World Journal of *Clinical Cases*

World J Clin Cases 2024 March 6; 12(7): 1196-1381





Published by Baishideng Publishing Group Inc

W J C C World Journal of Clinical Cases

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ABOUT COVER

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AIMS AND SCOPE

The primary aim of World Journal of Clinical Cases (WJCC, World J Clin Cases) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: Si Zhao; Production Department Director: Xiang Li; Editorial Office Director: Jin-Lei Wang,

NAME OF JOURNAL World Journal of Clinical Cases	INSTRUCTIONS TO AUTHORS https://www.wjgnet.com/bpg/gerinfo/204
ISSN	GUIDELINES FOR ETHICS DOCUMENTS
ISSN 2307-8960 (online)	https://www.wjgnet.com/bpg/GerInfo/287
LAUNCH DATE	GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH
April 16, 2013	https://www.wjgnet.com/bpg/gerinfo/240
FREQUENCY	PUBLICATION ETHICS
Thrice Monthly	https://www.wjgnet.com/bpg/GerInfo/288
EDITORS-IN-CHIEF	PUBLICATION MISCONDUCT
Bao-Gan Peng, Salim Surani, Jerzy Tadeusz Chudek, George Kontogeorgos, Maurizio Serati	https://www.wjgnet.com/bpg/gerinfo/208
EDITORIAL BOARD MEMBERS	ARTICLE PROCESSING CHARGE
https://www.wjgnet.com/2307-8960/editorialboard.htm	https://www.wjgnet.com/bpg/gerinfo/242
PUBLICATION DATE	STEPS FOR SUBMITTING MANUSCRIPTS
March 6, 2024	https://www.wjgnet.com/bpg/GerInfo/239
COPYRIGHT	ONLINE SUBMISSION
© 2024 Baishideng Publishing Group Inc	https://www.f6publishing.com

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World J Clin Cases 2024 March 6; 12(7): 1313-1319

DOI: 10.12998/wjcc.v12.i7.1313

ISSN 2307-8960 (online)

CASE REPORT

Reversal of complete atrioventricular block in dialysis patients following parathyroidectomy: A case report

Shan-Shan Xu, Li-Hai Hao, Yan-Meng Guan

Specialty type: Cardiac and cardiovascular systems

Provenance and peer review: Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0 Grade B (Very good): 0 Grade C (Good): C Grade D (Fair): 0 Grade E (Poor): 0

P-Reviewer: Juneja D, India

Received: October 19, 2023 Peer-review started: October 19, 2023

First decision: January 5, 2024 Revised: January 18, 2024 Accepted: February 4, 2024 Article in press: February 4, 2024 Published online: March 6, 2024



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Abstract

BACKGROUND

Refractory secondary hyperparathyroidism (SHPT) is a common complication observed in patients with end-stage renal disease and can result in ectopic calcification. Metastatic calcification involving the heart valves and the conduction system can easily lead to arrhythmias, including atrioventricular block. This case report describes a maintenance hemodialysis patient with refractory SHPT resulting in a complete atrioventricular block (CAVB), which was eventually reversed to a first-degree atrioventricular block.

CASE SUMMARY

We present the case of a 31-year-old Asian female who was receiving maintenance hemodialysis because of lupus nephropathy. She developed SHPT, and an electrocardiogram revealed a first-degree atrioventricular block. Then, she underwent parathyroidectomy (PTX) with autotransplantation. Unfortunately, a few years later, she developed SHPT again, and an electrocardiogram revealed a CAVB. A few years after the second PTX surgery, the calcification of the left atrium and left ventricle improved, and her CAVB was reversed.

CONCLUSION

This case revealed that metastatic cardiac calcification can result in complete atrioventricular blockage. Following parathyroid surgery, calcification of the cardiac conduction system improved, leading to reversal of the atrioventricular block. It is important for dialysis patients to optimize intact parathyroid hormone therapy and pay attention to calcification metastasis.

Key Words: Secondary hyperparathyroidism; Ectopic calcification; Atrioventricular block; Reversal; Case report

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Core Tip: Refractory secondary hyperparathyroidism (SHPT) is known to cause bone pain, itching, and muscle weakness and can lead to an increased incidence of pathological fractures, metastatic calcification, and cardiovascular events. Metastatic calcification involving the heart valves and the conduction system, such as the mitral valve ring, can easily lead to arrhythmias, including atrioventricular block. The current case report describes a maintenance hemodialysis patient with refractory SHPT resulting in a complete atrioventricular block, which was reversed to a first-degree atrioventricular block after parathyroidectomy. The importance of optimizing parathyroid hormone management is further highlighted by comparing the pre- and postreversal indices.

Citation: Xu SS, Hao LH, Guan YM. Reversal of complete atrioventricular block in dialysis patients following parathyroidectomy: A case report. World J Clin Cases 2024; 12(7): 1313-1319 URL: https://www.wjgnet.com/2307-8960/full/v12/i7/1313.htm DOI: https://dx.doi.org/10.12998/wjcc.v12.i7.1313

INTRODUCTION

Secondary hyperparathyroidism (SHPT) is a common complication observed in patients with end-stage renal disease and can result in ectopic calcification, significantly affecting quality of life and survival. Refractory SHPT is known to cause bone pain, itching, and muscle weakness and is associated with an increased incidence of pathological fractures[1], metastatic calcification, and cardiovascular events, all of which are significantly associated with increased mortality^[2]. Metastatic calcification can involve the heart valves or the atrium, and calcification of the conduction system at the mitral valve ring can easily lead to arrhythmias, including atrioventricular block. An increase in intact parathyroid hormone (iPTH) levels is associated with an increase in metastatic calcification and conduction block, underscoring the importance of timely management of hyperparathyroidism. This case report describes a maintenance hemodialysis patient with refractory SHPT resulting in a complete atrioventricular block (CAVB), which was eventually reversed to a first-degree atrioventricular block.

CASE PRESENTATION

Chief complaints

A 31-year-old female patient had been undergoing maintenance hemodialysis since 2009 and had bone pain and chest tightness for one year.

History of present illness

The patient had been undergoing maintenance hemodialysis at the department since 2009. In 2014, the patient developed SHPT with an iPTH level of 1400 pg/mL, for which calcitriol pulse therapy was applied. Over the next 5 years, the patient's iPTH levels increased progressively, leading to bone pain, skin itching, and difficult-to-correct hypercalcemia and hyperphosphatemia. In June 2018, a parathyroid B-ultrasound revealed parathyroid hyperplasia, and an electrocardiogram revealed a first-degree atrioventricular block (Figure 1). Total parathyroidectomy (PTX) with autotransplantation was performed, with four parathyroid glands completely removed during surgery and one resected parathyroid gland intramuscularly transplanted to the patient's right forearm. The iPTH levels decreased rapidly and were maintained at 40-80 ng/mL one month postsurgery and at 45-70 pg/mL on the side of the transplanted arm 3 months postsurgery. The patient's bone pain and skin itching were gradually relieved. However, 1.5 years later, the patient's iPTH level had risen again to 360-724 pg/mL, and the patient developed a serum calcium level of 2.37-2.86 mmol/L and a serum phosphorus level of 2.00-2.63 mmol/L. The patient was treated with intermittent use of paricalcitol and cinacalcet. The patient presented with bone pain, chest tightness and fatigue beginning in June 2020, and electrocardiography (ECG) revealed a third-degree or CAVB (Figure 2). The patient refused pacemaker therapy.

History of past illness

The patient had a medical history of systemic lupus erythematosus and lupus nephropathy. She had undergone regular hemodialysis for 12 years.

Personal and family history

The patient denied any family history of malignant tumors or other genetic conditions.

Physical examination

The vital signs of the patient were as follows: body temperature, 36.5 °C; heart rate, 78/min; respiratory rate, 18/min; and blood pressure, 100/65 mmHg.



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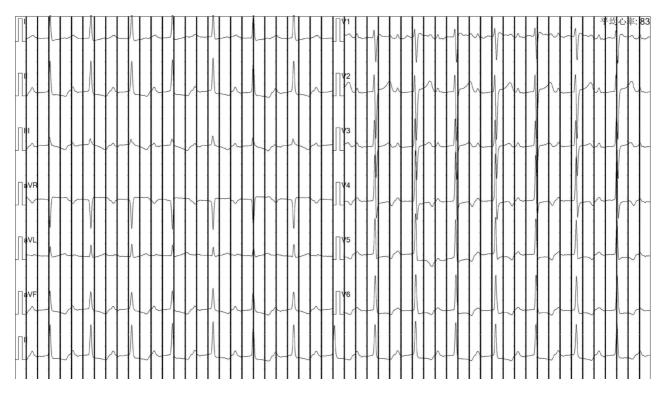


Figure 1 Her electrocardiograph from 2018 showed a first-degree atrioventricular block. The PR-interval was 310 ms.

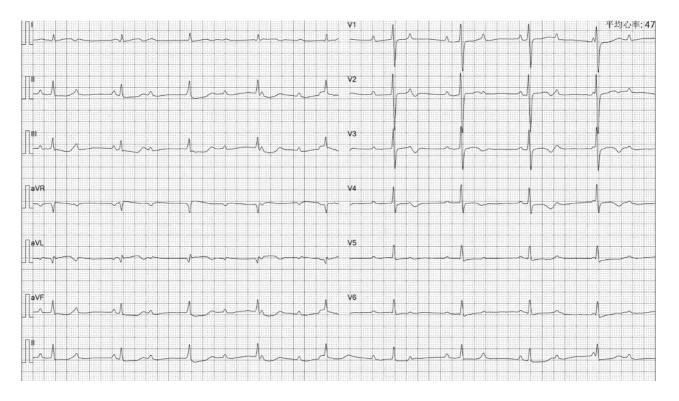


Figure 2 Her electrocardiograph in 2020 showed a complete atrioventricular block.

Laboratory examinations

Her iPTH was > 5000 µmol/L, her serum calcium was 3.08 mmol/L, and her serum phosphorus was 3.47 mmol/L.

Imaging examinations

ECG revealed a third-degree or CAVB. Parathyroid nuclear scintigraphy (ECT)/CT parathyroid scintigraphy revealed a parathyroid adenoma. Color Doppler echocardiography showed calcification in the posterior leaflet of the mitral valve (Figure 3A). Chest CT showed extensive calcifications of the left atrium and left ventricle (Figure 4B and D).

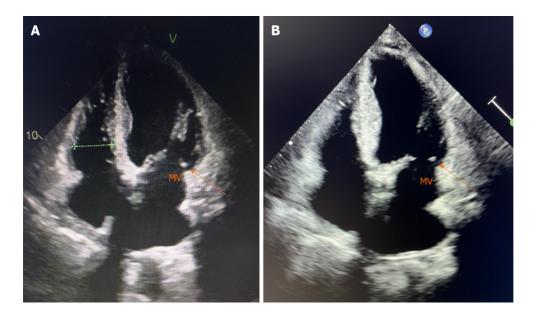


Figure 3 Comparison of Echocardiography data from 2021 and 2022. A and B: Color Doppler echocardiography showing calcification in the posterior leaflet of the mitral valve. The calcification point in the posterior mitral lobe was lower in A than in B. A was obtained beginning in 2021, and B was obtained beginning in 2022. Arrow: The calcified area of the mitral valve.

FINAL DIAGNOSIS

The patient was diagnosed with SHPT and CAVB.

TREATMENT

The patient underwent PTX again in March 2021. Postoperative pathology revealed hyperplastic parathyroid tissue.

OUTCOME AND FOLLOW-UP

Following surgery, her iPTH level decreased to 51 ng/mL, and the patient developed hypocalcemia. One month postsurgery, the patient's iPTH levels were maintained at 54-110 ng/mL, her serum calcium levels were 1.75-2.35 mmol/ L, and her blood phosphorus levels were 0.5-1.72 mmol/L. The patient's bone pain and itching significantly improved, but the patient still had a CAVB on the ECG. One year postsurgery, the iPTH levels were maintained between 50 and 100 ng/mL, without any significant discomfort. Chest CT showed improved calcification of the left atrium and left ventricle (Figure 4A and C), while the ECG showed a CAVB. Repeat ECG 2 years after surgery revealed that the patient's ECG was reversed to a first-degree atrioventricular block (Figure 5), and the calcification point in the posterior mitral lobe was reduced on cardiac ultrasound (Figure 3B). Unfortunately, the patient's EF was significantly lower than before (37%).

DISCUSSION

Bone mineral metabolism disorders are prevalent complications in patients undergoing maintenance hemodialysis. High levels of calcium and phosphorus secondary to hyperparathyroidism can lead to ectopic calcification. Extensive myocardial calcification can cause refractory heart failure, while calcium deposits in the cardiac conduction system can lead to progressive arrhythmias, heart failure, and increased mortality[3]. Dysfunction of the cardiac conduction system due to calcification is infrequent but can lead to first-, second-, or third-degree atrioventricular block, affecting patient survival. The etiology of atrioventricular block can be functional or structural. Functional CAVB tends to be reversible and can be caused by factors such as autonomic disease, metabolic/endocrine disease, or drug-related disease. The reversibility of atrioventricular block secondary to bone mineral metabolism disorders in maintenance hemodialysis patients is still controversial, despite metabolic diseases being the primary cause.

Literature reports on the pathogenesis of atrioventricular block in hemodialysis patients are rare; according to the current knowledge, the etiology tends to be metastatic calcification, which includes calcification of the conduction system and the mitral valve ring. Notably, an early case report by Dreher et al[4] in 1975 demonstrated complete recovery of firstdegree atrioventricular block following complete PTX in an end-stage hemodialysis patient with cardiac metastatic calcification. Similarly, Fujimoto et al[5] reported the case of a hemodialysis patient with third-degree SHPT and

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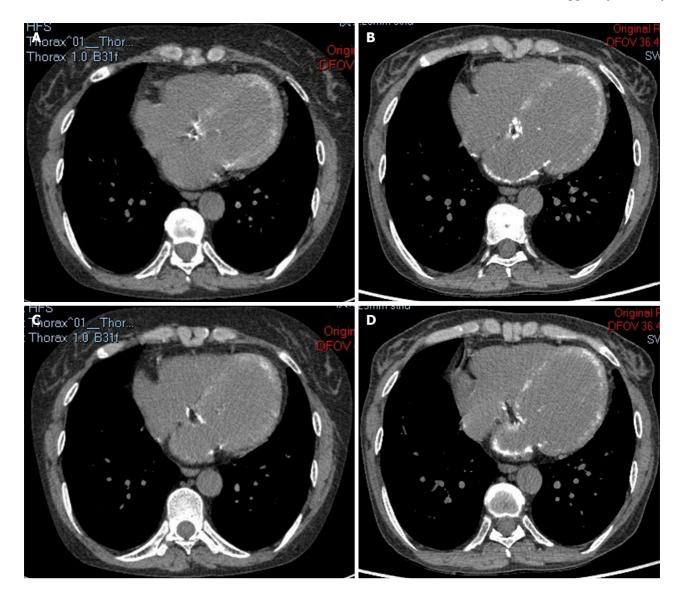


Figure 4 Comparison of chest computed tomography data from 2021 (B and D) and 2022 (A and C). Chest computed tomography revealed that the calcifications of the left atrium and left ventricle improved.

metastatic calcification resulting in atrioventricular block who died with extensive metastases despite hospitalization. Anam et al[3] recently described a case of CAVB secondary to severe hyperparathyroidism in a dialysis patient, highlighting the importance of suspecting metastatic cardiac calcification in cases of end-stage renal failure with elevated parathyroid hormone levels and concomitant first-degree atrioventricular conduction block and mitral annular calcification. To prevent progression of the atrioventricular block, measures should be taken to normalize parathyroid hormone, calcium, and phosphorus levels, possibly through exploration or removal of the parathyroid glands if the blockage worsens. In nonhemodialysis patients, primary hyperparathyroidism and hypercalcemia can also cause atrioventricular blockage due to calcium deposition, leading to atrioventricular node degeneration, as reported by Vosnakidis et al[6] in a case study involving dizziness, headache, and paroxysmal 2:1 atrioventricular block. Therefore, both primary and SHPT may lead to hypercalcemia and ultimately metastatic cardiac calcification, which is considered the primary mechanism underlying atrioventricular blockage.

Metastatic calcification in dialysis patients is a multifactorial condition, and its improvement is closely related to PTX. Uremic tumor calcinosis, corneal and conjunctival calcification, and arterial calcification are a few examples of conditions that are linked to this surgical intervention. Li et al^[7] reported complete relief of calcification in a patient with uremic tumor calcinosis two months after PTX. Another study confirmed that PTX in dialysis patients can rectify calcium and phosphorus metabolism disorders, reduce arterial calcification, and improve arteriosclerosis[8]. Additionally, a 2008 case report suggested that SHPT plays a critical role in the pathogenesis of tumor calcinosis in patients on hemodialysis[9]. Pessoa *et al*[10] reported the recovery of a patient with conjunctival and corneal calcification after PTX. In this case, the CAVB was caused by metastatic calcification of the heart, including metastatic calcification of the left atrium, left ventricle and mitral valve. The patient's atrioventricular block was reversed to a first-degree atrioventricular block 2 years after PTX surgery, and re-examination via chest CT and color ultrasound showed that the left atrial and left ventricular calcification was significantly better than before. Cardiac ultrasound showed that the calcification point in the posterior mitral lobe was reduced compared with that in the front; therefore, we considered that the reversal of CAVB in this

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Xu SS et al. Reversal of CAVB following parathyroidectomy

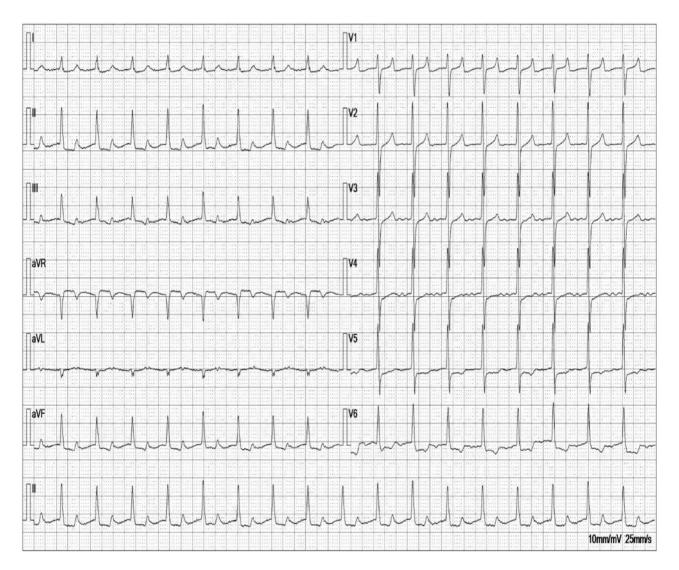


Figure 5 An electrocardiograph from 2022 showing a return to a first-degree atrioventricular block. The PR-interval was 342 ms.

patient was related to the improvement of metastatic calcification after PTX surgery. Since this patient refused to undergo pacemaker implantation, her cardiac function was reduced, and the PR interval was prolonged, although the CAVB block was reversed 2 years after the operation. Therefore, the author believes that in hemodialysis patients, the iPTH level should be optimized in the early stage, and patients should undergo relevant surgical treatment in a timely manner to improve their general condition.

CONCLUSION

This case reveals that metastatic cardiac calcification can result in complete atrioventricular blockage. Following parathyroid surgery, calcification of the cardiac conduction system improved, leading to reversal of the atrioventricular block. However, because cardiac function is significantly lower in these patients than before surgery, it is important for dialysis patients to receive optimized iPTH therapy and pay attention to calcification metastasis.

ACKNOWLEDGEMENTS

The authors thank the patient for participating in this study.

FOOTNOTES

Author contributions: Xu SS and Guan YM wrote the manuscript; Hao LH collected the data and drafted the manuscript; all the authors have read and approved the final manuscript.



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Supported by Weifang Health and Family Planning Commission Research Project, No. WFWSJK-2021-212.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors have no conflicts of interest to declare.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

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Country/Territory of origin: China

ORCID number: Yan-Meng Guan 0009-0004-0520-1490.

S-Editor: Gong ZM L-Editor: A P-Editor: Zheng XM

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