CASE REPORT

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SYNCHRONOUS PRIMARY ATYPICAL PULMONARY CARCINOID AND CECAL ADENOCARCINOMA: A CASE REPORT

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Abstract. *Pulmonary carcinoid tumors are a rare subgroup of neuroendocrine lung neoplasms, accounting for approximately 1-2% of all primary lung cancers. The occurrence of synchronous primary malignancies involving pulmonary carcinoid and colorectal adenocarcinoma is exceptionally uncommon. We report a 78-year-old woman initially diagnosed with an endobronchial atypical carcinoid tumor of the left lower lobe. After initial refusal of surgical treatment by the patient and following endoscopic management, a left lower lobectomy with systematic lymphadenectomy was performed. Histopathology confirmed a G2 atypical carcinoid (pT1cN0R0, stage I). One month later, the patient developed symptomatic anemia and abdominal pain. Colonoscopy revealed a stenosing adenocarcinoma of the cecum. A right hemicolectomy with en bloc resection of infiltrated adjacent structures was performed. Final staging demonstrated moderately differentiated adenocarcinoma (pT3N0R0, stage IIA). The postoperative course after both procedures was uneventful, and the patient was referred for adjuvant oncologic evaluation. Synchronous primary atypical pulmonary carcinoid and colorectal adenocarcinoma is rare. This case underscores the importance of distinguishing metastatic disease from synchronous independent primaries in elderly patients with newly diagnosed malignancies and highlights the role of multidisciplinary staged surgical management in achieving favorable outcomes.*

Key words: *atypical pulmonary carcinoid, synchronous malignancy, cecal adenocarcinoma, surgical treatment, double primary malignancy*

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INTRODUCTION

Neuroendocrine tumors (NETs) arise from diffuse neuroendocrine cells and most commonly originate in the gastrointestinal tract and bronchopulmonary system. Pulmonary neuroendocrine tumors account for approximately 20-25% of all NETs and are classified into typical carcinoid, atypical carcinoid, large cell neuroendocrine carcinoma, and small cell lung carcinoma based on mitotic activity and necrosis [1]. Pulmonary carcinoids represent 1-2% of all primary lung malignancies and generally demonstrate indolent biological behavior, although atypical carcinoids exhibit more aggressive features, including higher mitotic rates and increased risk of nodal involvement [2, 3]. Surgical resection with systematic lymph node evaluation remains the standard of care for localized pulmonary carcinoids, particularly for atypical histology, where nodal metastases occur more frequently compared to typical carcinoids [3, 4].

Colorectal cancer is the third most commonly diagnosed malignancy worldwide and a leading cause of cancer-related mortality [5]. Right-sided colon cancers, including cecal adenocarcinoma, often present with non-specific symptoms, such as anemia and abdominal discomfort, which may delay diagnosis [6].

The occurrence of synchronous primary malignancies is reported in approximately 2-8% of cancer patients, and individuals with neuroendocrine tumors have been described to possess an increased risk of developing second primary neoplasms [7, 8]. Recent data from the Bulgarian population further highlight the clinical burden and heterogeneity of thoracic malignancies, particularly in relation to pleural involvement and comorbid oncologic conditions [9]. Although second primary malignancies have been described in patients with neuroendocrine tumors, the coexistence of pulmonary carcinoid and colorectal adenocarcinoma remains exceptionally rare, with only isolated reports of similar pulmonary or gastrointestinal neuroendocrine–colorectal combinations.

We present a case of synchronous primary atypical pulmonary carcinoid and cecal adenocarcinoma in a 78-year-old woman successfully treated with staged surgical resections.

CASE PRESENTATION

A 78-year-old woman was admitted to the Department of Thoracic Surgery with a five-month history of persistent cough and copious malodorous purulent sputum. She had previously been hospitalized for left-sided

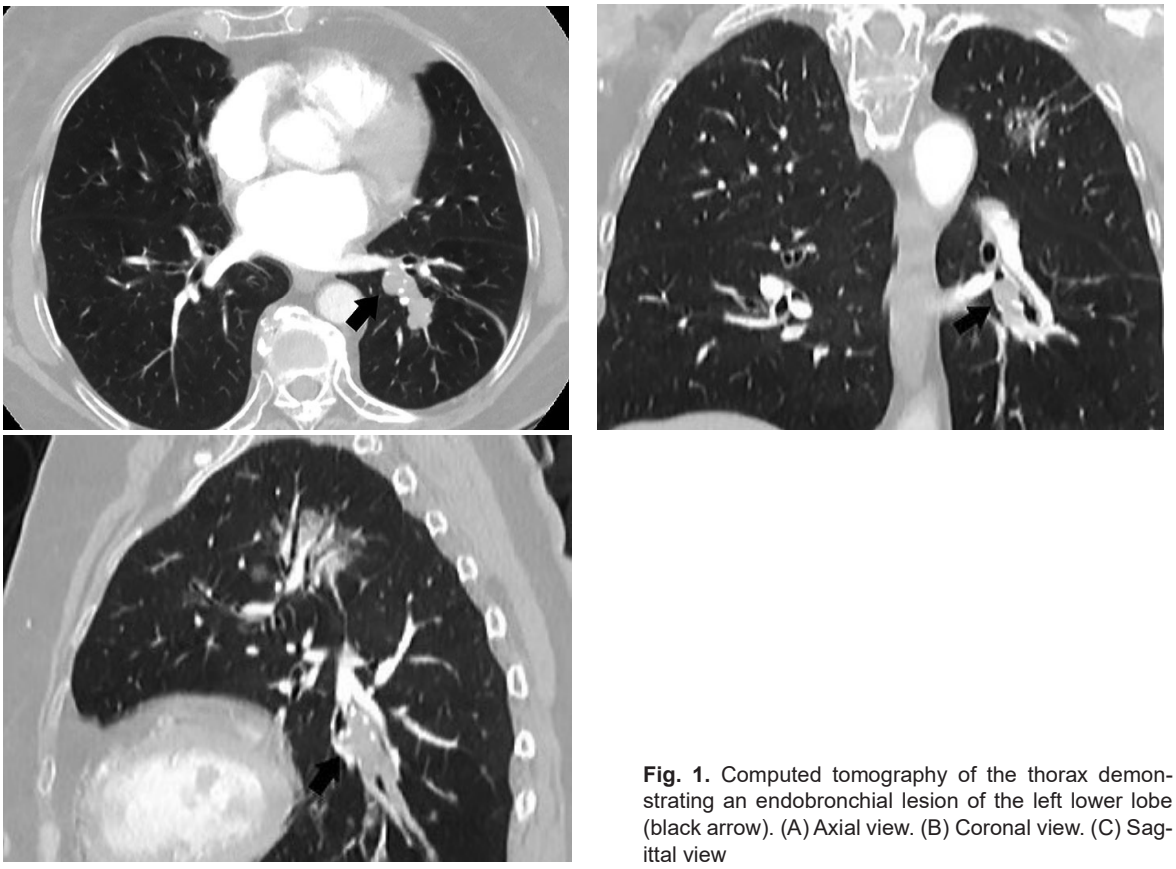


Fig. 1. Computed tomography of the thorax demonstrating an endobronchial lesion of the left lower lobe (black arrow). (A) Axial view. (B) Coronal view. (C) Sagittal view

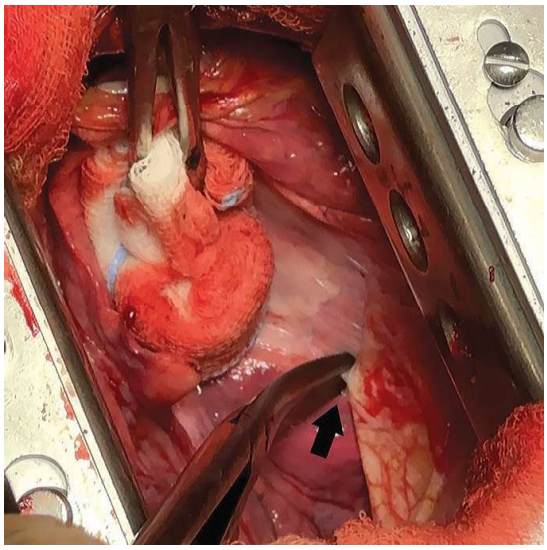
pneumonia with pleural effusion and later for suspected pulmonary thromboembolism. Despite adequate antimicrobial therapy, radiological abnormalities and elevated erythrocyte sedimentation rate (ESR) persisted.

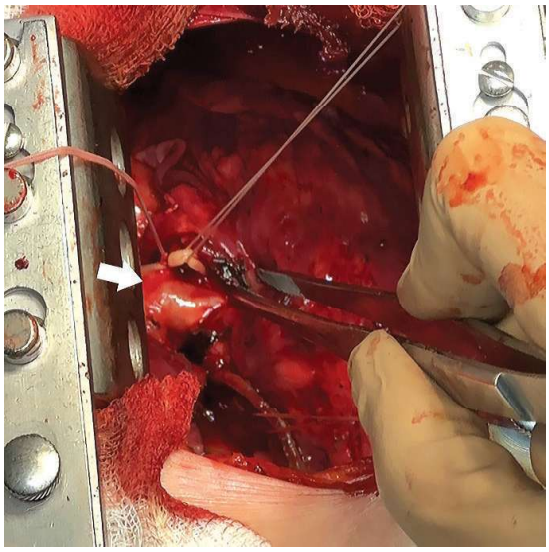
Single-photon emission computed tomography/computed tomography (SPECT/CT) demonstrated post-embolic subsegmental changes and residual pleural effusion. Flexible bronchoscopy (FBS) revealed an endobronchial lesion in the left lower lobe bronchus. Histopathological examination confirmed a carcinoid tumor. The patient initially declined surgical treatment.

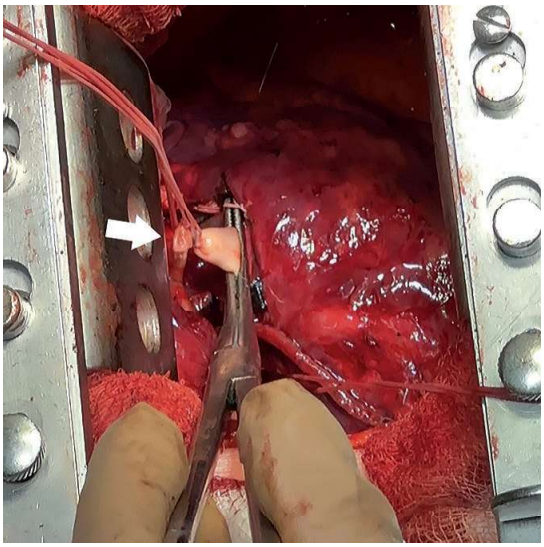
Five months later, follow-up bronchoscopy again demonstrated a lesion below the origin of B6, obstructing approximately 80% of the bronchial lumen. Transbronchial fine needle aspiration reconfirmed carcinoid tumor. Pulmonary function testing showed: vital capacity 1.69 L, FEV1 1.31 L, and Tiffeneau index 77.5%. Blood gas analysis was within normal limits.

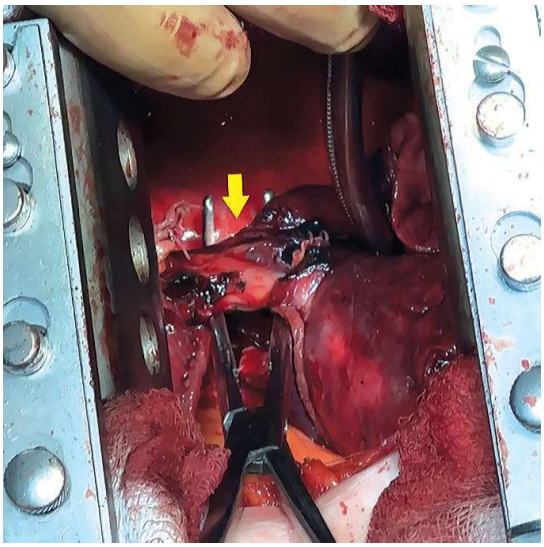
Chest CT angiography revealed a soft tissue lesion within the left lower lobe bronchus with distal extension into the segmental bronchi (Figure 1). In addition, irregular apical areas of consolidation and ground-glass opacities were observed in the left lung, corresponding to inflammatory changes in the stage of resorption. An enlarged right hilar lymph node measuring 17.2 × 16.6 mm in axial dimensions was also identified.

A left lateral minithoracotomy was performed. Intraoperatively, the left lower lobe was atelectatic and adherent to the parietal pleura (Figure 2). A left lower lobectomy with systematic lymphadenectomy (stations 5, 7, 10, and 11) was carried out (Figure 3). Frozen-section analysis of the bronchial margin confirmed tumor-free resection. The bronchial stump was closed using the Sweet technique.









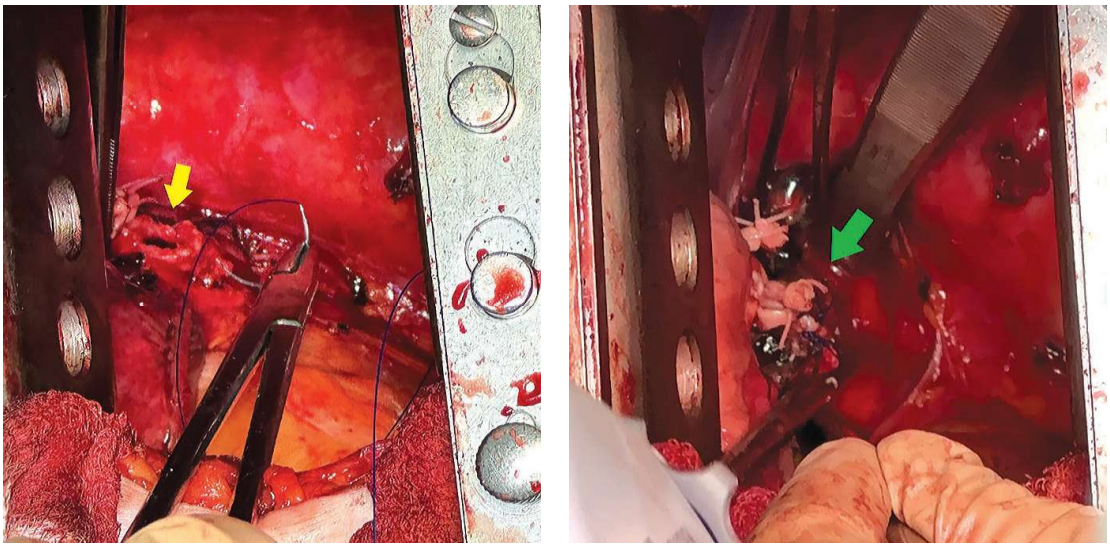


Fig. 2. Intraoperative findings during left lower lobectomy. A: Atelectatic left lower lobe adherent to the diaphragmatic pleura (black arrow indicates extrapleural dissection plane). B and C: Dissection and transection of the segmental artery (A6) and pyramidal artery (white arrow). D: Exposure of the lower lobe bronchus. (E) Closure of the bronchial stump using the Sweet technique. (F) Final hilar view after lobectomy (green arrow indicates arterial, venous, and bronchial stumps)

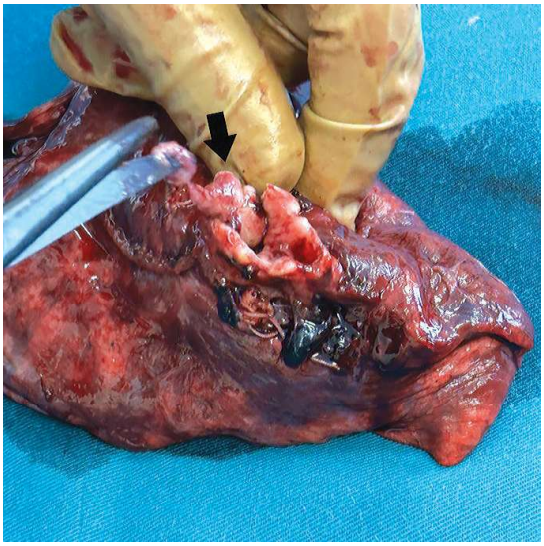


Fig. 3. Gross pathological specimen of the left lower lobe. The endobronchial atypical carcinoid is indicated by the black arrow

Histopathological examination confirmed atypical pulmonary carcinoid (G2) with a Ki-67 index of approximately 7%. Immunohistochemistry demonstrated diffuse chromogranin positivity, focal CD56 and INSM1 positivity, and negative TTF-1. All 16 dissected lymph nodes were negative for metastasis. Final staging was pT1cN0R0, Stage I.

One month postoperatively, the patient developed moderate anemia (hemoglobin 104 g/L) and colicky abdominal pain. A palpable mass was noted in the right abdomen. Abdominal CT demonstrated a 5-cm mass involving the cecum and terminal ileum, with suspected infiltration of adjacent retroperitoneal structures. Colonoscopy revealed a stenosing adenocarcinoma of the ascending colon.

A midline laparotomy was performed. Intraoperatively, a 5 × 5 cm tumor of the cecum was identified with adherence to the right psoas major muscle and the prerenal fascia. A right hemicolectomy was performed with en bloc resection of the infiltrated portion of the right psoas major muscle, together with adjacent retroperitoneal tissue, including the perirenal fascia and perirenal fat capsule of the right kidney (Figure 4). A latero-lateral ileotransversostomy was constructed. Due to the proximity of the tumor mass to the right infundibulopelvic ligament and the right adnexa, a

right adnexectomy was performed. Paracaval lymph nodes were not found, and one measuring about 7 × 8 mm in diameter at the border of the transition of the right iliac vein to the inferior vena cava was excised.

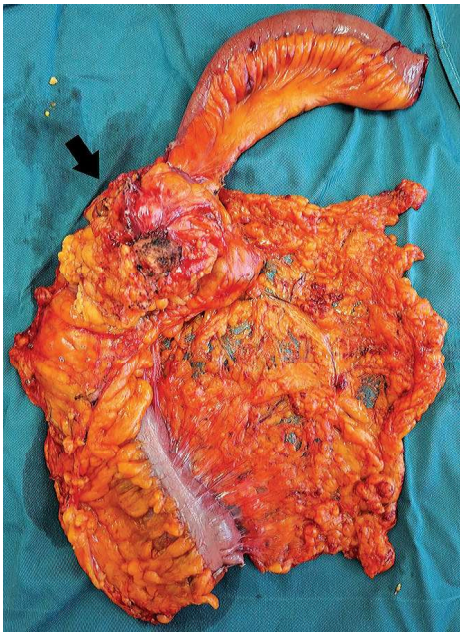


Fig. 4. Resected specimen following right hemicolectomy, including terminal ileum, cecum, ascending colon, omentum, portion of right psoas major muscle, and adjacent retroperitoneal tissue

Histopathology confirmed moderately differentiated adenocarcinoma of the cecum (G2), infiltrating through the muscularis propria into pericolic adipose tissue without visceral peritoneal involvement. All 20 harvested lymph nodes were negative for metastases. Resection margins were tumor-free. Final staging was pT3N0R0, stage IIA.

The postoperative course after both procedures was uneventful. The patient was discharged on postoperative days 8 and 12, respectively, and referred for multidisciplinary oncologic evaluation for adjuvant therapy.

At 6-month follow-up, no evidence of recurrence was detected.

DISCUSSION

Pulmonary carcinoid tumors are uncommon neoplasms accounting for 1-2% of all primary lung malignancies and approximately 20-25% of pulmonary neuroendocrine tumors [1, 2]. The potential complexity of pulmonary oncologic pathology is further illustrated by rare coexisting conditions that may pose diagnostic challenges [10]. Atypical carcinoids represent a more aggressive subgroup characterized by increased mitotic activity and the presence of necrosis, with a higher likelihood of nodal involvement and recurrence compared to typical carcinoids [3].

Surgical resection with systematic lymph node evaluation remains the gold standard for localized disease [4]. Endobronchial treatment is most effective in strictly intraluminal lesions smaller than 15 mm without bronchial wall invasion [11].

An important and unusual aspect of this case is the development of a second primary malignancy (cecal adenocarcinoma) within one month of lung surgery. The reported incidence of synchronous primary malignancies ranges between 2% and 8% among cancer patients. Population-level data also support the clinical relevance of second primary malignancies in patients with neuroendocrine tumors. Tsai et al. analyzed 1,350 patients with newly diagnosed NETs and found that 49 patients developed a metachronous second cancer, corresponding to an increased risk compared with the general population, particularly among patients aged 70 years or older [7]. Kamp et al. reported that 67 of 459 patients with gastroenteropancreatic NETs had a second primary cancer diagnosis, including synchronous tumors [8]. More recent registry data from England similarly showed increased rates of subsequent non-neuroendocrine malignancies, including lung and colon cancer, among patients with neuroendocrine neoplasms [12].

Several mechanisms may explain the association between neuroendocrine tumors and second primary malignancies. First, genetic susceptibility may be relevant in selected patients, particularly in hereditary endocrine tumor syndromes or in patients with defects in DNA repair pathways [13]. Second, shared carcinogenic exposures and host factors, including advanced age, smoking history, diet, chronic inflammation, and accumulated somatic mutations, may contribute to the independent development of both pulmonary and colorectal tumors [7]. Third, neuroendocrine tumors can produce bioactive peptides and growth factors, and several authors have proposed that tumor-related trophic signaling may create a permissive environment for additional neoplasia [7, 14]. Fourth, surveillance bias is plausible: once a NET is diagnosed, patients often undergo repeated imaging, endoscopic evaluation, and closer clinical follow-up, increasing the likelihood of detecting otherwise occult second primaries [7, 15]. In our case, however, the cecal tumor was not detected by routine surveillance alone, but by new-onset anemia, abdominal pain, and a palpable abdominal mass, suggesting that symptom-driven investigation was decisive.

Colorectal cancer is the third most common malignancy worldwide and remains a leading cause of cancer-related mortality [5, 6]. Right-sided colon cancers frequently present with anemia and non-specific abdominal symptoms, as observed in our patient. In this case, prompt colonoscopic evaluation following the onset of anemia led to early diagnosis of a locally advanced but node-negative tumor.

Reports specifically describing synchronous pulmonary carcinoid and colorectal adenocarcinoma are extremely limited. One of the closest published analogues is the case reported by Alghanmi, in which sigmoid adenocarcinoma occurred synchronously with two primary lung malignancies, including a pulmonary carcinoid tumor and lung adenocarcinoma [16]. Although that case differs from ours by the presence of an additional lung adenocarcinoma and by the colorectal tumor location, it is highly relevant because it illustrates the same diagnostic principle: pulmonary lesions in patients with colorectal cancer should not automatically be considered metastases, and histological confirmation may reveal independent primary tumors requiring separate staging and treatment.

From a surgical perspective, en bloc resection remains the standard treatment for locally advanced colon cancer involving adjacent structures [17]. Recent reviews have emphasized the diagnostic challenges and evolving therapeutic strategies in gastrointestinal malignancies, reflecting the broader complexity of oncologic management in the digestive tract [18, 19]. Although minimally invasive approaches have demonstrated oncologic equivalence in selected cases [20], an open approach was preferred in this patient due to suspected retroperitoneal infiltration. Complete (R0) resection was achieved, and no nodal metastases were identified.

The coexistence of atypical pulmonary carcinoid and colorectal adenocarcinoma within a short interval presents diagnostic and therapeutic challenges. A key clinical consideration is distinguishing between metastatic disease and true synchronous primary tumors. In this case, distinct histopathology, absence of metastatic spread, and independent TNM staging confirmed two separate primary malignancies.

Despite the patient's advanced age, staged surgical management resulted in favorable short-term and long-term outcomes. This case emphasizes the importance of comprehensive systemic evaluation in elderly patients diagnosed with pulmonary carcinoid tumors, particularly when new symptoms arise. Vigilance for second primary malignancies may allow timely intervention and improved prognosis.

CONCLUSION

Synchronous primary atypical pulmonary carcinoid and cecal adenocarcinoma is an exceptionally rare clinical entity. This case highlights the necessity of maintaining a high index of suspicion for second primary malignancies in patients diagnosed with neuroendocrine tumors, particularly when new systemic symptoms emerge. Accurate differentiation between metastatic disease and independent primary tumors is essential for optimal therapeutic planning. Staged radical surgical resection, guided by multidisciplinary evaluation, can achieve favorable outcomes even in elderly patients with complex oncologic presentations.

Ethics approval: Ethics committee approval was not required for this case report in accordance with institutional policy. The Helsinki Declaration was not relevant to this work.

COPE guidelines: Informed consent was obtained from the patient for all procedures. The admission consent form in the hospital implies agreement to educational/scientific activities (if no change in therapy/work-up plan is required). No change in therapy/work-up plan was required in this case.

Conflict of Interest Statement: *The authors declare no conflicts of interest related to this work.*

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