

Cardiogenic anorexia and underweighting in a child: case report

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Introduction

In this case, we present a child with a diagnosis of anorexia and no known cardiac disease, who was referred to our clinic for a routine cardiac monitoring. ‘Asymptomatic’ high degree of atrioventricular block (AVB) was revealed. She underwent successful pacemaker (PM) implantation and losing of the weight stopped.

Case presentation

In this case, the abrupt AVB with more than 6 s ventricular pauses without escape rhythm during the day-time most possible is a type of neurally mediated AVB which was presented by anorexia, underweighting, and refusing to eat in a 12-year-old girl. All symptoms disappeared after PM implantation, and weight gain was recorded during 6 months of follow-up.

Discussion

The causal link between AVB and anorexia is discussed. In this case, patient’s eating disorder was related to her AV conduction abnormality. The main reason of this case report is to emphasize, that in children with unexplained anorexia and underweighting cardiogenic origin must be excluded.

Keywords

Paroxysmal atrioventricular block • Anorexia • Child • Ventricular pause • Case report

Learning points

- A type of neurally mediated atrioventricular block (AVB) which may be presented by anorexia, underweighting and refusing to eat in a child. All symptoms disappeared after pacemaker (PM) implantation and weight gain was recorded during 6 months of follow-up. This case showed, patient’s eating disorder may be related to AV conduction abnormality.
- Change in eating behaviour, refusing to eat, anorexia, and underweighting may clinical presentation of AVB. So, AVB with this type of presentation should be considered as symptomatic, with absolute indication for pacing.
- The main reason of this case report is to emphasize, that in children with unexplained anorexia and underweighting cardiogenic origin must be excluded.

Case presentation

A 12-year-old girl was referred to our clinic with diagnosis of anorexia. She refused to eat during the last months and was obviously underweight (weight 31 kg, height 145 cm, body mass index (BMI)=14.7 kg/cm²). She had no palpitation, syncope or any other

cardiac complains. On admission, laboratory investigations revealed a normal complete blood count, electrolytes, sedimentation rate, liver and kidney function tests, amylase, cholesterol and thyroid-stimulating hormone. The follicle-stimulating hormone, luteinizing hormone and oestradiol were in normal levels for this age. To find the cause of underweighting, gastro-duodeno-oesophagoscopy and

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brain computer tomography (CT)-scan were performed. No abnormalities were found. Malabsorption syndrome characterized by anorexia, weight loss, and steatorrhoea was excluded. Anorexia nervosa was diagnosed by psychologist in an initial report, but it was called atypical, because does not clearly met common types of this pathology: restrictive or self-induced vomiting were not recorded. Commonly, restricting food intake may include excessive exercise, which was not reported in this case.

Twelve-lead resting electrocardiography revealed sinus rhythm with slight prolongation of PR interval (220 ms) (Figure 1). The echocardiography revealed normal structure of the heart. The 40-h ambulatory electrocardiography (ECG) monitoring showed more than 70 episodes of 2:1 AV block and high degree (up to 7:1) of AV block with no escape rhythm [ventricular pauses up to 6 s (Figure 2)]. Interestingly, all these episodes were recorded during the day-time and did not associate with syncope or dizziness. No AV nodal conduction abnormality was recorded during sleeping. Dual chamber pacemaker (PM) was successfully implanted after patient informed consent. 24-h ambulatory ECG monitoring showed ventricular pacing during day-time (Figure 3). The patient was under the follow-up during 6 months. According to PM follow-up cumulative percent of ventricular pacing was 6%. She started to eat normally just after implantation, and 5 kg weight gain was recorded after month. She was completely active, did well in math school, no any kind of physical or laboratory disorder were revealed, no eating disorder recorded during 6 months. She has been under the control in paediatric and cardiac clinics, no any structural changes reported at 6th month, weight 42 kg, height 148 cm, BMI = 19.2 kg/cm², which was categorized as normal weight. Parents' consent form was presented.

Timeline

Dates	Relevant past medical history and interventions
May 2017	A 12-year-old girl was referred to our clinic with diagnosis of anorexia. She refused to eat during the last months and was obviously underweight (weight 31 kg, height 145 cm, body mass index (BMI) = 14.7 kg/cm ²). She had no palpitation, syncope, or any other cardiac complains. To find the cause of underweighting gastro-duodeno-oesophagoscopy, brain computer tomography (CT)-scan, extended laboratory analyses were performed. No abnormalities were found. Anorexia nervosa was diagnosed by psychologist. Dates summaries from initial and follow-up visits
June 2017	Twelve-lead resting electrocardiography (ECG) revealed sinus rhythm with slight prolongation of PR interval (220 ms) (Figure 1). The echocardiography revealed normal structure of the heart. The 40-h ambulatory ECG monitoring showed more than 70 episodes of 2:1 AV block and high degree (up to 7:1) of AV block with no escape rhythm [ventricular pauses up to 6 s (Figure 2)]. Interestingly, all these episodes were recorded during the day-time and did not associate with syncope or dizziness. No AV nodal conduction abnormality was recorded during sleeping.
June 2017	Dual chamber pacemaker (PM) was successfully implanted after patient informed consent.
July 2017	She started to eat normally just after implantation and 5 kg weight gain was recorded after month.
Sept 2017	ECG, EchoCG, PM-follow-up data were in normal range, AsVp was up to 16%. Gastro-duodeno-oesophagoscopy and extended laboratory analyses were performed. No abnormalities were found. Psychologist confirmed no anorexia.
Nov 2017	She was completely active, did well in maths school, no any kind of physical or laboratory disorder were revealed, no eating disorder recorded. She has been under the control in paediatric and cardiac clinics, no any structural changes reported at 6th month, weight 42 kg, height 148 cm, BMI = 19.2 kg/cm ² , which was categorized as normal weight.

Discussion

The main clinical entity with abrupt complete heart block are:

- *Paroxysmal atrioventricular block (AVB)* is abrupt loss of AV conduction, which usually may lead to syncope. The main electrophysiological mechanism of paroxysmal AVB is a local phase 3 or 4 block in the His-Purkinje system. During a long pause (after premature atrial or ventricular beat), the fibres of the His-Purkinje system spontaneously depolarize and subsequent impulses are blocked due to sodium channel inactivation.¹ So, paroxysmal AVB is often initiated by atrial, His, or ventricular premature extrasystole that initiates the pause. Also tachycardia can initiate paroxysmal AVB via suppression of AV conduction.² Usually sinus acceleration has been observed during ventricular asystole. Paroxysmal AVB is a type of infranodal AVB when block is commonly unidirectional (anterograde) with preservation of retrograde conduction. That is why a ventricular premature beat is able to resume 1:1 AV conduction.³ In this case, no premature beats at the beginning or at the end of ventricular pause, that is why this cannot be called paroxysmal AVB. *Parasympathetic or vagal effect on the conduction system* includes gradual slowing of the sinus rate (P-P lengthening) and AV conduction (prolonging PR), which are occasionally followed by sinus arrest or complete AVB. This is type of nodal AVB and commonly bidirectional, usually met in rest and sleeping. Findings highly suggestive of vagal AVB is prolonging P-P interval during ventricular asystole. Vagally mediated AVB may be presented by Mobitz Type I, pseudo-Mobitz Type II also called apparent Type II block, 2:1, advanced-degree, complete AV block or a combination of different types of AV block.^{4,5} In this case, no gradual slowing of sinus rate, which is commonly present in vagally mediated AVB. Also AVB was recorded during day-time only.
- *Situational syncope AVB* (advanced-degree or complete) in patients with, such as swallowing or coughing syncope. Although vasovagal

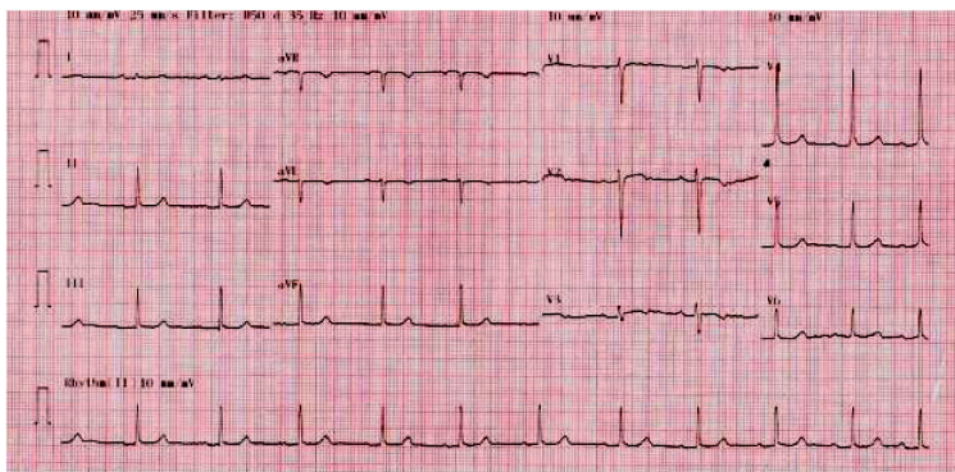


Figure 1 Twelve-lead resting electrocardiography revealed sinus rhythm with slight prolongation of PR interval (220 ms).

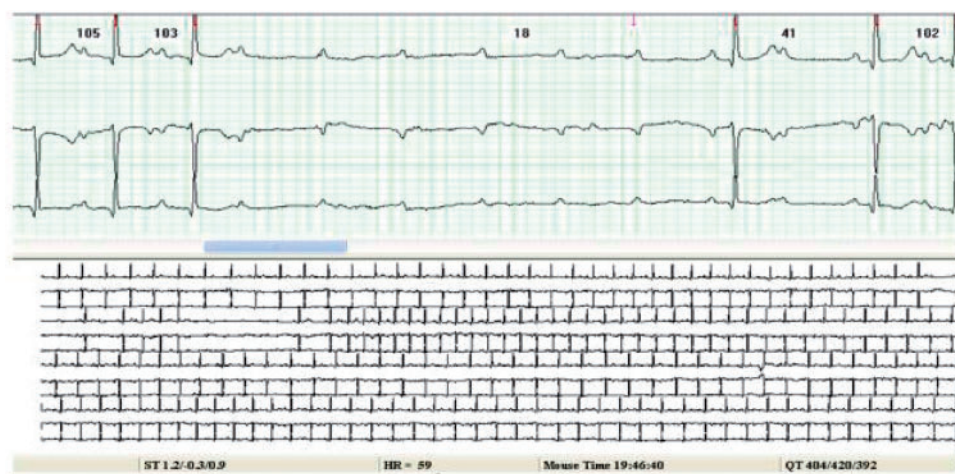


Figure 2 The strip from the 40-h ambulatory electrocardiography monitoring showed an episode of high degree (7:1) of atrioventricular block with no escape rhythm (ventricular pause is up to 6 s).

syncope occurs very frequently, swallow or deglutition syncope is an unusual condition whereby temporary loss of consciousness occurs with swallowing.⁶ These patients had normal heart, normal AV conduction and positive tilt testing. These syncopal episodes were diagnosed as *neurally mediated*. Physiologically, swallow syncope is produced by a vagovagal reflex. W. Omi and S. Mitra reviewed 63 and 17 reported cases of swallow syncope in children and adults. According to these reviews, swallow syncope has been occurred in patients with organic or functional disorders of the oesophagus (oesophageal spasm, stricture, achalasia, diverticula, cancer, hiatal hernia) or in the setting of cardiac diseases (acute myocardial infarction, rheumatic carditis, digoxin use, ascending aortic aneurysm, thoracic surgery). However, in a significant number of cases, no underlying disease was found. Pacemaker implantation was the common treatment method in most of the cases.^{7,8}

In our case, there is no any concomitant cardiac or oesophageal disease or situation pre-disposing to AVB.

- Complete AVB secondary to anatomical involvement of AV conduction or intrinsic AVB is historically known as no conduction through the AV node with complete dissociation of the atrial and ventricular activity.⁹ The ventricular escape mechanism can occur anywhere from the AV node to the Purkinje system.¹⁰ In this case, an AVB does not have a typical behaviour for complete AVB secondary to anatomical disease of AV node, as no escape rhythm was revealed, no any combination of different types of AV block, also it is unspecific to see intrinsic AVB only in day-time.

Anorexia nervosa is an eating disorder characterized by extremely low body weight, fear of gaining weight or distorted perception of body image, and amenorrhoea.¹¹ National Institute For Health and



Figure 3 The strip from the 24-h ambulatory electrocardiography monitoring after pacemaker implantation showed an episode of ventricular pacing in day-time.

Cardiac Excellence (NICE) guideline on eating disorders recommends referring people for assessment in case of unusual low BMI for age, rapid weight loss, family members report a change in eating behaviour. Physical signs and symptoms of anorexia may include: abnormal blood counts, cessation of menstruation after it has been established, irregular heart rhythms, low blood pressure, osteoporosis, etc. Diagnosis of anorexia mostly based on behavioural symptoms including attempts to lose weight by either: severely restricting food intake and may include excessive exercise or bingeing and self-induced vomiting to get rid of the food. Diagnosis may be more problematic in children and younger adolescents, as the existing diagnostic criteria are insufficiently sensitive.¹² Causes of anorexia nervosa are biological (genetic), psychological and environmental, but not structural changes. In children and adolescents with atypical presentations of an eating disorder, consideration should be given to the possibility of separate underlying physical pathology. In general, medical complications of anorexia nervosa include atrophy of the heart, brain, liver, intestines, kidneys, and muscles. According to published studies the most common cardiac findings in adolescents with anorexia nervosa are bradycardia and hypotension, a prolonged QTc interval.¹³ Electrolyte loss, interstitial oedema, decreased glycogen content in cardiac cells, myofibrillar atrophy, mitochondrial inflammation, and calcium-dependent proteinase activation are known mechanism of such findings.¹⁴ Structural abnormalities, including pericardial effusion and decreased left ventricular size are also common complications of anorexia nervosa. There is a case report presented a 12-year-old girl with anorexia (restrictive subtype) with a second-degree AVB, Mobitz Type I. This case report highlighted that although second-degree AVB (Mobitz Type I) may be an intrinsic conduction abnormality or caused by increased vagal activity as a consequence of the malnutrition secondary to anorexia.¹⁵ Another case report by Bravender *et al.* presented a 20-year-old woman with the second-degree AVB and anorexia. In this case, an AVB was thought to represent an intrinsic conduction abnormality unrelated to anorexia as it was present at low weight and after weight restoration.¹⁶ To overcome anorexia nervosa not too easy, and psychologist's usually work with patient and family. In this case, no any complications were reported, psychologist was failed to help working with patient and family. Another cause of substantial weight loss is malabsorption. Malabsorption syndrome is a complex of symptoms resulting

impaired absorption of nutrients from the gastrointestinal tract. Usually these patients suffering with chronic diarrhoea, nutritional deficiencies, and weight loss despite a healthy diet. Diagnosis of malabsorption is based on laboratory, radiographic tests, and endoscopy of gastrointestinal tract. Anaemia, weakness, and fatigue occur because iron, folic acid, and vitamin B12 are not absorbed in sufficient amounts. In this particular case no any abnormality was found to assist with malabsorption diagnosis.

This is the only report we could find in the literature with this type of presentation. Neither AVB presented by anorexia nor anorexia complicated by complete AVB did not reported before.

Summarizing, anorexia nervosa is the result of abrupt loss of AV conduction, but not the cause of AVB in this case. In the fact, a child has eating disorder which was eliminated by pacing. Most possible, this is some kind of neurally mediated AVB (involvement of vasovagal reflex cannot be excluded), and uncommonness is that this block was not clinically presented by syncope or dizziness. According to current guidelines only symptomatic bradycardia is an indication for pacemaker implantation, provided other causes of symptoms have been excluded.¹⁷ Alternative causes (apnoea, seizures, medication effects, and neurocardiogenic mechanisms) were excluded in our patient. In this case, AVB is associated with minimal physical activity including eating, as it is recorded only during day-time. Most likely AVB had been presented during the long period, and a child started to avoid all kinds of activities including eating. She could not explain own complains like adults, as she even did not know, that life can be otherwise. Fear of sudden death was stronger than instinct to eat. Change in eating behaviour, refusing to eat, anorexia, and underweighting were clinical manifestations of AVB in this particular case. So, this was symptomatic AVB, with absolute indication for pacing.

Conclusion

In this case, the abrupt AVB with more than 6 second ventricular pauses without escape rhythm during the day-time most possible is a type of neurally mediated AVB which was presented by anorexia, underweighting, and refusing to eat in a 12-year-old girl. All symptoms disappeared after PM implantation, and weight gain was recorded during 6 months of follow-up. In this case, patient's eating disorder was related to her AV conduction abnormality. The main

reason of this case report is to emphasize, that in children with unexplained anorexia and underweighting cardiogenic origin must be excluded.

Consent: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: none declared.

References

- Motte G, Desoutter P, Olive B, Bodereau P, Welti JJ. Paroxysmal block in phase 4 of the bundle of His. *Arch Mal Coeur Vaiss* 1977;**70**:797–807.
- Josephson ME, ed. *Clinical Cardiac Electrophysiology*, 4th edn. Philadelphia: Lippincott Williams & Wilkins; 2008. p110.
- Lee S, Wellens HJ, Josephson ME. Paroxysmal atrioventricular block. *Heart Rhythm* 2009;**6**:1229–1234.
- Massie B, Scheinman MN, Peters R, Desai J, Hirschfeld D, O'Young J. Clinical and electrophysiologic findings in patients with paroxysmal slowing of the sinus rate and apparent Mobitz type II atrioventricular block. *Circulation* 1978;**58**:305–314.
- Alboni P, Holz A, Brignole M. Vagally mediated atrioventricular block: pathophysiology and diagnosis. *Heart* 2013;**99**:904–908.
- Kakuchi H, Sato N, Kawamura Y. Swallow syncope associated with complete atrioventricular block and vasovagal syncope. *Heart* 2000;**83**:702–704.
- Omi W, Murata Y, Yaegashi T, Inomata J, Fujioka M, Muramoto S. Swallow syncope, a case report and review of the literature. *Cardiology* 2006;**105**:75–79.
- Mitra S, Ludka T, Rezkalla S, Sharma P, Luo J. Swallow syncope: a case report and review of the literature. *Clin Med Res* 2011;**9**:125–129.
- Narula OS, Scherlag BJ, Javier RP, Hildner FJ, Samet P. Analysis of the A-V conduction defect in complete heart block utilizing His bundle electrograms. *Circulation* 1970;**41**:437–448.
- Rosen KM, Dhingra RC, Loeb HS, Rahimtoola SH. Chronic heart block in adults. *Arch Intern Med* 1973;**131**:663–672.
- Klein D, Attia E. Anorexia nervosa in adults: diagnosis, associated clinical features, and assessment. In: DS Basow (ed.). *UpToDate*. Waltham, MA: UpToDate; 2013.
- Bryant-Waugh R. Overview of eating disorders. In: B Lask, R Bryant-Waugh (eds). *Anorexia Nervosa and Related Eating Disorders in Childhood and Adolescence*. Hove: Psychology Press; 2000. pp. 27–40.
- Nudel DB, Gootman N, Nussbaum MP, Shenker IR. Altered exercise performance and abnormal sympathetic responses to exercise in patients with anorexia nervosa. *J Pediatr* 1984;**105**:34–37.
- Tolnai S, von Althen I. Calcium-dependent proteolysis in the myocardium of rats subjected to stress. *Life Sci* 1987;**41**:1117–1122.
- Kanbur N, Goldberg E, Pinhas L, Hamilton R, Clegg R, Katzman D. Second-degree atrioventricular block (Mobitz Type I) in an adolescent with anorexia nervosa: intrinsic or acquired conduction abnormality. *Int J Eat Disord* 2009;**42**: 575–578.
- Bravender T, Kanter R, Zucker N. Anorexia nervosa and second-degree atrioventricular block (type I). *Int J Eat Disord* 2006;**39**:612–615.
- Brignole M, Auricchio A, Baron-Esquivias G, Bordachar P, Boriani G, Breithardt O, et al. 2013 ESC Guidelines on cardiac pacing and cardiac resynchronization therapy. The Task Force on cardiac pacing and resynchronization therapy of the European Society of Cardiology (ESC). Developed in collaboration with the European Heart Rhythm Association (EHRA). *Eur Heart J* 2013;**34**: 2281–2329.