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CASE REPORT

Troubleshooting During Temporary Epicardial Lead Implantation in a Child with an Erosive Twiddler's Syndrome and Multiple Sternotomies

A case report

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abstract: Temporary epicardial cardiac pacing in patients with bradyarrhythmias may be used as a bridge to implantation of a permanent pacemaker. The temporary epicardial lead placement may sometimes necessitate a sternotomy that may pose a challenge in patients who have had multiple earlier sternotomies. The difficulty in accessing the epicardium for urgent implantation of temporary epicardial pacing leads depends on the extent of adhesions in such patients. We report an 8-year-8-month-old girl with a pacemaker with an extruded pulse generator and difficult myocardial access due to 5 prior transsternal procedures. The child presented to a tertiary care hospital in Muscat, Oman, in 2021. A trouble-shooting technique was adopted to achieve temporary epicardial pacing to provide time for a course of antibiotic therapy administration. A permanent transvenous pulse generator system was implanted after 7 days of temporary pacing.

Keywords: Heart Block; Artificial Pacemaker; Implanted Electrodes; Case Report; Oman.

In patients with an earlier transsternal cardiac surgery and those in need of temporary epicardial pacing lead implantation, the difficulty in accessing the epicardium may increase depending on the extent of adhesions. This report details the case of an eroded permanent pacemaker in a pacemaker dependent child who had had 5 repeat sternotomies. Temporary epicardial pacing was needed for the interim period until a permanent endocardial pacemaker could be implanted. We describe a technique wherein transsternal temporary epicardial pacing was achieved averting a major invasive operative procedure.

Case Report

An 8-year-8-month-old girl (weight = 19 kg, height = 120 cm, body surface area = 0.8 m^2) after 5 repeat sternotomies presented to a tertiary care hospital in Muscat, Oman, in 2021 for the explantation of an eroded permanent pulse generator that was implanted 8 months previously [Figure 1]. The child had a prior complete repair of an atrioventricular canal defect with left atrioventricular valve repair, left diaphragmatic plication, mitral valve replacement and subsequent sub-aortic membrane resection. At 6-years-5-months of age, a permanent pacemaker was implanted for complete heart block. At the age of 8 years, the child was once again operated for a pannus removal of the prosthetic mitral valve and a new permanent pacemaker was implanted in the right epigastric region. The child had no history of any skin disease. Repeated itch-scratch cycles at the pacemaker insertion site resulted the pulse generator erosion. The chest radiographs after pacemaker insertion at 3 months, 6 months and 8 months (i.e. current admission) displayed changes in the axis of the pacemaker [Figure 2].

During the current admission the child was in complete heart block (40 beats per min). When the output of the pulse generator was increased to 7.5 V from the earlier 5 V, the capture by the pulse generator was regained. This suggested that the bipolar epicardial leads were still functional. The tentative plan of management was to implant temporary epicardial leads through a sternotomy urgently and explant the extruded permanent pulse generator. The blood counts and serum C-reactive protein levels on admission showed no signs of systemic infection (white blood cells = 7 × 10^9/L (normal range: 4.5–14.5], neutrophils = 4.9 × 10^9/L [normal range: 1.4–9], lymphocytes = 1.5 × 10^9/L [normal range: 1.9–9.8], serum C-reactive protein level = 12 mg/L [0.02–14.4]). The child was to receive prophylactic antibiotics and then undergo a permanent endocardial pacemaker implantation.

In the operation room, after attaching external defibrillator pads with pacing capability, general anaesthesia was administered under standard American Society of Anesthesia monitoring guidelines. A 6-Fr sheath was inserted through the right internal jugular vein to provide an access for transvenous

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Figure 1: Photograph of the abdomen on an 8-year-8-month-old girl showing the eroded permanent pulse generator.

pacing leads and right radial artery was accessed for invasive arterial pressure monitoring. An infusion of isoprenaline was kept ready.

A subxiphisternal incision was made and extended to the exposed pacemaker site. The pacemaker box, which was still functioning, was extruded. Although the pulse generator was implanted below the rectus abdominis, the anatomical planes were disrupted due to multiple interventions with the generator site. All debris and necrotic skin was excised and the area washed out with hydrogen peroxide and povidone iodine solution. The subxiphisternal incision was extended superiorly for another 2 cm and dissection was followed along the permanent leads in an attempt to approach the epicardium. The site of attachment of 1 lead was identified and the epicardium around that site was exposed. Then 2 temporary pacemaker leads (TPW30, Ethicon Inc., Raritan, New Jersey, USA) were implanted and an attempt to pace the heart was made. Despite increasing the output voltage of the external pulse generator (Dual Chamber Temporary External Pacemaker, Model 5392, Medtronic, Minneapolis, USA], the heart rhythm could not be captured. This

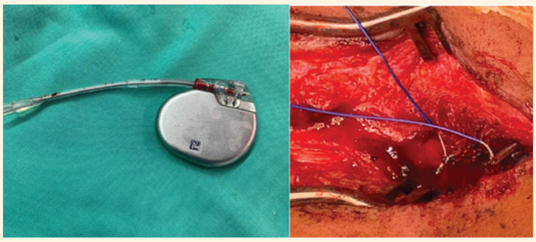

Figure 3: Photographs showing the **(A)** explanted pulse generator and **(B)** connection of the positive and negative leads to the temporary epicardial leads for pacing in single chamber ventricular pacemaker mode.

was probably due to fibrotic tissue on the surface of the heart because of dense adhesions as a result of multiple sternotomies. It was apparent that further dissection of the adherent myocardium needed cardiopulmonary bypass support. Gaining a femoral vascular access for institution of cardiopulmonary bypass was not an option as the earlier cardiac catheterisation reports mentioned multiple collateral pathways of both the femoral vessels.

As a trouble-shooting technique, it was decided that after explantation of the defective pulse generator, the permanent leads would be excised as close to the myocardium as possible. Then the temporary pacemaker leads would be connected to the denuded surface of the permanent leads and the junction of the leads would be insulated with polyvinyl tubing [Figure 3].

During this period when the pulse generator was explanted, the child's underlying heart rate was approximately 40 beats per minute with an arterial mean pressure of 52 mmHg. Once the leads were in place, the heart could be paced at a rate of 100 beats per minute (single chamber ventricular pace maker, 12 V) with a marginal improvement in the mean arterial blood pressure (57 mmHg). The child's trachea was extubated after skin closure. The child was administered intravenous amoxicillin with clavulanic acid (25 mg/kg every 6 hours) for 10 days. The child continued to show no signs of systemic infection as shown by normal blood counts C-reactive protein

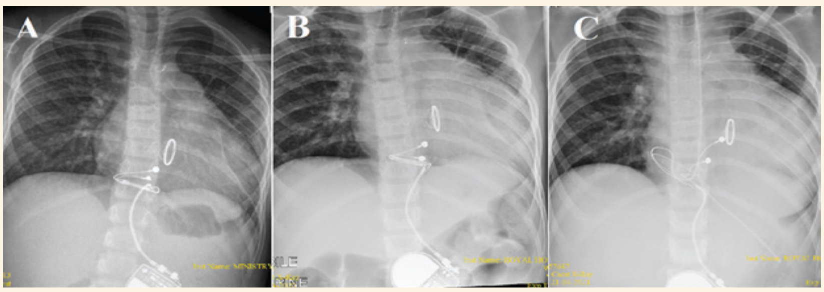

Figure 2: Chest radiograph showing **(A)** the permanent epicardial pacemaker system at 3 months follow-up, rotation of the pacemaker compared to **(B)** the original orientation at 6 months follow-up and coiling of the epicardial leads with displacement of the pacemaker at **(C)** 8 months (i.e. at the time of current admission).

levels. The blood culture and wound cultures showed no bacterial growth. After a week of antibiotic treatment, a single chamber endocardial pacemaker (Medtronic) was placed, and the temporary pacing was discontinued. The epicardial leads were excised flush with the skin after application of traction.

The institutional ethical committee approval (SRC#CR35/2021) as well as an informed written consent from the parents was obtained to publish this report.

Discussion

A technique for achieving temporary epicardial pacing in a patient with Twiddler's syndrome and repeated sternotomies is described.

Twiddler's syndrome is defined as pacemaker malfunction in the setting of device lead dislodgement due to physical manipulation.[1] There are 3 distinct variants of Twiddler's syndrome which have been described.[2–5] An erosive subtype of the Twiddler's syndrome with pacemaker externalisation without lead dislodgement and pacemaker failure was previously reported in an adult patient.[6] In the current patient, the pacemaker was eroded with loss of capture of the heart rate. However, the heart rhythm could be captured with a higher voltage indicating that the permanent epicardial leads were functional but needed a higher voltage for rhythm capture. This was probably due to onset of fibrotic changes at the site of implantation. The authors suggest that this current condition in the child may be considered as an erosive form of Twiddler's syndrome.

An attempt to implant temporary epicardial pacing wires on the exposed myocardium though the limited sternotomy failed to capture the heart probably due to dense fibrotic tissue. Further dissection for better access was deemed hazardous without cardiopulmonary bypass support. Institution of cardiopulmonary bypass support via the femoral vascular access that was subjected to multiple catheterisations was considered a drastic measure just for the implantation of the temporary epicardial leads. A lateral thoracotomy approach was a possibility that was not pursued due to pleural adhesions.[7] The other alternatives to achieve temporary pacing in the current situation was insertion of a Micra leadless pacemaker (Medtronic) or a screw-in lead transvenous pacemaker (Tendril 1888 TC, St Jude Medical, Saint Paul, Minnesota, USA). The option of a leadless pacemaker was not considered as the sheath that was needed with the available system in the institution was too big for the child. A screw-in lead transvenous pacemaker was not available. Hence, attaching the temporary lead cables to the existing epicardial leads was considered a viable option. This approach met the main objective of achieving a temporary heart rate control while simultaneously avoiding major surgical dissections.

A high stimulation threshold was accepted during the epicardial pacing as it was a temporary solution. This was done in order to buy time until a permanent transvenous pacemaker system could be implanted after the administration of a course of antibiotic. The course of antibiotic was administered to overcome any nidus of infection at the site of pulse generator extrusion. Secondly, it was opted to avoid an intravenous lead placement albeit temporary, up until the time of implantation of the transvenous permanent pacemaker system. Hence, despite the stimulation threshold being high, an elective insertion of a temporary transvenous pacemaker system was avoided and was kept as an emergency standby.

The current report had the following shortcomings: (1) in hind sight, it may be suggested that in view of the multiple prior sternotomies, explantation of the permanent pacemaker and clipping the permanent leads as close as possible to the heart followed by the attachment of the temporary pace maker leads to the clipped ends may have been planned; (2) as it was an urgent procedure, neither appropriate radiological evaluation of the relation of cardiac structures to the sternum nor evaluation of the patency of the femoral vasculature was done. This may have been done for a better assessment of the cardiovascular structures and it was missed in the current child; (3) parts of the permanent epicardial leads were left in situ although they were excised after retracing them to the safest possible extent. This was done under the presumption that the child had no suggestion of local or systemic infection.

Conclusion

A simple technique of using the functioning bipolar permanent epicardial leads for temporary epicardial pacing is reported. The technique helped bridge the immediate crisis of explantation of an eroded permanent pulse generator in a child who was pacemaker dependent. The technique enabled the implantation of a permanent endocardial pacemaker after institution of antibiotic therapy.

Authors' Contribution

MMM contributed towards conception of the work; clinical management, acquisition of images and data; drafting of manuscript; and intellectual content. IAA contributed towards clinical management; critical review of the manuscript; and helped in responding to the reviewers' comments. MHP contributed towards clinical management; acquisition of data and images; and help in responding to the reviewers' comments. AMA contributed towards clinical management. SZ contributed towards conception of the work; clinical management; and critical review of the manuscript. All authors approved the final version of the manuscript.

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