 years, 2 cases of CS occurred in our institution, which caused much alarm and led to the study's objectives.

Methods: All patients who underwent implantation of an epicardial pacemaker from January 1992 to March 2012 were included. There were no exclusion criteria. Health records were used to locate all subjects and gather all retrospective data. Prospectively, subjects without a chest radiograph from the previous 2 years were approached for imaging.

Results: This study included 86 patients retrospectively, and 84 patients prospectively. There was a 2.3% incidence, and a 1.2% mortality, related to CS. A pattern of posterior looping of the ventricular lead was seen in radiographs of both CS-diagnosed patients. Five variables were significantly associated with an outcome of CS ($P = .0153$).

Conclusions: Our data indicate that the 2 cases of CS were not caused by a lack of follow-up but by a lack of consistent imaging for diagnosis. This conclusion is supported by the 8 cases of CS found in the English-language literature. If the patient is age ≤6 months at the time of implantation, particular attention should be given to the placement of leads and follow-up. (J Thorac Cardiovasc Surg 2015;149:522-7)

See related commentary on pages 528-9.

Cardiac strangulation (CS) is a mechanical complication that can occur with epicardial pacemaker (EP) systems in any individual undergoing somatic growth, by causing the entrapment of the patient's heart or great vessels. CS occurs when the epicardial leads of a pacemaker adhere to the heart in a circumferential manner and over time constrict the encompassed structures. As the patient grows, the lead tightens around the heart, and depending on the location of maximal compression, this may lead to coronary stenosis, valvular insufficiency, or ventricular dysfunction, with the potential of cardiac arrest and death.[1-8] As somatic growth is necessary for CS to occur, this mechanical complication is limited to the pediatric population.

Globally, CS is considered a rare pediatric complication, with only 8 published cases.[1-8] However, there are 2 known cases at our institution, suggesting a much higher local incidence than the number of cases currently reported in the literature. Thus, the primary objective of the current study was to investigate the true incidence of CS, with the hypothesis that, based on the local incidence of CS, this complication has simply been underreported on a global scale as a result of its rarity on an individual institutional basis. The secondary objective was to develop a clinical pathway to aid in the diagnosis and follow-up of patients at risk of CS. As chest radiographs are noninvasive and have a minimal number of associated complications, yet provide some of the most explicit images of the epicardial leads, a standard posteroanterior (PA) and lateral chest radiograph was presumed to be the most practical method of identifying early signs of CS.[9]

METHODS

This study goes back 20 years, for 2 reasons: to cover at least one cohort of patients transitioning from pediatric to adult cardiac care; and because patients are not known to be clear of risk of CS at any specific age. The subject inclusion criteria were being a patient who had an epicardial pacemaker (EP) implantation, from January 1, 1992, to May 1, 2012, at BC Children's Hospital. Health records at this hospital were searched to

From the Division of Pediatric Cardiovascular and Thoracic Surgery,[a] Department of Surgery, BC Children's Hospital, Provincial Health Services Authority, Vancouver, British Columbia, Canada; Division of Cardiovascular Surgery,[b] Department of Surgery, Faculty of Medicine, University of British Columbia, Vancouver, British Columbia, Canada; Division of Cardiology,[c] Department of Pediatrics, BC Children's Hospital, Provincial Health Services Authority, Vancouver, British Columbia, Canada; Division of Cardiology,[d] Department of Pediatrics, Faculty of Medicine, University of British Columbia, Vancouver, British Columbia, Canada; and Department of Measurement & Reporting,[e] Provincial Health Service Authority, Vancouver, British Columbia, Canada.

This work was supported by a grant from Innovations in Acute Care and Technology (iACT), Vancouver, British Columbia, Canada.

Disclosures: Authors have nothing to disclose with regard to commercial support.

Received for publication May 15, 2014; revisions received Oct 17, 2014; accepted for publication Oct 20, 2014; available ahead of print Dec 10, 2014.

Address for reprints: Andrew I. M. Campbell, BSc, MD, FRCS(C), BC Children's Hospital, Provincial Health Services Authority, AB306-4480 Oak St, Vancouver, British Columbia, Canada V6H 3V4 (E-mail: acampbell@cw.bc.ca).

0022-5223/$36.00

http://dx.doi.org/10.1016/j.jtcvs.2014.10.094

Abbreviations and Acronyms
CI = confidence interval
CS = cardiac strangulation
CT = computed tomography
EP = epicardial pacemaker
PA = posteroanterior
SD = standard deviation

locate all patients who met these criteria. Medical records from all possible sources were reviewed to obtain all retrospective data on these patients, including cardiac surgical charts, pediatric cardiology charts, pacemaker interrogation charts, radiological records, and transfer charts. Transfer charts are for those who have been transferred to the Pacific Adult Congenital Heart clinic in St Paul's Hospital. This search process identified 86 patients.

This investigation included both retrospective and prospective data collection. In the retrospective portion, the patients' health records were abstracted for demographics, surgical details, complications, and all available follow-up data. The prospective portion involved reviewing a standard set of PA and lateral chest radiographs for all patients, but only if such a set or the fluoroscopic equivalent had not been obtained within the past 2 years. This process left 48 subjects without a recent chest radiograph or fluoroscopic equivalent. These subjects were invited to participate in the study and provide informed consent for imaging to be obtained. The chest radiographs were then reviewed to determine if potential CS was present or suspected. If so, a low-dose, contrast computed tomography (CT) scan was recommended to the patient's cardiologist or corresponding doctor. In all cases, subjects had imaging results explained to them. The Research Ethics Board of the University of British Columbia for the Children's & Women's Health Centre of British Columbia approved this study (case number H12-01641).

Statistics

Exploratory data analysis was performed using descriptive measures. Categoric variables were expressed in terms of percentages with 95% confidence interval (CI). Continuous variables were expressed as medians with ranges because of their skewed distribution.

The incidence was reported with Clopper-Pearson exact CI. The strength of a statistical association was measured by use of the exact logistic regression with penalized likelihood method, by Firth. To ensure equal exposure, these analyses were restricted to cases that had ≥3 years of follow-up, which left 66 subjects from the total subject pool of 86. The 2-sided P value of <.05 was considered statistically significant. All analyses were performed using SAS software, version 9.3 (SAS Institute Inc, Cary, NC).

RESULTS

A classic pattern for an epicardial lead was recognized to exist in all CS-positive patients, both in the literature and in the local cases being presented. The pattern is denoted by posterior looping of the atrial and ventricular leads, forming a ''heart-shaped'' appearance on PA chest radiography (Figure 1, *A*).

Literature Review

The English-language literature was reviewed for reported cases of CS and published incidence. Key characteristics of reported cases were summarized (Table 1). A total of 100,900 epicardial leads from one manufacturer have been implanted in the United States between October 1981 and August 6, 2012 (Katie Lasch, Medtronic, personal communication, 2013). Assuming all implantations were of the most common type—a 2-lead implantation—and knowing that only 8 cases have been reported in the literature, the published global incidence was estimated to be 0.016%. Two of the 86 identified patients had CS, resulting in a local incidence of 2.33% (95% CI, 0.64%-8.09%), which is statistically significantly larger than 0.01% ($P < .001$). As 1 of the 2 cases is deceased, the resulting CS mortality rate was 1.16% (95% CI, 0.21%-6.30%).

Case Details

Within the study period, there were 2 cases of CS, and 1 case that required further investigation beyond the standard PA and lateral chest radiographs to eliminate CS as a potential risk. Patient 1 presented 3 years after implantation of an EP system for congenital heart block with new-onset

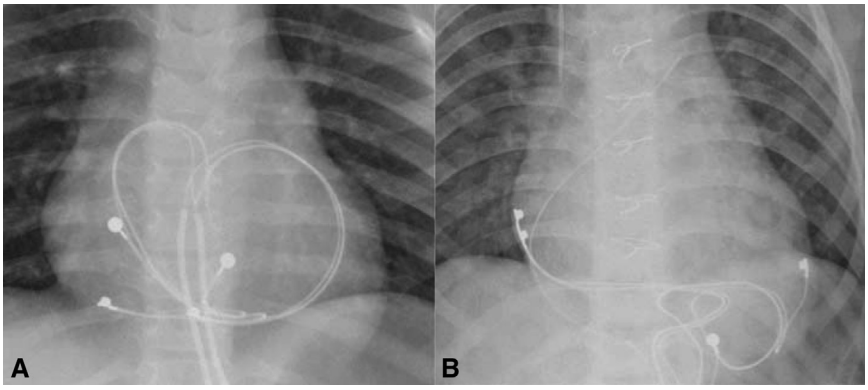

FIGURE 1. A, The last posteroanterior chest radiograph of patient 1, confirming the diagnosis of CS, showing a classic pattern of posterior looping by the atrial and ventricular leads. B, Posteroanterior chest radiograph post–lead replacement, showing patient 1 clear of any possible CS with the leads well situated at the bottom of the pericardium.