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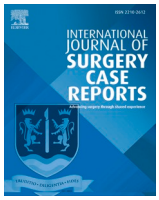

Case report

Gastric bronchogenic cyst mimicking adrenal Phaeochromocytoma: a case report

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ABSTRACT

Background: Bronchogenic cysts (BCs) are rare foregut-derived cystic malformations that can develop within the respiratory tract. While they are commonly found in the mediastinum or lungs, their occurrence at ectopic sites, such as the stomach, is extremely rare. This case report highlights the challenges in diagnosing a gastric bronchogenic cyst and the potential for misdiagnosis a pheochromocytoma, especially when associated with hypertension.

Case presentation: A 47-year-old male presented with a 6-day history of headache and nausea, and was found to have elevated blood pressure. Imaging studies, including computed tomography (CT) scans, suggested the possibility of a pheochromocytoma located near the left adrenal gland. However, subsequent surgical exploration revealed a cystic lesion near the posterior gastric wall, contiguous with the posterior gastric fundus. Pathological examination confirmed the diagnosis of a bronchogenic cyst in the gastric fundus.

Discussion: Bronchogenic cysts are congenital malformations that can present diagnostic challenges, especially when located in atypical sites like the stomach or when associated with hypertension, potentially mimicking pheochromocytoma. Accurate diagnosis relies on imaging, laboratory tests for metanephrines, and careful clinical assessment to differentiate from other tumors.

Conclusions: Correct differentiation between gastric bronchogenic cysts and pheochromocytoma is crucial, emphasizing the need for thorough diagnostic workup and considerate surgical approach.

1. Introduction

Bronchogenic cysts (BCs) are foregut-derived cystic malformations derived from the foregut and develop within the respiratory tract. They are typically located in the mediastinum at during early gestation or in the lungs at later stages [1]. However, BCs can occur at ectopic sites along the developmental pathway of the foregut. Notably, they are known for their clinical and radiological heterogeneity. Nevertheless, it is extremely rare for gastric BCs to arise from the posterior wall of the stomach [2]. Compared to the right adrenal gland, the left adrenal gland is surrounded by more complex adjacent structures, including the posterior gastric wall, the tail of the pancreas, the spleen, and the splenic flexure of the colon [3]. Misdiagnosis of left adrenal tumors as tumors adjacent to the adrenal gland is not uncommon. Herein, we present a case of a gastric bronchogenic cyst associated with hypertension that was misdiagnosed as a pheochromocytoma (PCC). This case serves as a reminder for urologists to reflect on such diagnostic challenges to avoid similar errors in the future. This case report has been reported according to the revised SCARE guidelines, 2023 [4].

2. Case presentation

A 47-year-old male with a 20-year history of smoking presented with a 6-day history of headache and nausea. On physical examination, his blood pressure was 190/110 mmHg. An adrenal CT revealed a soft-tissue density mass measuring 4.0 × 3.0 cm at its maximum diameter, located above the left adrenal gland, exhibiting no contrast enhancement (Fig. 1). An enhanced CT scan suggested pheochromocytoma, with a CT

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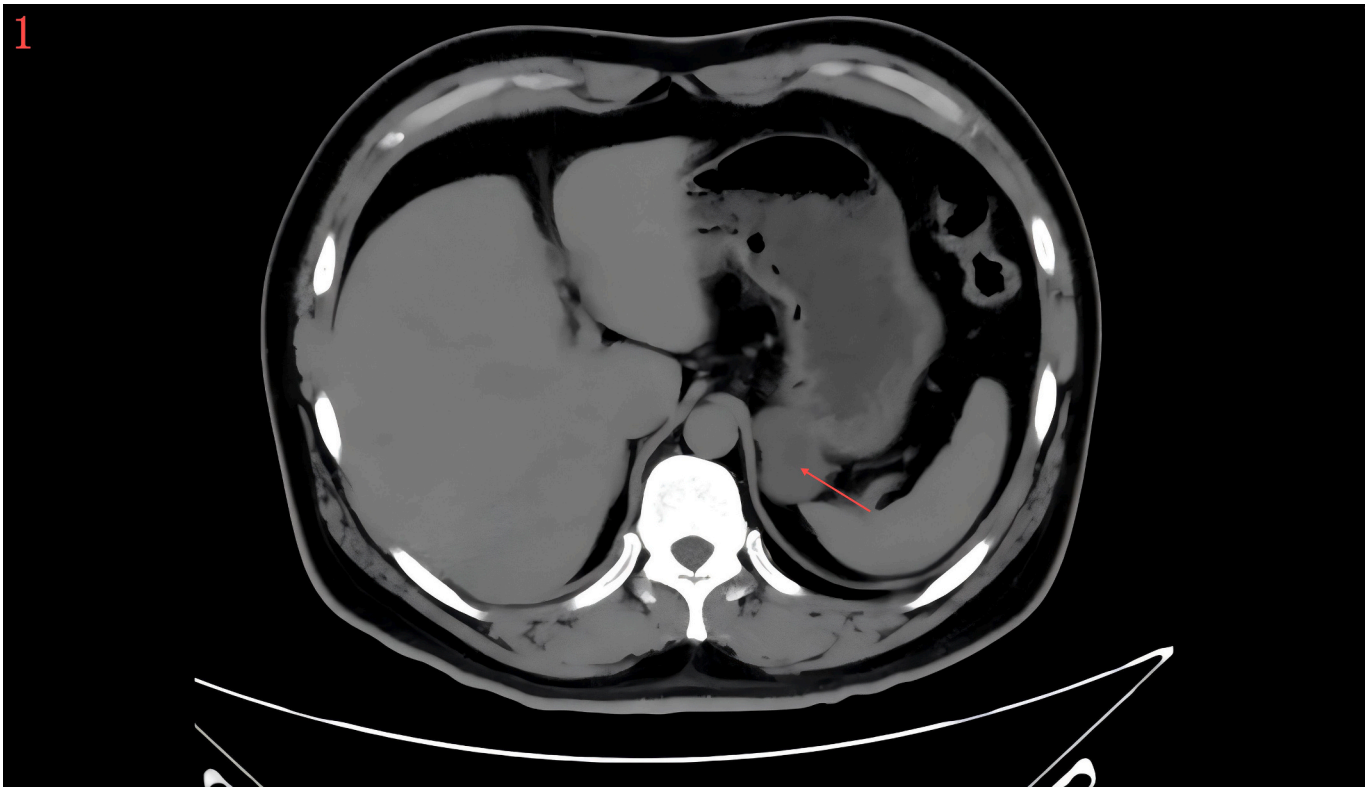

Fig. 1. An adrenal CT revealed a soft-tissue density mass measuring 4.0 × 3.0 cm at its maximum diameter, located above the left adrenal gland, exhibiting no contrast enhancement.

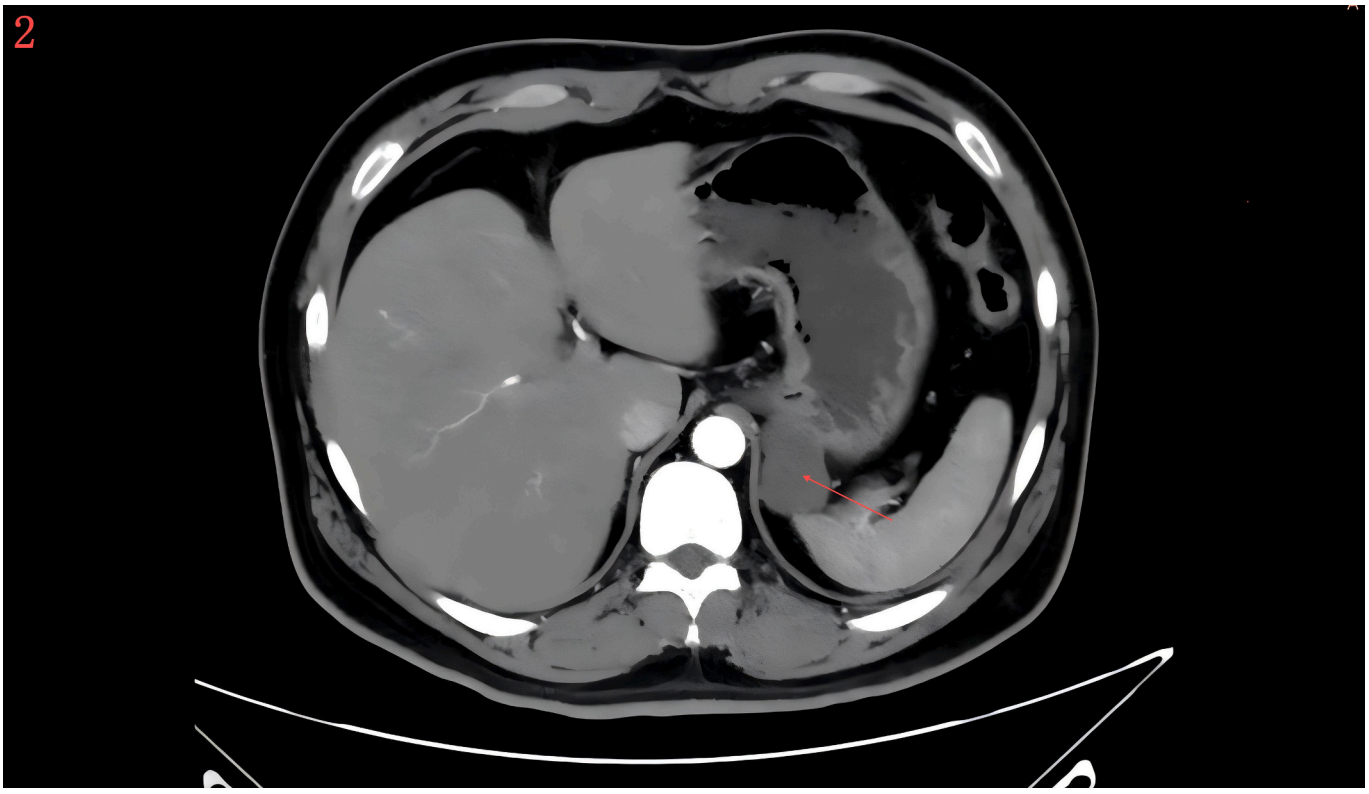

Fig. 2. An enhanced CT scan suggested pheochromocytoma, with a CT value of approximately 20 HU.

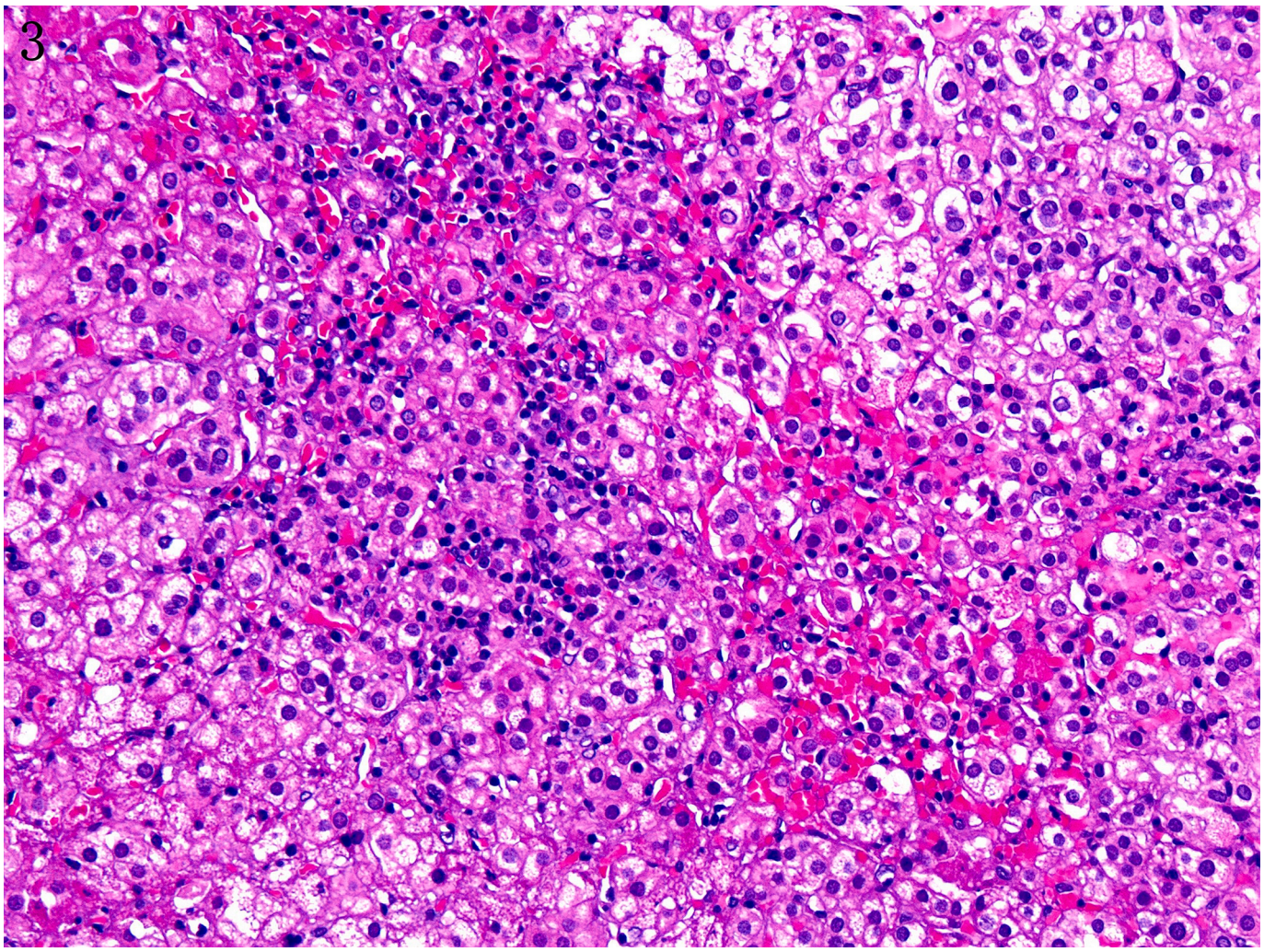

Fig. 3. Pathological examination confirmed the diagnosis of a bronchogenic cyst.

value of approximately 20 HU (Fig. 2). Laboratory results showed elevated vanillylmandelic acid (VMA) at 6.38 mg (normal 1.4–5.5 mg), renin at 1269.50 ng/L (normal 3.8–38.8 ng/L), and 24-hour urinary free cortisol at 765.60 nmol/24 h (normal 100–379 nmol/24 h). Vanillylmandelic acid (VMA), as one of the most important catecholamine metabolites, is commonly used to aid in diagnosis of pheochromocytoma [5]. Plasma catecholamine tests were normal. Based on these findings, the patient was diagnosed with pheochromocytoma. Oral antihypertensive medications were ineffective. After comprehensive preoperative preparation for oral alpha blockade and volume expansion therapy, a transperitoneal laparoscopic left adrenalectomy for pheochromocytoma was thus performed. However, no tumor was found in the left adrenal gland region. With assistance from a general surgeon, a cystic lesion measuring 4.5 × 3.0 × 3.0 cm was uncovered near the posterior gastric wall, adjacent to the tail of the pancreas, and found to be contiguous with the posterior gastric fundus. The cystic mass had a smooth surface and a large base filled with thick yellow fluid. The lesion was excised, and the patient had no postoperative complications. Pathological examination confirmed the diagnosis of a bronchogenic cyst in the gastric fundus (Fig. 3). The postoperative reconstructed image shows a round, low-density mass beneath the gastric fundus, clearly demarcated by a distinct margin, with a maximum diameter of approximately 2.6 cm (Fig. 4). The patient showed favorable recovery at the two-month follow-up, with normalized blood pressure levels.

3. Discussion

During the fourth week of fetal development, the primitive anterior foregut divides into a ventral tract and a dorsal tract. Bronchogenic cysts originate from abnormal budding in the primitive foregut and are typically located in the posterior mediastinum [6]. These benign congenital malformations of the primitive ventral foregut are lined with ciliated epithelium and contain focal areas of hyaline cartilage, smooth muscle, and bronchial glands within their walls. While most BCs are found in the mediastinum, they can also develop in the bronchial tree, in almost any intrathoracic location, and other areas such as the cervical region, thoracic cavity, abdomen, and skin [7]. Gastric bronchogenic cysts, a rare condition, are often initially misdiagnosed as gastrointestinal tumors, such as gastric stromal tumors. The symptoms they cause are usually non-specific, primarily arising from complications like local compression [8]. The modest cortisol rise was likely due to acute stress and/or compression of the adrenal outflow. After surgery, the patient's blood pressure returned to normal. A 24 - h urine collection revealed normal levels of VMA and urine - free cortisol. Plasma catecholamines and renin levels were also normal. We hypothesize that this normalization may be related to the removal of local compression on the left adrenal gland.

Gastric bronchogenic cysts associated with hypertension can be challenging to differentiate from left-sided adrenal tumors, such as pheochromocytoma, especially when presenting with atypical clinical features. Distinguishing between gastric bronchogenic cysts with

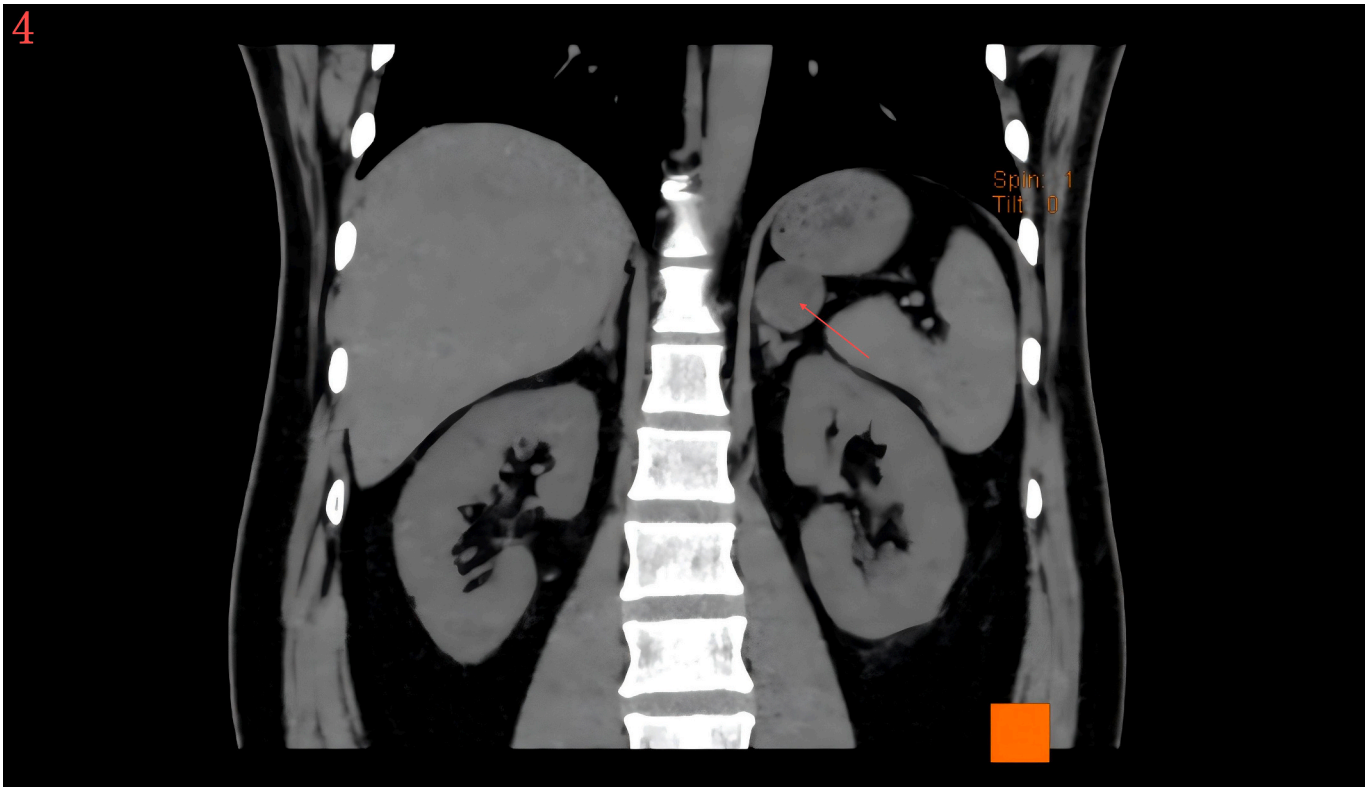

Fig. 4. The postoperative reconstructed image shows a round, low-density mass beneath the gastric fundus, clearly demarcated by a distinct margin, with a maximum diameter of approximately 2.6 cm.

hypertension and pheochromocytoma is critical for accurate diagnosis. The imaging features and laboratory results associated with pheochromocytoma provide essential criteria for its diagnosis and differentiation from other conditions. PCC is a rare neuroendocrine tumor characterized by the excessive production of catecholamines, such as epinephrine and norepinephrine, which are typically released episodically, leading to non-specific but potentially fatal clinical signs [9]. The diagnosis of PCC primarily relies on imaging localization and laboratory investigations. Enhanced CT scans typically show characteristic intense enhancement and an attenuation value of up to 190 HU in the venous phase, offering valuable diagnostic information for PCC [10]. An adrenal incidentaloma exhibiting high attenuation (>130 HU) on post-contrast CT is clinically suggestive of PCC [11]. In contrast, the CT enhancement effect of gastric bronchogenic cysts is not pronounced. MRI findings may present as iso- or hypointense on T2-weighted images, contrary to the belief that they always appear markedly hyperintense on T2-weighted images [12]. Diagnosing PCC in the left adrenal gland requires greater caution than for the right gland due to its complex anatomical relationships. We noted that larger tumors are often found in the left adrenal gland. Therefore, routine CT scans with three-dimensional reconstruction are crucial to rule out adjacent organ tumors.

Measurements of free metanephrine levels in plasma are the most accurate testing methods for detecting or excluding PCC. Clinically, many physicians still routinely measure deconjugated metanephrines (including both free and conjugated forms) along with VMA in 24 - h urine samples. Compared to tests for free metanephrines levels in plasma, tests for VMA and deconjugated metanephrines in 24 - h urine samples tend to yield more false - positive results. This finding supports a shift toward measuring free metanephrines instead [13]. Free metanephrines, especially plasma ones, have a higher diagnostic accuracy (sensitivity [SE] 97.9 % and specificity [SP] 94.2 %) than urinary and salivary ones [14]. Achote and et al. previously reviewed catecholamine metabolism in sympathetic tissues, the adrenal medulla, and pheochromocytoma tissue, providing convincing evidence that plasma free metanephrines are more specific for detecting PCC [15]. When plasma MN concentration is only 2–4 times above normal values, a clonidine suppression test can be performed, measuring plasma MN levels at baseline and three hours after oral administration of 0.3 mg clonidine, with suppression of plasma MN levels below 40 % suggestive of PCC [16]. It is important to note that urinary VMA levels may be affected by factors, such as smoking and variations in renal perfusion pressure [17]. Consequently, for patients presenting with left-sided PCC and insufficient evidence from endocrine examinations (when plasma MN concentration is only 2–4 times above normal values or perfectly normal), it is crucial to remain vigilant for abnormalities in endocrine examinations that may result from compression by adjacent tumors of the adrenal gland. This strategy could help avoid a hasty diagnosis of PCC.

Urologists often use the adrenal gland as a landmark for tumor localization and simultaneously perform total ipsilateral adrenalectomy. However, in this patient's situation, the removal of the adrenal gland could have been completely avoided. Dr. Nasim reported a case of a subdiaphragmatic bronchial cyst that masqueraded as an incidental adrenal tumor [18], which emphasizes the importance for urologists, general surgeons, and even thoracic surgeons to be aware of such conditions. When dealing with high-positioned left adrenal tumors with an ambiguous diagnosis, we strongly recommend choosing a transabdominal approach over a retroperitoneal approach for the surgical procedure. When the patient is in the right lateral decubitus position, resecting the short gastric vessels and the right gastroepiploic vessels causes the gastric fundus to shift to the right under the influence of gravity, fully exposing the pancreas and spleen. This approach avoids the need for extensive exposure of the pancreas and spleen, thereby simplifying the surgical procedure.

4. Limitations

Compared with enhanced CT, MRI has little advantage in adrenal tumor, but MRI may be more valuable for bronchogenic cyst. In the recent years, PET has become a key multimodality molecular imaging technique in the assessment of PCC, but PET-CT have Lower resolution than CT, variable performance across subtypes, costly, Exposure to ionizing radiations and lesions smaller than 3 to 5 mm are hard to detect [10]. Its conventional utility is still limited.

5. Conclusion

Bronchogenic cysts located at non-typical regions and, when accompanied by hypertension, might easily be misdiagnosed as PCC. Therefore, comprehensive CT scans with three-dimensional reconstruction, appropriate laboratory analyses, and selecting a suitable surgical approach are essential to avoid misdiagnosis and mistreatment. Moreover, adjunctive PET - CT or radiomics analysis may be considered in ambiguous cases to further clarify the lesion origin.

Author contribution

Fan-Fan Li and Xiao-Ping Li contributed equally to this work as co-first authors.

Constructing hypothesis for the manuscript: Fan-Fan Li and Xiao-Ping Li.

Logical interpretation and presentation of the results: Yong-Shun Li.

Organizing and supervising the course of the article: Xiao-Ping Wang.

Construction of the whole or body of the manuscript and taking responsibility: XiaoChun Yang.

All the authors read and agreed with the published version of the manuscript.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request. The privacy of patient is protected in this article.

Ethical approval

The case report was exempt from ethical approval by the Ethics Committee of Gansu Provincial Hospital, because we have obtained informed consent from the patient, which is not the design of the clinical trial. We have obtained informed consent from the patient and told the patient not at any risk to him. On the basis of full patient understanding and consent, informed consent was signed and consent was provided with the medical information to be used for scientific research and publications.

Guarantor

The guarantor is Xiao-Chun Yang.

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Declaration of competing interest

There is no conflict of interest.

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