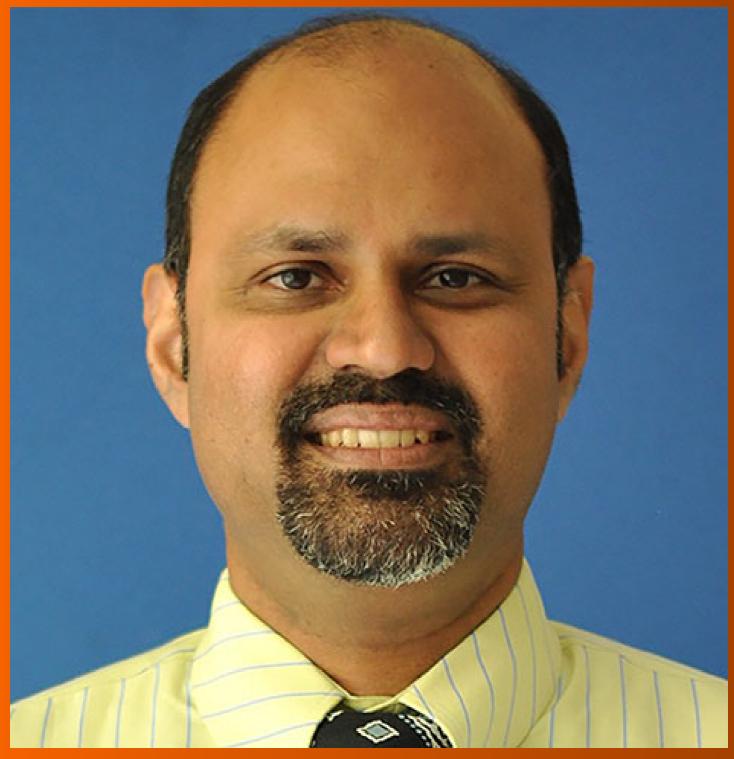
World J Clin Cases 2022 August 6; 10(22): 7620-8056



Contents

Thrice Monthly Volume 10 Number 22 August 6, 2022

OPINION REVIEW

7620 Whipple's operation with a modified centralization concept: A model in low-volume Caribbean centers Cawich SO, Pearce NW, Naraynsingh V, Shukla P, Deshpande RR

REVIEW

7631 Role of micronutrients in Alzheimer's disease: Review of available evidence

Fei HX, Qian CF, Wu XM, Wei YH, Huang JY, Wei LH

MINIREVIEWS

7642 Application of imaging techniques in pancreaticobiliary maljunction

Wang JY, Mu PY, Xu YK, Bai YY, Shen DH

7653 Update on gut microbiota in gastrointestinal diseases

Nishida A, Nishino K, Ohno M, Sakai K, Owaki Y, Noda Y, Imaeda H

7665 Vascular complications of pancreatitis

Kalas MA, Leon M, Chavez LO, Canalizo E, Surani S

ORIGINAL ARTICLE

Clinical and Translational Research

7674 Network pharmacology and molecular docking reveal zedoary turmeric-trisomes in Inflammatory bowel disease with intestinal fibrosis

Zheng L, Ji YY, Dai YC, Wen XL, Wu SC

Case Control Study

7686 Comprehensive proteomic signature and identification of CDKN2A as a promising prognostic biomarker and therapeutic target of colorectal cancer

Wang QQ, Zhou YC, Zhou Ge YJ, Qin G, Yin TF, Zhao DY, Tan C, Yao SK

Retrospective Cohort Study

7698 Is an oplasty superior to scar revision surgery for post-hemorrhoidectomy anal stenosis? Six years of

Weng YT, Chu KJ, Lin KH, Chang CK, Kang JC, Chen CY, Hu JM, Pu TW

Retrospective Study

7708 Short- (30-90 days) and mid-term (1-3 years) outcomes and prognostic factors of patients with esophageal cancer undergoing surgical treatments

Shi MK, Mei YQ, Shi JL



WJCC https://www.wjgnet.com

Contents

Thrice Monthly Volume 10 Number 22 August 6, 2022

7720 Effectiveness of pulsed radiofrequency on the medial cervical branches for cervical facet joint pain Chang MC, Yang S

7728 Clinical performance evaluation of O-Ring Halcyon Linac: A real-world study Wang GY, Zhu QZ, Zhu HL, Jiang LJ, Zhao N, Liu ZK, Zhang FQ

7738 Correlation between the warning symptoms and prognosis of cardiac arrest Zheng K, Bai Y, Zhai QR, Du LF, Ge HX, Wang GX, Ma QB

7749 Serum ferritin levels in children with attention deficit hyperactivity disorder and tic disorder Tang CY, Wen F

7760 Application of metagenomic next-generation sequencing in the diagnosis of infectious diseases of the central nervous system after empirical treatment

Chen YY, Guo Y, Xue XH, Pang F

7772 Prognostic role of multiple abnormal genes in non-small-cell lung cancer Yan LD, Yang L, Li N, Wang M, Zhang YH, Zhou W, Yu ZQ, Peng XC, Cai J

7785 Prospective single-center feasible study of innovative autorelease bile duct supporter to delay adverse events after endoscopic papillectomy

Liu SZ, Chai NL, Li HK, Feng XX, Zhai YQ, Wang NJ, Gao Y, Gao F, Wang SS, Linghu EQ

Clinical Trials Study

7794 Performance of Dexcom G5 and FreeStyle Libre sensors tested simultaneously in people with type 1 or 2 diabetes and advanced chronic kidney disease

Ólafsdóttir AF, Andelin M, Saeed A, Sofizadeh S, Hamoodi H, Jansson PA, Lind M

Observational Study

7808 Complications of chronic pancreatitis prior to and following surgical treatment: A proposal for classification

Murruste M, Kirsimägi Ü, Kase K, Veršinina T, Talving P, Lepner U

7825 Effects of comprehensive nursing on postoperative complications, mental status and quality of life in patients with glioma

Dong H, Zhang XL, Deng CX, Luo B

Prospective Study

7832 Predictors of long-term anxiety and depression in discharged COVID-19 patients: A follow-up study Boyraz RK, Şahan E, Boylu ME, Kırpınar İ

META-ANALYSIS

7844 Same-day single-dose vs large-volume split-dose regimens of polyethylene glycol for bowel preparation: A systematic review and meta-analysis

П

Pan H, Zheng XL, Fang CY, Liu LZ, Chen JS, Wang C, Chen YD, Huang JM, Zhou YS, He LP

Contents

Thrice Monthly Volume 10 Number 22 August 6, 2022

7859 Rectal nonsteroidal anti-inflammatory drugs, glyceryl trinitrate, or combinations for prophylaxis of postendoscopic retrograde cholangiopancreatography pancreatitis: A network meta-analysis

Shi QQ, Huang GX, Li W, Yang JR, Ning XY

7872 Effect of celecoxib on improving depression: A systematic review and meta-analysis

Wang Z, Wu Q, Wang Q

CASE REPORT

7883 Rectal mature teratoma: A case report

Liu JL, Sun PL

7890 Antibiotic and glucocorticoid-induced recapitulated hematological remission in acute myeloid leukemia: A case report and review of literature

Sun XY, Yang XD, Yang XQ, Ju B, Xiu NN, Xu J, Zhao XC

Non-secretory multiple myeloma expressed as multiple extramedullary plasmacytoma with an 7899 endobronchial lesion mimicking metastatic cancer: A case report

Lee SB, Park CY, Lee HJ, Hong R, Kim WS, Park SG

- 7906 Latamoxef-induced severe thrombocytopenia during the treatment of pulmonary infection: A case report Zhang RY, Zhang JJ, Li JM, Xu YY, Xu YH, Cai XJ
- 7913 Multicentric reticulohistiocytosis with prominent skin lesions and arthritis: A case report Xu XL, Liang XH, Liu J, Deng X, Zhang L, Wang ZG
- 7924 Brainstem abscesses caused by Listeria monocytogenes: A case report

Wang J, Li YC, Yang KY, Wang J, Dong Z

7931 Primary hypertension in a postoperative paraganglioma patient: A case report

Wei JH, Yan HL

7936 Long-term survival of gastric mixed neuroendocrine-non-neuroendocrine neoplasm: Two case reports

Woo LT, Ding YF, Mao CY, Qian J, Zhang XM, Xu N

7944 Percutaneous transforaminal endoscopic decompression combined with percutaneous vertebroplasty in treatment of lumbar vertebral body metastases: A case report

Ran Q, Li T, Kuang ZP, Guo XH

7950 Atypical imaging features of the primary spinal cord glioblastoma: A case report

Liang XY, Chen YP, Li Q, Zhou ZW

7960 Resection with limb salvage in an Asian male adolescent with Ewing's sarcoma: A case report

Lai CY, Chen KJ, Ho TY, Li LY, Kuo CC, Chen HT, Fong YC

7968 Early detection of circulating tumor DNA and successful treatment with osimertinib in thr790met-positive leptomeningeal metastatic lung cancer: A case report

Ш

Xu LQ, Wang YJ, Shen SL, Wu Y, Duan HZ

Contents

Thrice Monthly Volume 10 Number 22 August 6, 2022

7973 Delayed arterial symptomatic epidural hematoma on the 14th day after posterior lumbar interbody fusion: A case report

Hao SS, Gao ZF, Li HK, Liu S, Dong SL, Chen HL, Zhang ZF

- 7982 Clinical and genetic analysis of nonketotic hyperglycinemia: A case report Ning JJ, Li F, Li SQ
- 7989 Ectopic Cushing's syndrome in a patient with metastatic Merkel cell carcinoma: A case report Ishay A, Touma E, Vornicova O, Dodiuk-Gad R, Goldman T, Bisharat N
- 7994 Occurrence of MYD88L265P and CD79B mutations in diffuse large b cell lymphoma with bone marrow infiltration: A case report

Huang WY, Weng ZY

- 8003 Rare case of compartment syndrome provoked by inhalation of polyurethane agent: A case report Choi JH, Oh HM, Hwang JH, Kim KS, Lee SY
- 8009 Acute ischemic Stroke combined with Stanford type A aortic dissection: A case report and literature review

He ZY, Yao LP, Wang XK, Chen NY, Zhao JJ, Zhou Q, Yang XF

- 8018 Compound-honeysuckle-induced drug eruption with special manifestations: A case report Zhou LF, Lu R
- 8025 Spontaneous internal carotid artery pseudoaneurysm complicated with ischemic stroke in a young man: A case report and review of literature

Zhong YL, Feng JP, Luo H, Gong XH, Wei ZH

- Microcystic adnexal carcinoma misdiagnosed as a "recurrent epidermal cyst": A case report 8034 Yang SX, Mou Y, Wang S, Hu X, Li FQ
- 8040 Accidental discovery of appendiceal carcinoma during gynecological surgery: A case report Wang L, Dong Y, Chen YH, Wang YN, Sun L
- 8045 Intra-ampullary papillary-tubular neoplasm combined with ampullary neuroendocrine carcinoma: A case report

ΙX

Zavrtanik H, Luzar B, Tomažič A

LETTER TO THE EDITOR

8054 Commentary on "Primary orbital monophasic synovial sarcoma with calcification: A case report" Tokur O, Aydın S, Karavas E

Contents

Thrice Monthly Volume 10 Number 22 August 6, 2022

ABOUT COVER

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CASE REPORT

Primary hypertension in a postoperative paraganglioma patient: A case report

Jian-Hui Wei, Hai-Li Yan

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Abstract

BACKGROUND

Primary hypertension is a common clinical disease. Pheochromocytoma and paraganglioma is a rare cause of secondary hypertension. The diagnosis of the latter is still difficult, and the relationship between the two is not clear. The successful diagnosis of this case confirmed that standardized etiological investigation of secondary hypertension is necessary, contributes to the accurate diagnosis of rare diseases, and is conducive to the formulation or optimization of treatment plans. It shows an example of the coexistence of primary hypertension and secondary hypertension.

CASE SUMMARY

The patient was a 54-year-old male and was hospitalized with high blood pressure for 4 years. The patient's blood pressure was measured at 150/100 mmHg during physical examination 4 years ago and had no paroxysmal or persistent elevated blood pressure, no typical triad of headache, palpitation, and sweating, without postural hypotension. After taking nifedipine sustained release tablets intermittently, the blood pressure did not meet the standard. Physical examination revealed blood pressure of 180/120 mmHg. There was no abnormality in cardiopulmonary and abdominal examination. The results of blood and/or urinary catecholamines/metanephrine and normetanephrine before and after operation were normal. Fundus examination revealed retinal arteriosclerosis in both eyes. There was a history of paraganglioma diagnosed by pathology after retroperitoneal tumor resection, a family history of hypertension, and a history of passive smoking. The clinical diagnosis was subclinical paraganglioma, primary hypertension, and hypertensive fundus lesions. The patient's blood pressure was regulated, blood lipid was reduced, and anti-inflammatory, and symptomatic support were given. After treatment, the blood pressure was stable and up to standard without discomfort symptoms.

CONCLUSION

Subclinical paraganglioma and primary hypertension can coexist. The holistic thinking in clinical practice is helpful to the early diagnosis of rare diseases.

Key Words: Paraganglioma; Primary hypertension; Secondary hypertension; Diagnosis and Differential diagnosis; Genetic; Case report

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Core Tip: Pheochromocytoma and paraganglioma (PPGL) is a rare cause of secondary hypertension, and early and accurate diagnosis is still facing challenges. A case of subclinical paraganglioma (PGL) complicated with essential hypertension was analyzed retrospectively. A typical and subclinical pheochromocytoma and PGL should be paid more attention due to the lack of clinical features. At the same time, standardized etiological investigation of secondary hypertension is also an indispensable part of an accurate diagnosis. Clinical practice has proven that subclinical PGL and essential hypertension are two independent diseases that can coexist. After reviewing the literature, it is considered that genetic susceptibility is the same pathogenic factor.

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INTRODUCTION

Paraganglioma (PGL) is a rare cause of secondary hypertension that is manifested as a hypertensive crisis and easily leads to target organ damage. The reported prevalence of pheochromocytoma and paraganglioma (PPGL) is 0.2%-0.6%[1], with an incidence closely related to a germline gene mutation [2]. Surgery is the first option after diagnosis, resulting in a generally normal postoperative blood pressure. In this study, we present our findings on a confirmed case of combined postoperative PGL and primary hypertension.

CASE PRESENTATION

Chief complaints

A 54-year-old male had high blood pressure for 5 mo.

History of present illness

The patient's blood pressure was 186/100 mmHg at a physical examination 5 mo ago without discomfort. However, taking a 20 mg nifedipine sustained-release tablet once daily did not normalize his blood pressure. He had stopped the medication 3 mo prior to the time of admission. Since the disease onset, he had maintained a good diet and slept without snoring.

History of past illness

He had a history of PGL resection (Figure 1) and postoperative pathological diagnosis of PGL (Figure 2).

Personal and family history

He had a history of passive smoking, and family history of hypertension, but no PPGL.

Physical examination

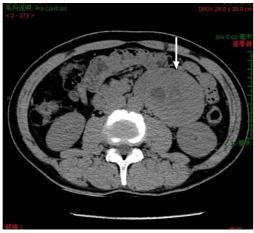
The physical examination at admission revealed a body temperature of 36.4 °C, pulse of 86 beats/min, breathing of 18 breaths/min, blood pressure of 188/108 mmHg, waist circumference of 96 cm, body mass index of 27.8 kg/m², clear mind, good spirit, and no murmur in neck and umbilical blood vessels. No abnormality was detected in the heart, lungs, and abdomen. No edema was found in both lower limbs, and positive nervous system signs were observed.

Laboratory examinations

Laboratory examinations revealed normal macrobiochemical parameters, thyroid function, parathyroid

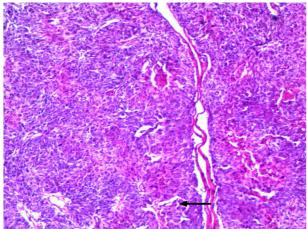


7932



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Figure 1 Computed tomography scan of paraganglioma. Performed on September 1, 2016: 64-slice computed tomography plain scan + enhanced scan (arrow). A mass of approximately 84 mm × 61 mm (right and left × back and forth) was observed below the left renal artery and vein, the abdominal aorta, the left psoas major muscle and the front of the left kidney. The edge was smooth, with an uneven density. The plain scan computed tomography value was within 17-41 HU. The arrow indicates the location, shape and size of the mass.



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Figure 2 Histopathological features of paraganglioma. The tumor represents characteristic nest-like structure (arrow, hematoxylin and eosin × 40). The physician who completed the pathological diagnosis was the chief physician, who had been engaged in pathological diagnosis for 31 years. The arrow refers to the typical pathological feature of paraganglioma - nest like structure.

hormone levels, cortisol and adrenocorticotropic hormone levels, and rhythm as well as normal prolactin, antinuclear antibody spectrum, and 24-h urine protein levels. Blood aldosterone and renin were determined by chemiluminescence measurements (11.31). Metanephrine was 31.9 ng/L (reference range < 96.60 ng/L), detected by liquid chromatography-tandem mass spectrometry. Normetanephrine was 68.9 ng/L (reference range < 163.00 ng/L).

Imaging examinations

Adrenal and renal artery computed tomography: Bilateral adrenal hyperplasia and right renal artery stenosis. Brain magnetic resonance imaging and chest, abdomen, and pelvic computed tomography were normal.

FINAL DIAGNOSIS

(1) Postoperative PGL; and (2) Primary hypertension with hypertensive retinopathy stage 2.

TREATMENT

Felodipine sustained-release tablet of 5 mg was administered once daily combined with olmesartan/hydrochlorothiazide tablet of 20 mg/12.5 mg once daily.

OUTCOME AND FOLLOW-UP

The patient had no symptoms. At the last follow-up examination on April 23, 2021, the blood pressure was normal and stable.

DISCUSSION

Hypertension was defined as systolic blood pressure ≥ 140 mmHg and/or diastolic pressure ≥ 90 mmHg measured three times on a different day in the absence of antihypertensive medications. Secondary hypertension refers to hypertension caused by certain diseases or causes, accounting for about 5% of all hypertension, is characterized by endocrine dysfunction, such as primary aldosteronism and PPGL[3]. PGL patients, accounting for 15%-20% of all PPGL cases, have normal blood pressure without symptoms. They are distributed in the abdomen, chest, pelvic, neck, and brain tissue, especially in the retroperitoneum. A malignant tendency of PGL development has been reported, with a malignant transformation rate of 24%-50%[4]. No typical clinical manifestation of PPGL was observed in the present case.

The blood metanephrine was normal, and only the abdominal computed tomography revealed a left retroperitoneal mass. Importantly, postoperative pathological diagnosis of PGL should be differentiated from adrenocortical eosinophilic and low-grade neuroendocrine tumors[5]. The main difference among the three tumors is the intensity of the neuroendocrine markers; hence, we considered it was nonfunctional subclinical PGL, which was consistent with the results of previous studies[6,7]. PGL is curable secondary hypertension in which resection is to be performed after the diagnosis, which results in achieving normal postoperative blood pressure. This case completely differed from PPGL, with symptoms and positive examination results. It had high concealment, with no PPGL triad of headache, palpitation, sweating, and hypertension, and the specific marker of blood metanephrine was normal. The increase in the blood pressure occurred 3 years after the PGL operation.

Differential diagnosis and screening of PGL metastasis were performed based on the specific medical history, clinical manifestations, etiology of secondary hypertension, distribution of PGL, and the site of metastasis. Renal parenchymal hypertension, renovascular hypertension, primary aldosteronism, sleep apnea hypopnea syndrome, hypercortisolism, pituitary tumors, thyroid and parathyroid dysfunction, pharmacogenic hypertension, and connective tissue disorders, such as vasculitis and systemic sclerosis, were excluded[8,9]. No recurrence or metastasis was observed in PGL, and the diagnosis of primary hypertension was clear.

Meanwhile, because of the lack of family history of hypertension, middle age, short course of disease, mild target organ damage, and PGL history, this case was different from the commonly known primary hypertension. High blood pressure occurred after the PGL operation. The diagnosis of the combination of postoperative PGL and primary hypertension was confirmed by recurrence and metastasis screening. After reviewing the literature[9], the diagnosis of postoperative PGL was clear albeit rare in clinical practice. No related report was available of subclinical postoperative PGL and primary hypertension, and thus we had to make the differential diagnosis. This case has deepened the clinician's understanding that primary hypertension and secondary hypertension can coexist. In the era of precision medicine, holistic thinking is helpful to the diagnosis and treatment of diseases.

CONCLUSION

Despite its rare occurrence, postoperative PGL patients can develop primary hypertension. The screening, diagnosis, and differential diagnosis of PPGL should be performed in cases with adrenal incidentaloma, retroperitoneal mass, or carotid body tumor. Pathological diagnosis is the gold standard for PPGL diagnosis.

FOOTNOTES

Author contributions: Wei JH conceived the idea, designed the experiments, and interpreted the data; Wei JH and Yan HL performed the experiments, analyzed the data, and wrote the manuscript; all authors reviewed and approved the manuscript.



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