An incessant atrial tachycardia originating from epicardial left atrial appendage in a 12-year-old girl: ablation or excision?

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Abstract

Aims The left atrial appendage (LAA) is one of the major sources of atrial tachycardias (ATs) in children. However, endocardial ablation in LAA may fail or even cause fatal tamponade; and epicardial ablation with subsequent surgical appendectomy (SAE) may be required. There is no relevant report in children. We aimed to evaluate the feasibility of epicardial ablation with SAE in children. Methods and results The epicardial ablation with SAE was performed in a 12-year-old girl with an incessant AT. Endocardial mapping demonstrated endocardial activation time of -112 msec and -105 msec (relative to the onset of the A wave at CS9-10) in the right ventricular outflow tract and LAA, respectively. But multiple ablation attempts at these sites did not terminate the AT. After the left-side pericardium opened, the earliest activation (-120 msec) during AT was found at the apex of upper lobe. At this site, ablation eliminated the tachycardia within 5 seconds of onset of energy. After ablation, the LAA was excited, followed by a continuous over and over suture to definitely close the resection line. The free of AT and a decrease in LAD and NT-proBNP were achieved during the 12-month follow-up. Conclusion The epicardial ablation with subsequent SAE was successfully performed in a child. The heart function of the patient improved after a 12-month follow-up. The excision of LAA may be an ideal strategy for children with incessant AT originating from epicardial LAA. However, the long-term safety and efficacy of SAE in children should be further estimated.

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Introduction

Left atrial appendage (LAA) is one of the major sources of atrial arrhythmias in children[1]. But atrial tachycardias (AT) with an epicardial LAA origin are uncommon and usually arise from the base [2]. Catheter ablation(CA) may be an important strategy for patients with atrial arrhythmias [3], especially due to the LAA firing[4]. Due to a large number of trabeculations and true epicardial location, endocardial CA in LAA may fail or even cause fatal tamponade[2, 5]. The epicardial CA may be an alternative approach [6]. Because the LAA has a very thin wall and may be prone to perforation, caution should be taken when LAA ablation is performed[4], even if epicardial ablation. There may be recurrence of AT, and surgical left atrial appendectomy (SAE) may be required.

We report one child with incessant AT that was found to be arising from epicardial LAA, in whom CA was successfully done through epicardial approach with subsequent surgical appendectomy without any complications. We aimed to evaluate the feasibility of epicardial ablation with subsequent SAE in children.

Methods

This study was approved by Ethics Committee of Xinhua Hospital Affiliated to Shanghai Jiao tong University School of Medicine (approval number: XHEC-D-2020-191) and performed in accordance with the Declaration of Helsinki.

Study population

A 12-year-old girl weighing 46 kg and 165 cm in height (with a body surface area of 1.45 m^2) presented with almost incessant, drug-refractory AT was referred for radiofrequency catheter ablation. She had a 5-month history of palpitations, and the electrocardiogram (ECG) showed persistent atrial tachycardia with variable (3-1:1) conduction, and a maximum heart rate of 150 beats/min (Figure 1).

The echocardiogram revealed enlarged left atrium diameter (LAD, 37 mm; normal range $<$ 35 mm). The left ventricular end-diastolic and end-systolic diameters (LVEDD and LVESD) were 48 (normal range $<$ 48 mm) and 30 mm, respectively, with a normal left ventricular ejection fraction (LVEF) of 67% (Table 1). The markers of myocardial injury were normal, and no inflammatory edema of myocardial tissue was found by echocardiography. The ECG was repeated, and the results showed the AT was in the range of 140-180 beats/minute without dynamic ST change. The atrial tachycardia lasted for more than 2 days with

elevated NT-proBNP (2586 pg/ml, normal range < 285 pg/ml), elevated GPT (109 U/L, normal range < 75 U/L), and elevated GOT (43 U/L, normal range < 38 U/L) level.

After ruling out reversibility and other causes, the patient was admitted to the hospital for electrophysiology examination and ablation. Transesophageal echocardiography (TEE) was performed before ablation to verify the absence of a left atrial thrombus and guide LAA resection. Patients had been treated with anticoagulants before ablation.

Electrophysiological Study

As previously described[2], an electrophysiological procedure was performed with standard catheter positions in the coronary sinus, His bundle region and high anterolateral right atrial wall after informed consent was obtained. Following a single transseptal puncture by use of the modified brockenbrough technique and an 8 Fr SL1 transseptal sheath (St. Jude Medical), systemic anticoagulation was made with intravenous heparin (50 u/kg) was administered, with additional 1000 u boluses every hour after the first infusion. An ablation catheter was positioned at the left atrium through a transseptal puncture. Then activation mapping with the Ensite Precision (St Jude Medical, Inc., Minnesota, USA) cardiac mapping system was performed at the left atrium.

Excision of the LAA

As previously described[2, 7], the patient underwent general anesthesia with a double lumen endotracheal tube for selective single-lung ventilation, and then was placed in the supine position with bilateral forearms alongside the body and slightly below the table to expose the axillary regions. After right single-lung ventilation, the procedure was begun on the left side in the fourth intercostal space on the midaxillary line. The left-side pericardium was opened at 2 cm posterior to the phrenic nerve to expose LAA. Then activation mapping and ablation was performed on the epicardial surface of LAA.

After ablation, the LAA atriotomy is closed between two 4–5 mm wide strips of Teflon felt to reinforce the fragile tissue, using a mattress suture followed by a continuous over and over sutures to definitely close the resection line. Then the LAA was excised at the orifice leaving a remnant of 5 mm under gentle traction of both sutures[2].

Results

The repeated ECG showed that the P-wave morphology was negative in leads I and aVL and positive in the inferior leads and lead V_1, suggestive of the origin of left atrial localization[1].

First ablation

The cycle length of AT was 400-500 ms long or so. Activation mapping revealed a distal- to- proximal atrial activation sequence in coronary sinus. Further endocardial mapping demonstrated activation time of -112 msec and -105 msec (relative to the onset of the A wave at $CS_{9\text{-}10}$) in the right ventricular outflow tract (RVOT) and LAA, respectively.

A TactiCath Quartz ablation catheter (St. Jude Medical, St. Paul, MN, USA) was used for ablation. Radiofrequency energy applications at multiple sites with early endocardial activation in RVOT did not terminate the tachycardia. Then the ablation catheter was placed at an early activation site of LAA, and multiple attempts using a temperature of 43°C and power settings of 17 to 25 W at this site did not terminate the AT either.

Second ablation

The patient was referred for surgery. After the left-side pericardium opened, LAA with multiple lobes was seen. Then, mapping was performed on the epicardial surface of LAA, and demonstrated that the earliest activation (-120 msec) during tachycardia was found at the apex of upper lobe. LAA angiography demonstrated that this site was adjacent to the endocardial site of earliest activation (Figure 3). At this epicardial site, radiofrequency application using a FlexAbility irrigated ablation catheter (St Jude Medical, St Paul,

MN, USA) eliminated the tachycardia within 5 seconds of onset of energy (a temperature of 43degC and power settings of 30 W). After adequate power achieved, the AT could not be induced.

Excision of the LAA

After ablation, retraction sutures were used to facilitate the exposure. The LAA excision was performed. From macroscoping findings, the middle sites of LAA epicardial surface had lesions from catheter ablation (Figure 3). The surgical procedure took 90 minutes. The patient has remained free of symptomatic arrhythmias during 12-month follow-up.

Assessment of heart function

After the procedure, the echocardiogram showed that LAD, LVEDD, and LVESD decreased to 29, 48, and 32mm, respectively, and the LVEF increased to 61%. The New York Heart Association functional class was II before the procedure, which improved to I after the procedure (Table1).The blood chemistry data were improved with normal NT-ProBNP (133pg/ml), GPT (47 U/L), and GOT (28 U/L) level.

Discussion

In the current case, AT originated at the apex of upper lobe on LAA. The AT could not be terminated by endocardial CA but eliminated by epicardial CA. To the best of our knowledge, this is the first report on one child illustrating an AT with an epicardial LAA origin that was successfully ablated through epicardial approach with subsequent surgical appendectomy. The termination of AT and a decrease in LAD and NT-proBNP were achieved during the 12-month follow-up. On the whole, the preliminary results were inspiring.

Characteristics of LAA in children

The LAA is the remnant of the original embryonic left atrium that develops during the third week of gestation[8], so it is a structure anatomically appended to the main body of the LA, which locate between the left upper pulmonary vein and the left ventricle. The LAA has a wide perimeter that interfaces with the atrial musculature[4]. In the LAA, thick cords of pectinate muscle intervene with thin-walled tissue[8]. The anisotropic junctional tissue, with a complex fiber orientation, results in electrophysiological properties that may predispose this region to be the source of the arrhythmia[3].

In children, left atrium in size is small, and increase as ages. The LAA lies within the confines of the pericardium, and thus its emptying and filling may be affected by left ventricular function.

Indications in children

Currently, no relevant guideline exists for the epicardial LAA AT treatment in children. Although, endocardial CA was considered to be the best strategy for the child in the present study if AT could be terminated without any complications. The complex anatomy of the left atrial appendage makes it a difficult intracardiac structure in which to manipulate catheters safely[9]. In one report, firm forward pressure was exerted on the ablating catheter, causing the appendage to straighten[9]. However, this maneuver could result in LAA perforation[10] and thus was not attempted in the reported case.

Thus, the epicardial CA may be an alternative approach to endocardial CA. CA using an epicardial access by a pericardial puncture may be necessary and is a proven feasible route for the management of a variety of arrhythmias[6]. But the response to radiofrequency applications in epicardial areas differs markedly from the response in endocardial areas because incomplete ablation might result from a lack of contact with the ablation electrode and the thermal homeostatic effect of cavitary saline infusion on the LAA muscles[11]. And the limited space of pericardial cavity makes it still difficult to manipulate catheter safely.

Consideration of surgical appendectomy

The LAA is one of the major sources of ATs in children[1]. The mechanism of AT originating in the LAA is unknown. Intracellular recordings demonstrated that the mechanism was abnormal automaticity[2]. Histology showed that spontaneous activity arose in an area with abnormal cells[2]. However, CA in an atrial appendage

may fail or there may be recurrence of AT; and surgical appendectomy may be required[2, 5]. Because the LAA has a very thin wall and may be prone to perforation, caution should be taken when LAA ablation is performed[4], even if epicardial ablation.

In the current case, because of its favorable risk–benefit ratio rather than a second attempt at catheter ablation, we chose epicardial catheter ablation with subsequent SAE. The patient's heart function improved with significantly decreased LVEDD, LAD and enhanced NYHF.

Conclusions

The epicardial ablation with subsequent surgical appendectomy was successfully performed in the child. The heart function of the patient improved after a 12-month follow-up. The excision of LAA may be an ideal strategy for children with incessant AT originating from epicardial left atrial appendage. However, the long-term safety and efficacy of SAE in children should be further estimated.

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Conflict of interest

None declared

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Figure legends

Figure 1: 12 lead ECG of baseline tachycardia at a rate of 150 bmp

From ECG, the P-wave morphology showed a negative P-wave in leads I and aVL and a positive P-wave in the inferior leads and lead V_1.

Figure 2: Intracardiac electrograms (a), electroanatomic maps (b) and angiograms (c) of endocardial surface of the LAA

(a) Activation at the apex of the LAA is the earliest. (b)The earliest activation at the apex of the LAA was 112 ms ahead of $CS_{9\text{-}10}$. (c) The tip of ablation catheter was at the apex of LAA. The Lasso catheter was placed at base of LAA.

ABL, ablation catheter; CS, coronary sinus; PV, decapolar circular mapping Lasso catheter

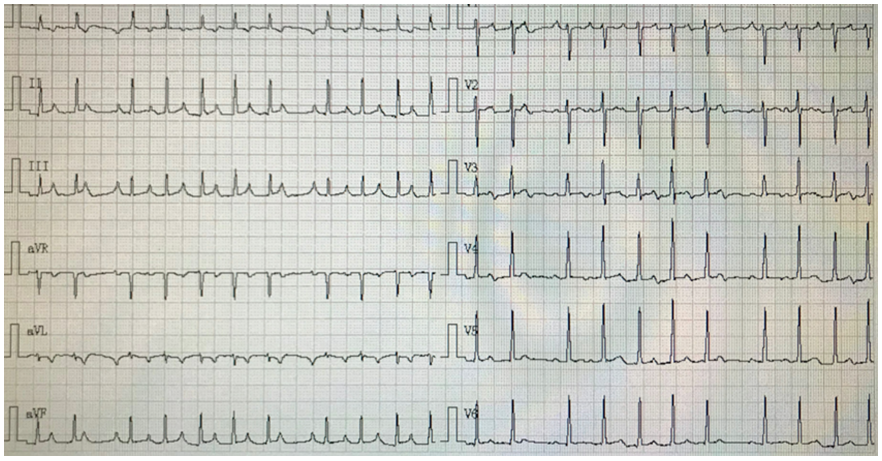
Figure 3: Electroanatomic maps (a) of epicardial surface of the LAA, surgical approach (b), a macroscoping findings(c) and angiograms of LAA (d)

(a) The earliest activation at the epicardial apex of the LAA was 120 ms ahead of $CS_{9\text{-}10}$. The maroon tags represent radiofrequency applications at the earliest activation sites which terminated the AT (white arrow). (c) The middle sites of LAA epicardial surface had lesions from catheter ablation (black arrow).

ABL, ablation catheter; CS, coronary sinus; PV, decapolar circular mapping Lasso catheter

Table 1: Echocardiography parameters before and after the procedure

LAD, left atrium diameter; LVEDD, left ventricular end-diastolic diameter; LVESD, left ventricular end-systolic diameter; LVEF, left ventricular ejection fraction; NYHA, New York Heart Association.



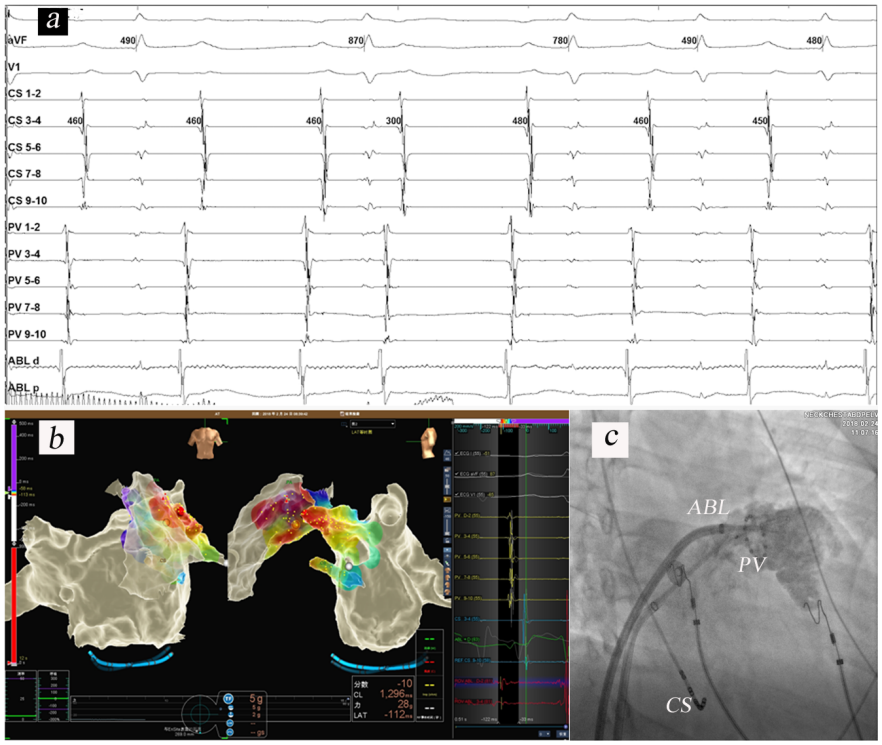


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Table 1.docx available at https://authorea.com/users/380844/articles/562635-an-incessant-atrial-tachycardia-originating-from-epicardial-left-atrial-appendage-in-a-12-year-old-girl-ablation-or-excision