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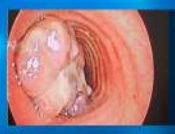

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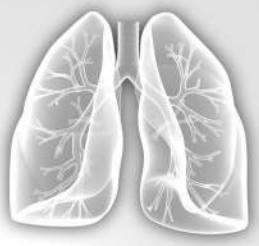

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Plastic Bronchitis Following Massive Hemothorax and Hemorrhagic Shock: A Case Report and Mechanistic Insights

Masif Hemotoraks ve Hemorajik Şok Sonrası Gelişen Plastik Bronşit: Olgu Sunumu ve Mekanistik Bulgular

Shengquan Wei, Jinghe Chen, Mingjun Wu

Abstract

Type 1 fibrinous plastic bronchitis (PB) is rare in adults, especially following massive hemothorax. A 62-year-old smoker developed extensive fibrinous bronchial casts after thoracoscopic evacuation of a massive right hemothorax (6,500 mL) with hemorrhagic shock, massive transfusion, and consumptive coagulopathy. Bronchoscopy on postoperative day 4 revealed branching casts in the segmental bronchi. Repeated mechanical extraction, saline lavage, and viscoelastic-guided correction of coagulopathy, without the use of intrabronchial fibrinolytics, led to resolution. Histopathology confirmed Type 1 inflammatory PB. Massive hemorrhage may trigger Type 1 PB through airway epithelial injury, a procoagulant milieu, and neutrophilic inflammation. Prompt bronchoscopic clearance with targeted hemostatic correction may be key when fibrinolysis is deferred because of bleeding risk.

Keywords: *Plastic bronchitis, hemothorax, hemorrhagic shock, coagulopathy, bronchoscopy.*

Öz

Tip 1 fibröz plastik bronşit (PB), özellikle masif hemotoraks sonrasında yetişkinlerde nadir görülmektedir. Altmış iki yaşında sigara içen bir hastada, hemorajik şok, yoğun kan transfüzyonu ve tüketim koagülopatisi ile birlikte sağda masif hemotoraksın (6500 mL) torakoskopik olarak boşaltılmasından sonra yaygın fibröz bronşiyal yapılar ortaya çıktı. Ameliyat sonrası 4. günde yapılan bronkoskopide segmental bronşlarda dallanan yapılar görüldü. Tekrarlanan mekanik ekstraksiyon, intrabronşiyal fibrinolitikler kullanılmadan sadece serum fizyolojik lavajı ve viskoelastik yönlendirmeli koagülasyonun düzeltilmesi, iyileşmeyi sağladı. Histoloji, Tip 1 inflamatuvar PB'yi doğruladı. Masif kanama, hava yolu epitel hasarı, prokoagülan ortam ve nötrofilik inflamasyon Tip 1 PB'i tetikleyebilir. Bu hastalarda, fibrinoliz olmaksızın, hemostatik dengenin düzeltilmesi ile hızlı bronkoskopik temizlik çok önemlidir.

Anahtar Kelimeler: *Plastik bronşit, hemotoraks, hemorajik şok, koagülopati, bronkoskopi.*

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Plastic bronchitis (PB) is a rare condition characterized by the formation and expectoration or bronchoscopic removal of firm, branching endobronchial casts that partially or completely obstruct the tracheobronchial tree (1,2). Two principal phenotypes have been described: Type 1 (inflammatory), consisting of fibrin-rich material with dense cellular infiltrates, and Type 2 (lymphatic/mucinous), which is often acellular and associated with lymphatic abnormalities (1,3). Although PB is most frequently reported in pediatric populations, particularly following Fontan palliation for congenital heart disease, adult cases, though rare, have been described in association with diverse etiologies, including lymphatic disorders, infections, and inflammatory conditions (4,5).

Recent advances in understanding PB pathogenesis have highlighted the potential role of neutrophil extracellular traps (NETs) in stabilizing fibrin matrices and promoting cast persistence within the airway lumen (6). However, the pathophysiologic mechanisms linking systemic hemorrhagic events to the development of Type 1 fibrinous PB remain poorly characterized. Here, we present an adult case of Type 1 PB that developed following massive intrathoracic hemorrhage and hemorrhagic shock, discuss mechanistic considerations grounded in contemporary literature, and describe a multimodal management approach that integrated mechanical clearance, inhaled anti-inflammatory therapy, and hemostatic correction while deferring fibrinolytic therapy because of ongoing bleeding risk.

CASE

A 62-year-old man with a 40-pack-year smoking history presented with acute right-sided pleuritic chest pain and progressive dyspnea following a forceful coughing episode. The patient reported a fall four days before symptom onset. Initial computed tomography (CT) imaging demonstrated a large right hemothorax with mediastinal shift and passive atelectasis of the right lower lobe (Figure 1). Emergency thoracoscopic evacuation was performed, with an estimated intraoperative blood loss of 6,500 mL.

The postoperative course was complicated by refractory hemorrhagic shock necessitating massive transfusion (12 units of packed red blood cells, 8 units of fresh frozen plasma, and 2 pools of platelets), vasopressor support, and invasive mechanical ventilation. The patient did not experience hemoptysis during the hospital course, although bronchoscopy revealed blood-tinged sputum, findings attributable to the underlying intrapleural bleeding and airway mucosal irritation rather than primary pulmonary hemorrhage.

On postoperative day 4, persistent lobar collapse and worsening hypoxemia (PaO2/FiO2 ratio, 150 mmHg)

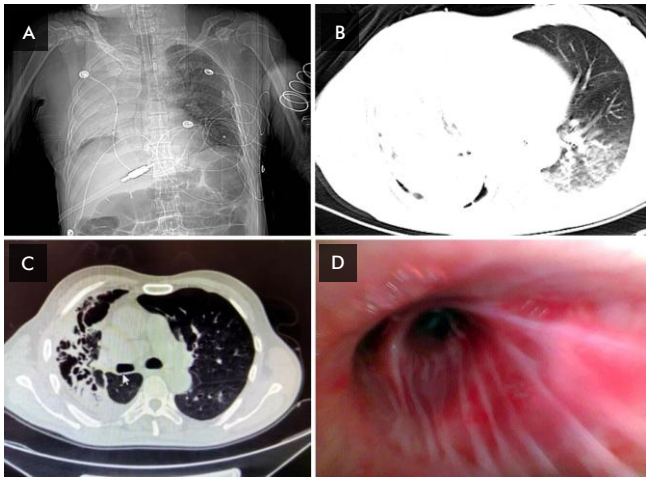

Figure 1: Preoperative chest radiograph showing a large right-sided hemothorax with associated lung compression, leftward mediastinal shift, and passive atelectasis of the right lower lobe (A); postoperative day 3 chest CT (axial view) following thoracoscopic evacuation showing persistent right opacification and cast-related lobar collapse despite pleural drainage (B); postoperative week 5 chest CT (axial view) demonstrating complete resolution of hemothorax, re-expansion of the right lung, and absence of residual pleural fluid (C); bronchoscopic view at week 5 confirming complete clearance of airway casts with restoration of normal bronchial mucosa (D).

prompted urgent flexible bronchoscopy. Endoscopic examination revealed extensive yellowish, focal fibrinous bronchial casts admixed with blood-tinged sputum, with branching casts occluding the posterior segmental bronchi of the right lower lobe and the apical segment of the right upper lobe (Figure 2). The casts were cohesive and rubbery in consistency. Under general anesthesia, mechanical extraction was performed using a cryoprobe and flexible forceps, followed by segmental saline lavage.

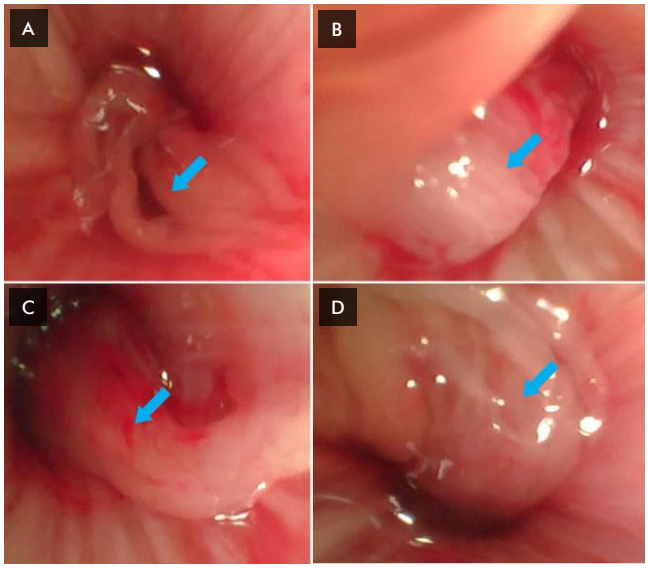

Figure 2: Endobronchial casts extracted by bronchoscopy. Representative photographs of fibrinous bronchial casts retrieved during serial bronchoscopic procedures on postoperative days 4–12. The casts exhibited characteristic yellowish-tan coloration, branching morphology reflecting segmental airway architecture, and rubbery consistency. Multiple sub-branches corresponding to distal bronchi were visible.

Given the inflammatory nature of the casts and the patient's smoking history, adjunctive inhaled therapy was initiated, comprising budesonide 1 mg nebulized twice daily and salbutamol 2.5 mg with 2 mL of normal saline nebulized twice daily. Serial bronchoscopic procedures were performed every other day to address recurrent cast formation, with the gross cast yield progressively declining after each session. By week 2, the airway fibrinous material was markedly reduced; however, the patient declined continued nebulized therapy. Bronchoscopic clearance was continued three times weekly during weeks 3–5 until airway casts were no longer visible endoscopically (Figure 1D).

Laboratory evaluation revealed a platelet nadir of 26 $\times$ 10^9/L, fibrinogen $<$0.8 g/L, and an international normalized ratio (INR) of 2.3, consistent with consumptive coagulopathy. Rotational thromboelastometry (ROTEM)-guided hemostatic interventions, including fibrinogen repletion with cryoprecipitate, platelet transfusion, and prothrombin complex concentrate as indicated, were instituted to correct systemic coagulopathy.

Histopathologic examination of the extracted casts demonstrated dense interlacing fibrin strands with abundant neutrophilic infiltration and negligible mucin content, confirming the diagnosis of Type 1 inflammatory PB (Figure 3).

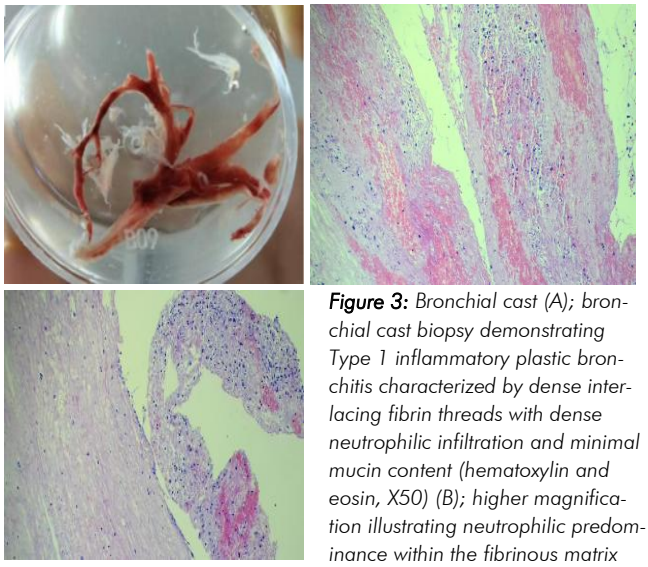

Figure 3: Bronchial cast (A); bronchial cast biopsy demonstrating Type 1 inflammatory plastic bronchitis characterized by dense interlacing fibrin threads with dense neutrophilic infiltration and minimal mucin content (hematoxylin and eosin, X50) (B); higher magnification illustrating neutrophilic predominance within the fibrinous matrix (H&E, X100) (C).

Bronchoalveolar lavage cultures and multiplex viral polymerase chain reaction (PCR) testing were negative for pathogenic organisms. Histopathologic examination of the right pleural lesion (parietal pleura surgical specimen) showed edematous fibroadipose tissue with focal lymphocytic infiltration, extensive subpleural hemorrhage, and fibrin deposition on the pleural surface, consistent with hemorrhagic pleuritis; acid-fast staining was negative for mycobacteria (Figure 4).

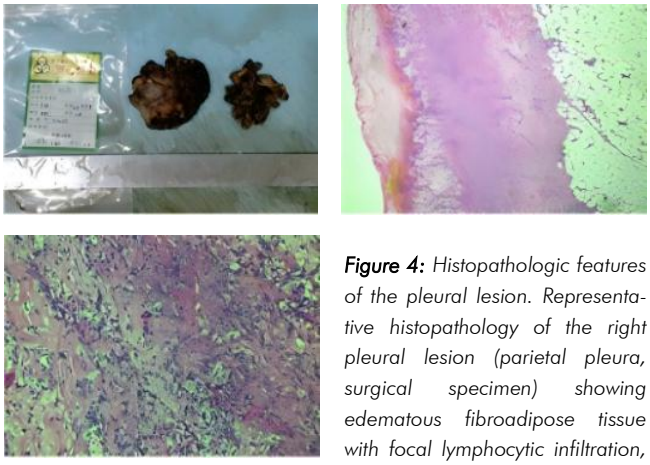

Figure 4: Histopathologic features of the pleural lesion. Representative histopathology of the right pleural lesion (parietal pleura, surgical specimen) showing edematous fibroadipose tissue with focal lymphocytic infiltration, extensive subpleural hemorrhage, and fibrin deposition on the pleural surface (hematoxylin and eosin stain, X50) (A,B); acid-fast staining negative for mycobacteria (C).

Given the potential association between PB and hemodynamic perturbations, transthoracic echocardiography was performed and revealed normal biventricular systolic function, no valvular abnormalities, and no evidence of pulmonary hypertension or right heart dysfunction. These findings excluded a significant cardiac contribution to cast formation in this case.

In view of the ongoing systemic bleeding risk, intrabronchial fibrinolytic therapy was withheld. Management focused on four key elements: (a) repeated mechanical clearance to restore and maintain airway patency, (b) segmental saline lavage to facilitate removal of residual fibrinous material, (c) inhaled corticosteroids and bronchodilators to attenuate airway inflammation and hyperresponsiveness, and (d) protocol-driven correction of systemic hemostatic derangements guided by viscoelastic testing.

Cast production diminished progressively, with substantial reduction by week 2 and complete clearance by week 5. Oxygenation improved (PaO2/FiO2 ratio, 500 mmHg), vasopressors were successfully weaned by day 12, and the patient was extubated on day 16. He was discharged to a rehabilitation facility on day 24 and remained free of recurrent respiratory symptoms at the three-month follow-up. Pulmonary function testing was not completed due to patient-related factors.

DISCUSSION

This case documents the occurrence of adult Type 1 fibrinous PB following massive intrathoracic hemorrhage and hemorrhagic shock, a scenario that illustrates how systemic hemorrhagic events and associated coagulopathy can create a profibrinogenic intrabronchial environment. Our multimodal management approach, combining serial bronchoscopic mechanical clearance, inhaled anti-inflammatory therapy, and targeted correction of

systemic hemostasis while deferring airway fibrinolytic therapy, proved successful and aligns with contemporary management strategies reported in the literature (4,7,8).

The pathophysiology of PB in this context likely involves three convergent processes. First, extensive intrathoracic bleeding and ischemia-reperfusion injury to the bronchial epithelium permit the ingress of fibrinogen-rich plasma into the airway spaces, with local coagulation activation favoring fibrin polymerization and deposition within the bronchial lumen. The patient's 40-pack-year smoking history may have predisposed him to preexisting airway epithelial vulnerability, potentiating fibrinogen leakage and impairing normal mucociliary clearance mechanisms. Second, systemic consumptive coagulopathy and transfusion-related hemostatic perturbations may create a regional procoagulant milieu, with an imbalance between coagulation and fibrinolysis, while stasis within poorly ventilated or collapsed lung segments facilitates cast consolidation. Third, the dense neutrophilic infiltrate observed histologically suggests neutrophil-mediated scaffold formation through the release of proteases and neutrophil extracellular traps (NETs), which have been implicated in stabilizing fibrin matrices and rendering casts resistant to mechanical disruption in other vascular contexts (6). Although specific immunohistochemical staining for NET markers was not performed, representing a limitation of this report, the histologic findings provide a plausible substrate for this mechanism, which warrants validation in future studies employing dedicated NET detection methodologies.

The management of PB requires a tailored approach based on cast pathology and clinical context. Ntiamoah et al. (4) proposed a diagnostic and therapeutic algorithm that emphasizes supportive care, bronchoscopic extraction, and targeted pharmacologic interventions according to cast type. For inflammatory (Type 1) casts, inhaled corticosteroids and bronchodilators may be beneficial, particularly in patients with underlying airway hyperresponsiveness or allergic predisposition. In the present case, the combination of budesonide and salbutamol was employed as adjunctive therapy to address both the inflammatory component of cast formation and potential bronchospastic elements related to the patient's smoking history. This approach was consistent with the observation that conservative medical management, including inhaled therapies, provides symptomatic relief and facilitates cast resolution in Type 1 PB (4,9).

Flexible bronchoscopy remains essential for diagnosis, enabling direct visualization and therapeutic removal of casts, although repeated procedures may be required to achieve and maintain airway patency (7,8). Mechanical extraction using forceps, cryoprobes, or snares, combined with segmental saline lavage, represents a pragmatic first-line strategy when systemic hemorrhagic risk precludes pharmacologic intervention. Intrabronchial fibrinolytic agents have demonstrated benefit in selected case reports, particularly in pediatric populations, but carry nontrivial hemorrhagic risk and are generally reserved for patients in whom bleeding risk is acceptably low (2,10). When systemic bleeding risk is present, targeted hemostatic correction guided by viscoelastic testing may reduce ongoing intrabronchial fibrin formation and support bronchoscopic clearance efforts, as observed in this case.

Clinicians should consider PB in the differential diagnosis of adults with refractory lobar collapse or unexplained hypoxemia following massive hemothorax or chest trauma. The present case highlights several key management principles: prompt bronchoscopic evaluation, serial mechanical clearance as needed, adjunctive inhaled anti-inflammatory therapy where appropriate, and correction of systemic coagulation abnormalities with the aid of viscoelastic testing. Deferring fibrinolytic therapy was considered safe and potentially advisable in the setting of ongoing bleeding risk, provided that mechanical clearance was thorough, hemostasis was optimized, and anti-inflammatory therapy was instituted. Future studies should systematically measure airway fibrinolytic activity and inflammatory mediators, including NET components, to better define the molecular mechanisms underlying cast formation and persistence.

Limitations

This is a single case report and cannot establish causality. Owing to technological and equipment-related constraints, airway fibrinolytic biomarker measurements and dedicated lymphatic imaging were not performed. Specific immunohistochemical staining for NET markers, such as citrullinated histone H3 and myeloperoxidase, was also not performed. Consequently, the proposed NET-mediated mechanism remains speculative despite the dense neutrophilic infiltrate observed histologically.

CONCLUSION

This case demonstrates that Type 1 fibrinous plastic bronchitis may complicate massive intrathoracic hemorrhage and hemorrhagic shock in adults. A pathophysiologic triad—airway epithelial injury with blood exposure, a local procoagulant microenvironment, and neutrophil-driven scaffold formation—likely underlay cast formation in this case. Early bronchoscopic diagnosis, repeated mechanical clearance, adjunctive inhaled corticosteroid and bronchodilator therapy, and targeted correction of systemic coagulopathy represent effective management strategies and may obviate the need for fibrinolytic thera-

py when bleeding risk is elevated. Increased awareness of this rare complication following hemorrhagic events may facilitate prompt diagnosis and improve patient outcomes.

ACKNOWLEDGEMENTS

We thank the bronchoscopy nursing team and pathology department for timely specimen handling and the cardiothoracic surgery and intensive care teams for multidisciplinary care

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - S.W., J.C., M.W.; Planning and Design - S.W., J.C., M.W.; Supervision - S.W., J.C., M.W.; Funding - J.C., M.W.; Materials - J.C., M.W.; Data Collection and/or Processing - S.W., J.C., M.W.; Analysis and/or Interpretation - S.W., J.C., M.W.; Literature Review - S.W., J.C.; Writing - S.W., J.C.; Critical Review - S.W., J.C., M.W.

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RESPIRATORY CASE REPORTS

Primary Malignant Melanoma of the Trachea: A Rare Case and Endobronchial Management

Trakeanın Primer Malign Melanomu: Nadir Bir Olgu ve Endobronşiyal Tedavi Yönetimi

Zehra Buşra Özbolat, Mehmet Akif Özgül, Efsun Gonca Uğur Chousein, Demet Turan, Ayşe Koyukan, Erdoğan Çetinkaya

Abstract

Primary malignant melanoma of the trachea is an exceptionally rare tumor that may cause life-threatening airway obstruction. This report presents a rare case of primary tracheal malignant melanoma successfully managed by rigid bronchoscopy. A 58-year-old man presented with dyspnea, cough, and inspiratory stridor. Computed tomography revealed a severely obstructive intraluminal tracheal mass. Rigid bronchoscopy demonstrated a vascular polypoid lesion obstructing approximately 85% of the tracheal lumen. Mechanical debulking, argon plasma coagulation, and cryotherapy were performed with therapeutic intent, achieving complete airway recanalization and immediate clinical improvement. Histopathologic examination confirmed malignant melanoma with positivity for S-100, Melan-A, and HMB-45. Dermatologic, ophthalmologic, and radiologic evaluations excluded another primary tumor site or distant metastasis, supporting the diagnosis of primary tracheal malignant melanoma. No complications, recurrence, or disease progression were observed during the 12-month follow-up period. This case highlights the importance of interventional bronchoscopy in rare central airway tumors.

Keywords: *Primary malignant melanoma, trachea, airway obstruction, bronchoscopy, APC.*

Öz

Primer trakeal malign melanom, yaşamı tehdit eden santral hava yolu obstrüksiyonuna yol açabilen oldukça nadir bir tümördür. Bu yazıda nadir görülen bir primer trakeal malign melanom olgusu ve rigid bronkoskopi ile yönetimi bildirilmiştir. Elli sekiz yaşında erkek hasta; dispne, öksürük ve inspiratuvar stridor ile başvurdu. Bilgisayarlı tomografide ciddi obstrüksiyona neden olan intratrakeal kitle saptandı. Rijid bronkoskopide trakea lümeninin yaklaşık %85'ini obstrükte eden vasküler polipoid lezyon izlendi. Küratif amaçla mekanik debulking, argon plazma koagülasyonu ve kriyoterapi uygulanarak tam hava yolu rekanalizasyonu ve hızlı klinik düzelme sağlandı. Histopatolojik inceleme S-100, Melan-A ve HMB-45 pozitifliği ile malign melanom tanısını doğruladı. Dermatolojik, oftalmolojik ve radyolojik değerlendirmelerde başka primer odak veya uzak metastaz saptanmadı. On iki aylık takip sürecinde komplikasyon, nüks veya hastalık progresyonu izlenmedi. Bu olgu, nadir santral hava yolu tümörlerinde girişimsel bronkolojinin önemini göstermektedir.

Anahtar Kelimeler: *Primer malign melanom, trakea, hava yolu obstrüksiyonu, bronkoskopi, APC.*

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Primary malignant tumors of the trachea account for less than 0.1% of all respiratory tract malignancies, and primary tracheal malignant melanoma is exceedingly rare (1). Clinical manifestations are generally nonspecific and include cough, dyspnea, stridor, and hemoptysis, often leading to delayed diagnosis. We report a rare case of primary tracheal malignant melanoma successfully managed by interventional bronchoscopy.

CASE

A 58-year-old man presented with progressive cough, dyspnea, and inspiratory stridor. Chest computed tomography demonstrated an obstructive intraluminal tracheal mass causing severe airway narrowing (Figure 1).

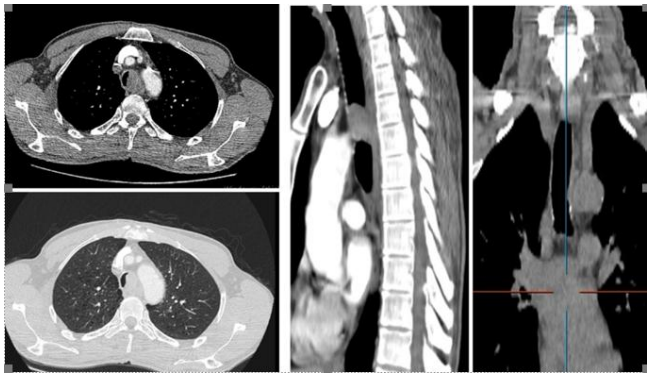


Figure 1: Chest CT demonstrating an obstructive intraluminal tracheal mass (sagittal and coronal view)

Rigid bronchoscopy revealed a vascular polypoid lesion arising from the left lateral tracheal wall and obstructing approximately 85% of the tracheal lumen over a 3–4 cm segment. The bronchoscopic intervention was performed with therapeutic intent to achieve complete airway recanalization. Mechanical debulking, argon plasma coagulation (APC), and cryotherapy were successfully applied, resulting in complete restoration of airway patency and immediate clinical improvement (Figure 2 and Videos 1 and 2).

Histopathologic examination confirmed malignant melanoma with positivity for S-100, Melan-A, and HMB-45. Dermatologic, ophthalmologic, and whole-body radiologic evaluations revealed no evidence of another primary tumor site or distant metastasis, supporting the diagnosis of primary tracheal malignant melanoma (Figure 3). The patient was referred for oncologic evaluation and adjuvant treatment planning.

The patient was followed for 12 months after the procedure. No early or late procedure-related complications, recurrent airway obstruction, or disease progression were observed during follow-up.

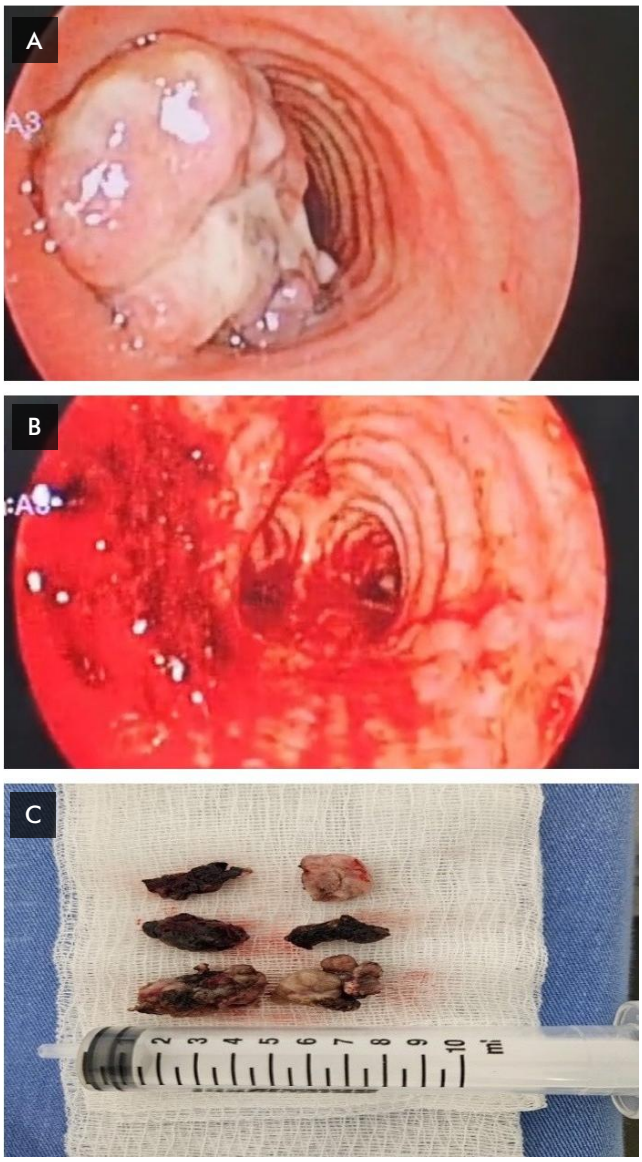
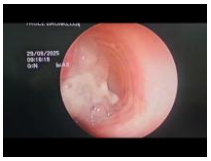


Figure 2: Rigid bronchoscopy views: intraluminal mass obstructing the tracheal lumen (A); restored airway after intervention (B); tumor specimens obtained for analysis (C)

Videos:



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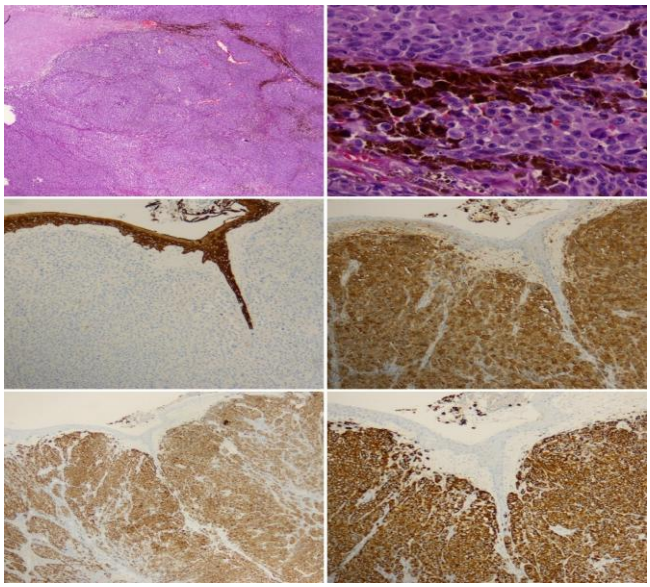

Figure 3: Histopathology: H&E, X40 (A); H&E, X400, showing pigmented tumor cells (B); H&E, X100, panoramic view (C); AE1/AE3 positivity on the epithelial surface, X100 (D); S-100 positivity in tumor cells, X100 (E); MART-1-positive cytoplasmic staining, X100 (F); HMB-45-positive melanocytic staining, X100 (G)

DISCUSSION

Primary malignant melanoma of the trachea is extremely rare, with only a limited number of cases reported in the literature (1,2). Differentiating primary from metastatic tracheal melanoma is essential but challenging, since histopathologic findings alone are insufficient for definitive distinction (3). Diagnostic criteria include the absence of cutaneous, mucosal, or ocular melanoma and the presence of a solitary tracheal lesion without systemic spread. In our patient, the diagnosis of primary tracheal melanoma was established based on the absence of skin or ocular lesions, a negative history of melanoma, and lack of distant metastatic disease (4).

Regardless of histologic subtype, tracheal tumors may lead to critical airway obstruction requiring urgent intervention (5). Restoration and maintenance of airway patency therefore represent a fundamental component of management. In the present case, bronchoscopic intervention was performed with therapeutic intent in the absence of detectable systemic disease. The patient presented with severe airway compromise associated with inspiratory stridor, and complete endobronchial tumor removal was achieved using rigid bronchoscopy combined with APC and cryotherapy. Immediate symptomatic and clinical improvement was observed following the procedure.

Rigid bronchoscopy offers both diagnostic and therapeutic advantages in central airway tumors. Mechanical debulking combined with ablative modalities such as APC and cryotherapy has been shown to effectively restore airway patency in selected cases. Importantly, no early or late procedure-related complications occurred in our patient, and no recurrent airway obstruction or disease progression was observed during the 12-month follow-up period. These findings support the role of interventional bronchoscopy as an effective and safe therapeutic modality in carefully selected patients with localized tracheal malignancies (5,6).

This case highlights the importance of interventional bronchoscopy in rare central airway tumors. Rigid bronchoscopy provides rapid airway stabilization as well as simultaneous diagnostic and therapeutic benefit in selected patients with critical airway obstruction.

Due to the rarity of primary tracheal melanoma, standardized treatment guidelines are lacking. Surgical resection may be considered in suitable patients, whereas adjuvant chemotherapy, radiotherapy, or immunotherapy may be required because of the risk of occult micrometastatic disease (6). Immune checkpoint inhibitors such as nivolumab and ipilimumab have demonstrated promising results in advanced melanoma and may also contribute to disease control in airway melanomas (7).

CONCLUSION

Primary malignant melanoma of the trachea is an exceptionally rare malignancy that may result in life-threatening airway obstruction. Early diagnosis and prompt bronchoscopic intervention are essential for both airway management and tissue diagnosis. In selected patients without systemic disease, interventional bronchoscopic techniques may provide complete airway recanalization and local disease control. A multidisciplinary approach remains crucial because of the absence of standardized management strategies for this rare entity.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - Z.B.Ö., E.Ç., M.A.O., E.G.U.C., D.T., A.K.; Planning and Design - Z.B.Ö., M.A.O., E.G.U.C., D.T., A.K., E.Ç.; Supervision - M.A.O., Z.B.Ö., E.G.U.C., D.T., A.K., E.Ç.; Funding -; Materials - Z.B.Ö.; Data Collection and/or Processing - Z.B.Ö.; Analysis and/or Interpretation - Z.B.Ö., M.A.O.; Literature Review - Z.B.Ö.; Writing - Z.B.Ö.; Critical Review -.

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Occult Intrapulmonary Sewing Needle Coexisting with Adjacent Bronchiolar Adenoma in an Adult: A Case Report

Yetişkin Bir Hastada Bronşiol Adenom ile Komşu Gizli Kalmış İntrapulmoner Dikiş İğnesi: Olgu Sunumu

Shengquan Wei[1], Bao Wei[2], Yu Chen[2], Baojun Zhang[2], Mingjun Wu[2]

Abstract

We report a rare case of an asymptomatic intrapulmonary foreign body (FB) coexisting with a benign neoplasm in a 29-year-old nonsmoking woman. The patient presented with nonspecific back pain without respiratory symptoms or a history of trauma. Chest CT incidentally revealed a 4.0-cm linear metallic density (2,761 HU) and a 5-mm subsolid nodule in the right lower lobe. Preoperative hookwire localization followed by video-assisted thoracoscopic surgery (VATS) enabled successful en bloc resection. A 4.0-cm sewing needle was extracted intact. Histopathology confirmed organizing pneumonia with chronic foreign body reaction and, notably, a benign bronchiolar adenoma adjacent to the FB. To our knowledge, this is the first reported coexistence of an occult intrapulmonary sewing needle and bronchiolar adenoma. Although the spatial proximity raises etiologic questions, the absence of histologic transition suggests a coincidental relationship. VATS with histopathologic correlation was essential for definitive diagnosis and management.

Keywords: *Intrapulmonary foreign body, Bronchiolar adenoma, VATS.*

Öz

Sigara içmeyen 29 yaşında bir kadında, iyi huylu bir neoplazm ile birlikte görülen asemptomatik intrapulmoner yabancı cisim olgusunu sunuyoruz. Hasta, solunum semptomları veya travma öyküsü olmaksızın, spesifik olmayan sırt ağrısı şikayeti ile başvurdu. Göğüs BT'sinde tesadüfen sağ alt lobda 4,0 cm uzunluğunda doğrusal metalik yoğunlukta bir alan (2.761 HU) ve 5 mm çapında subsolid bir nodül tespit edildi. Ameliyat öncesi tel ile işaretleme ve ardından video yardımlı torakoskopik cerrahi (VATS) ile başarılı bir en blok rezeksiyon yapıldı. Dört cm'lik dikiş iğnesi sağlam bir şekilde çıkarıldı. Histopatolojik incelemede organize pnömoni, kronik yabancı cisim reaksiyonu ve özellikle yabancı cisme bitişik iyi huylu bronşiolar adenom tespit edildi. Bildiğimiz kadarıyla, bu, gizli intrapulmoner dikiş iğnesi ile bronşiolar adenomun birlikte görüldüğü ilk olgudur. Mekansal yakınlık etiyolojik soruları gündeme getirirken, histolojik geçişin olmaması tesadüfi bir ilişki olduğunu düşündürmektedir. Histopatolojik korelasyonu olan VATS kesin tanı ve tedavi için gereklidir.

Anahtar Kelimeler: *İntrapulmoner yabancı cisim, bronşial adenom, VATS.*

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Retained intrapulmonary metallic foreign bodies (FBs) in adults are uncommon and typically result from penetrating trauma, aspiration, or iatrogenic procedures (1,2). Truly occult presentations, in which patients have no recollection of a causative event, are exceptionally rare and pose unique diagnostic and etiologic challenges (2,3). Furthermore, chronic FB retention can induce reactive parenchymal changes, potentially complicating the clinical and radiologic picture (4).

Bronchiolar adenoma (BA), also referred to as ciliated muconodular papillary tumor (CMPT), is a recently characterized benign peripheral lung neoplasm that typically manifests as a subsolid nodule on high-resolution CT (5,6). First described in a comprehensive case series in 2018, BA/CMPT is defined by its distinct bilayered epithelial architecture, comprising ciliated columnar cells overlying a continuous layer of basal cells (7).

Herein, we report a unique case of a truly occult intrapulmonary sewing needle discovered incidentally in a young woman, with the unexpected finding of an adjacent bronchiolar adenoma, a combination that, to our knowledge, has not been previously documented in the literature. This case is distinguished by three salient features: (i) a truly occult intrapulmonary metallic FB with no history of trauma or aspiration; (ii) the incidental discovery of an adjacent BA/CMPT, a neoplasm of uncertain pathogenesis; and (iii) the successful application of preoperative CT-guided hookwire localization to facilitate single-stage, minimally invasive resection of both lesions. We detail the clinical presentation, imaging characteristics, surgical management, and histopathologic findings while critically evaluating potential mechanisms of FB migration and the potential relationship between the FB and the coexisting adenoma.

CASE

A 29-year-old nonsmoking woman presented with a one-week history of intermittent back pain without cardiorespiratory symptoms. She denied fever, weight loss, hemoptysis, or a smoking history. Her medical history was unremarkable, and she specifically denied any history of trauma, aspiration, acupuncture, or sewing-related injury. Physical examination and routine laboratory tests were normal.

Chest CT images (lung window, 1.0-mm slice thickness) revealed a thin, linear hyperdense structure measuring 4.0 cm in length, located within the posterior basal segment (S10) of the right lower lobe (Figure 1A and B). The object demonstrated metallic attenuation (2,761 HU), with subtle surrounding ground-glass opacity suggestive of a localized inflammatory response. No direct pleural penetration site was evident; however, focal dorsal pleural thickening was noted. Adjacent to the needle tip, a well-defined 5-mm subsolid nodule was identified (Figure 1C). There was no evidence of mediastinal or hilar lymphadenopathy. Repeated and detailed questioning failed to elicit any potential source of the foreign body.

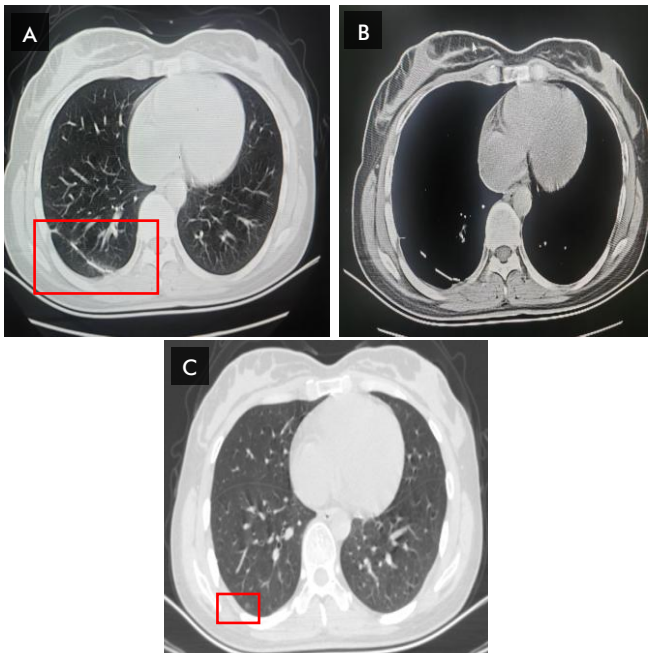

Figure 1: Preoperative chest computed tomography (CT), axial views, demonstrating an occult linear metallic hyperdensity (red box) in the posterior basal segment of the right lower lobe (A,B); adjacent 5-mm subsolid nodule (red box) in the lateral basilar segment, with minimal right dorsal pleural thickening (C).

Given the unexplained metallic FB and the indeterminate subsolid nodule, surgical intervention was planned. Preoperative CT-guided percutaneous placement of a hookwire was successfully performed to localize the small subsolid nodule adjacent to the needle tip. Under general anesthesia with double-lumen endotracheal intubation, a three-port VATS procedure was performed. Mild pleural adhesions over the posterior basal segment were lysed. The metallic FB was visualized embedded within the parenchyma and carefully extracted intact, revealing a 4.0-cm sewing needle (Figure 2A). The preplaced hookwire facilitated precise localization of the adjacent subsolid nodule, which was nonpalpable and not visible on thoracoscopic inspection. Wedge resection encompassing the needle tract and the localized nodule was performed using endoscopic staplers. The procedure was completed with minimal blood loss (estimated at 20 mL), and the patient was discharged uneventfully on postoperative day 3.

The wedge resection specimen measured 4.5 × 3.0 × 2.0 cm. A straight metallic sewing needle measuring 4.0 cm in length was embedded within the pulmonary parenchyma (Figure 2A). Adjacent to the needle tip, a firm,

grayish-white, well-circumscribed subpleural nodule measuring 0.5 cm in diameter was identified (Figure 2B).

Microscopic examination: Sections from the needle tract (Figure 2D) revealed organizing pneumonia characterized by intraluminal fibroblastic plugs (Masson bodies), dense chronic lymphohistiocytic infiltrates, abundant hemosiderin-laden macrophages, and scattered multinucleated foreign body-type giant cells. These findings were consistent with a chronic foreign body reaction. No granulomatous inflammation or necrosis was observed.

The 0.5-cm nodule (Figure 2C) demonstrated a papillary architecture lined by a distinctive bilayered epithelium: a luminal layer of ciliated columnar cells overlying a continuous basal cell layer. Foci of intraluminal mucin were present. No significant cytologic atypia, mitotic activity, or necrosis was identified.

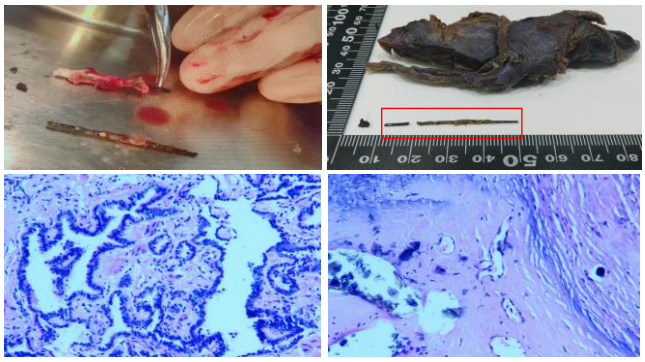

Figure 2: *Gross and microscopic pathology of the VATS resection specimen. Formalin-fixed wedge resection specimen showing the metallic sewing needle embedded within the pulmonary parenchyma (A); adjacent 5-mm subsolid nodule (red box) corresponding to bronchiolar adenoma (B); histopathology of bronchiolar adenoma demonstrating papillary architecture with bilayered epithelium, with inset showing p40 immunohistochemical positivity in basal cells (H&E, X100) (C); histopathology of the needle tract revealing organizing pneumonia with multinucleated foreign body-type giant cells and hemosiderin deposition (H&E, X50) (D).*

The basal cell layer demonstrated strong, continuous positivity for cytokeratin 5/6 (CK5/6) and p40, whereas the luminal columnar cells showed nuclear positivity for thyroid transcription factor-1 (TTF-1). This immunophenotype (basal cells: CK5/6+, p40+; luminal cells: TTF-1+) is considered pathognomonic for bronchiolar adenoma/CMPT (6,7).

DISCUSSION

This case is distinguished by three salient features: (i) a truly occult metallic FB with no history of trauma or aspiration, confirmed by comprehensive history-taking and the absence of chest wall scars; (ii) the incidental discovery of an adjacent bronchiolar adenoma (BA), a neoplasm of uncertain pathogenesis; and (iii) the successful application of preoperative CT-guided hookwire localization to facilitate single-stage, targeted VATS resection of both lesions.

The complete absence of respiratory symptoms, antecedent trauma, or identifiable risk factors rendered this metallic FB truly "silent" (1,3). Its incidental discovery during evaluation for vague back pain, likely referred pain from diaphragmatic or pleural irritation caused by the chronic inflammatory reaction (4), underscores the need for a broad differential diagnosis in patients with atypical thoracic findings. The coexistence of a 5-mm subsolid nodule further complicated the diagnostic pathway, as such lesions are radiologically suspicious for preinvasive neoplasms such as adenocarcinoma in situ, thereby necessitating tissue diagnosis (5,6).

Bronchiolar adenomas, formally recognized as benign peripheral lung neoplasms in the 2021 WHO classification of thoracic tumors (5), can recur locally if incompletely excised. Their presentation as subsolid nodules on imaging frequently mimics adenocarcinoma in situ or minimally invasive adenocarcinoma, making histopathologic confirmation the diagnostic gold standard (6). The defining feature distinguishing BA from its malignant mimics, particularly mucinous adenocarcinoma, is its characteristic bilayered epithelium with a preserved continuous basal cell layer (5,7).

The mechanism by which a sewing needle enters the lung without a remembered event remains unclear. Based on the foreign body's location in the posterior basal segment of the right lower lobe and the finding of focal dorsal pleural thickening, three plausible mechanisms were considered.

Transcutaneous migration with intrathoracic penetration: This is the most compelling explanation. Sharp foreign bodies, such as sewing needles or Kirschner wires, can penetrate the chest wall, particularly in the posterior thorax where soft tissue coverage is thinner, and subsequently migrate under the influence of respiratory motion, gravity, and negative intrathoracic pressure (8-10). Sato et al. (8) documented a striking case of intrathoracic needle migration from a left subclavian entry site, crossing the mediastinum to the right thoracic cavity, demonstrating the remarkable migratory potential of sharp objects within the thorax. They emphasized that respiratory and cardiac motion can propel needles along tissue planes, sometimes causing dynamic intraoperative displacement. Topaloğlu et al. (9) further showed that sewing needles can migrate through chest wall muscle planes, with the posterior thorax being particularly susceptible. The dorsal pleural thickening adjacent to the FB in our case likely represents the healed transcutaneous entry tract, providing supportive imaging evidence for this migratory pathway (8,9).

Silent aspiration with parenchymal penetration: It is possible that a sharp object could be silently aspirated, penetrate the bronchial wall during a bout of coughing or deep inspiration, and then migrate peripherally, driven by respiratory motion and negative pressure. Abraham et al. (11) documented that the high expiratory flow rates during coughing, reaching up to 12 L/s, can propel foreign bodies distally, and irregularly shaped objects may subsequently pierce the bronchial mucosa. Although well documented in children, this mechanism is exceptionally rare in adults without a witnessed aspiration event (2,3). The absence of respiratory symptoms and the peripheral parenchymal location do not preclude this possibility, as inorganic foreign bodies can evoke minimal inflammation and remain clinically silent for years (3,11).

Retrograde transdiaphragmatic migration: Although theoretically possible, this mechanism is unlikely in our case. Transdiaphragmatic migration is typically associated with gastrointestinal tract pathology, and migrated objects are more commonly found in the left thoracic cavity due to the anatomic relationship of the diaphragm with the stomach and spleen (12). The absence of gastrointestinal symptoms, subdiaphragmatic pathology, and a left-sided location in our patient makes this explanation improbable.

Clinicopathologic relationship between the foreign body and adenoma

The spatial proximity of the needle tract and the BA, approximately 5 mm, raises the intriguing question of a potential pathogenetic link. However, any causal relationship must be interpreted with extreme caution, as the evidence remains speculative and does not imply causation.

Chronic foreign body reactions can establish a proinflammatory microenvironment characterized by cytokine release, reactive oxygen species, and sustained epithelial injury, which could theoretically promote neoplastic transformation in susceptible bronchial epithelium (12). It is noteworthy that bronchiolar adenomas themselves frequently exhibit associated chronic inflammation on low-power histologic examination (7), suggesting that inflammatory microenvironments may play a role in their pathogenesis or progression. Despite this, several observations in our case support a coincidental rather than causal relationship: (i) the patient's young age and absence of smoking history minimize her baseline risk for neoplastic transformation; (ii) there is no histologic continuity or transitional morphology between the FB-induced inflammatory tract and the adenomatous lesion; (iii) no epithelial dysplasia, metaplasia, or other precancerous changes are present at their interface; (iv) BA is recognized to originate de novo from terminal bronchiolar epithelium, independent of exogenous stimuli (7); and (v) the duration of FB retention is unknown, creating temporal uncertainty. Therefore, although the hypothesis that chronic inflammation may promote adenoma development is biologically plausible, it remains conjectural and would require further investigation through additional case observations or experimental models.

To contextualize our findings, we reviewed previously reported cases of occult intrapulmonary metallic foreign bodies in adults. Sato et al. (8) reported intrathoracic needle migration from a remote entry site, highlighting the dynamic migratory capacity of sharp objects; however, their case involved a known iatrogenic source (acupuncture) rather than a truly occult presentation. More recently, Topaloğlu et al. (9) reported a case of a sewing needle found in the chest wall, emphasizing the potential for transcutaneous migration without a remembered event. Abraham et al. (11) documented spontaneous expulsion of an intrabronchial metallic pin in a pediatric patient, illustrating the potential for spontaneous migration but in a completely different clinical context. To our knowledge, no prior report has described the coexistence of an occult intrapulmonary FB with a bronchiolar adenoma, underscoring the unique nature of the present case.

Preoperative CT-guided hookwire localization was critical for the successful targeting of the subcentimeter subsolid nodule, which was neither visible nor palpable during VATS (13). This technique enables precise wedge resection while maximally preserving healthy pulmonary parenchyma. The efficacy of CT-guided percutaneous localization is well established; a large retrospective study by Guo et al. (13), including 599 patients, reported a 96.6% success rate for hookwire localization, with pneumothorax occurring in 27.2% and parenchymal hemorrhage in 33.9%, complications that were generally mild and self-limited. A 2025 meta-analysis by Wang et al. (14) comparing hookwire with anchored needle localization confirmed that both techniques are effective, although anchored needles may offer a slightly lower complication rate. The three-port VATS approach provided adequate visualization for simultaneous FB extraction and parenchymal-sparing resection of the nodule, achieving minimal morbidity and rapid recovery, outcomes consistent with contemporary standards for minimally invasive thoracic surgery (14,15).

CONCLUSION

This case raises two key issues: the origin of an asymptomatic intrapulmonary sewing needle, most plausibly through unrecognized transcutaneous migration, and its incidental adjacency to a bronchiolar adenoma. The absence of transitional morphology or histologic continui-

ty favors a coincidental relationship rather than inflammation-driven causation. Clinically, unexplained thoracic symptoms may prompt consideration of occult foreign bodies; CT-guided hookwire localization combined with VATS enables precise, minimally invasive removal, while histopathology is essential for distinguishing reactive changes from true neoplasms.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - S.W., B.W., Y.C., M.W., B.Z.; Planning and Design - S.W., B.W., Y.C., M.W., B.Z.; Supervision - S.W., B.W., Y.C., M.W., B.Z.; Funding - B.W., Y.C., B.Z., M.W.; Materials - B.W., Y.C., B.Z., M.W.; Data Collection and/or Processing - S.W., B.W., Y.C., B.Z.; Analysis and/or Interpretation - B.W., Y.C., M.W., B.Z.; Literature Review - S.W., B.W., Y.C.; Writing - S.W., B.W., Y.C.; Critical Review - S.W., B.W., M.W.

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Intrathoracic Extrapulmonary Vertebral Hydatid Cyst Causing Paraparesis: A Case Report

Paraparezi Gelişimine Yol Açan İntratorasik Ekstrapulmoner Vertebral Kist Hidatik Olgusu

Mustafa Kuzucuoğlu[1], Mehmet Unal[1], İsmail Ertan Sevin[2], Keramettin İbrahim Taylan[3], Bayram Çağrı Sakarıya[1]

Abstract

Hydatid disease is a parasitic infection caused by Echinococcus species. It is endemic in certain regions of the world and most commonly affects the liver and lungs, although cysts may develop in various organs. Intrathoracic extrapulmonary involvement is rare, and vertebral localization within this region is exceedingly uncommon. In spinal hydatid disease, in which neurologic symptoms may be prominent, surgical intervention remains the most effective treatment. Herein, we present the case of a 22-year-old patient treated for an intrathoracic extrapulmonary vertebral hydatid cyst, along with a review of the relevant literature.

Keywords: *Hydatid cyst, extrapulmonary, spinal, paraparesis.*

Öz

Kist hidatik, ekinekokların neden olduğu paraziter bir hastalıktır. Dünyanın bazı bölgelerinde endemik olan hastalık sıklıkla karaciğer ve akciğer olmak üzere vücudun birçok yerinde kistlerin gelişimine yol açabilmektedir. İntratorasik ekstrapulmoner yerleşimi nadir olup bu bölgelerden vertebra yerleşimli kist hidatikler daha da nadir görülmektedir. Nörolojik semptomları ön planda olan spinal kist hidatiklerde en etkili tedavi cerrahidir. Bizde 22 yaşında intratorasik vertebral kist hidatik nedeniyle tedavi ettiğimiz olgumuzu literatürler eşliğinde sunduk.

Anahtar Kelimeler: *Kist hidatik, ektrapulmoner, spinal, paraparezi.*

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Hydatid disease is a parasitic infection most commonly caused by Echinococcus granulosus and typically presents with cystic lesions in the liver and lungs, and less frequently in the brain. Rarely, the disease may present with intrathoracic extrapulmonary involvement, with cysts located in the mediastinum, pleura, or diaphragm. Vertebral involvement accounts for only 0.2–1% of all hydatid cyst cases and is therefore considered extremely rare. Within the vertebral column, the thoracic spine is most frequently affected, followed by the lumbosacral and cervical regions (1,2).

In this report, we present a rare case of an intrathoracic extrapulmonary hydatid cyst located at the cervicothoracic junction in a 22-year-old patient, along with a review of the literature.

CASE

A 22-year-old female patient presented to the neurosurgery clinic with progressive lower-extremity weakness that had begun approximately 6–7 months earlier during the postpartum period. Cervical magnetic resonance imaging (MRI) revealed a cystic mass lesion causing destruction of the C7–T1 vertebrae and resulting in a pathologic fracture. The lesion completely filled the left C6–C7 neural foramen and extended into the right neural foramen (Figure 1). Contrast-enhanced thoracic computed tomography (CT), performed for further evaluation, demonstrated multiple cystic lesions extending into the upper mediastinum at the cervicothoracic junction, with destruction of the C7 vertebral body (Figure 2).

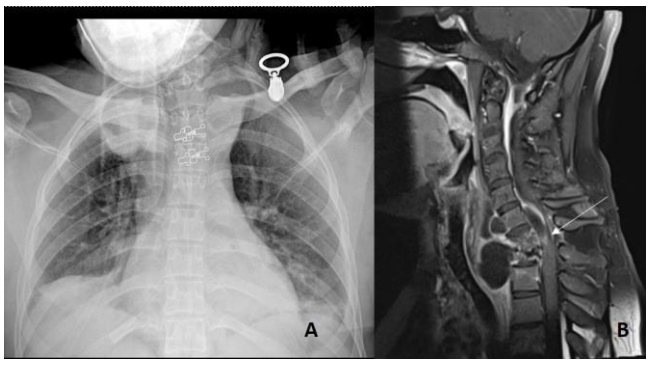

Figure 1: Chest radiograph showing bilateral apical opacities associated with mediastinal widening (A); cervical MRI demonstrating vertebral destruction (B).

The patient was jointly evaluated with the neurosurgery department. Due to the presence of paraparesis, an initial neurosurgical procedure was performed, including screw fixation at the right C4–C6 and T2–T4 levels and the left C4–C5 and T1–T4 levels, followed by total C7 laminectomy, C7–T1 corpectomy, and cage placement. During the procedure, part of the cyst content was aspirated, and the cystic components within the spinal canal were removed.

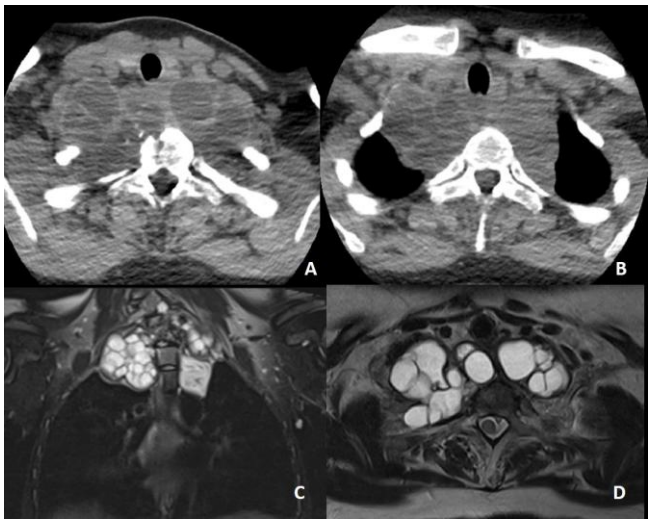

Figure 2: Thoracic CT (A,B) and MRI (C,D) showing an intrathoracic extrapulmonary cyst located in the apical region with vertebral destruction.

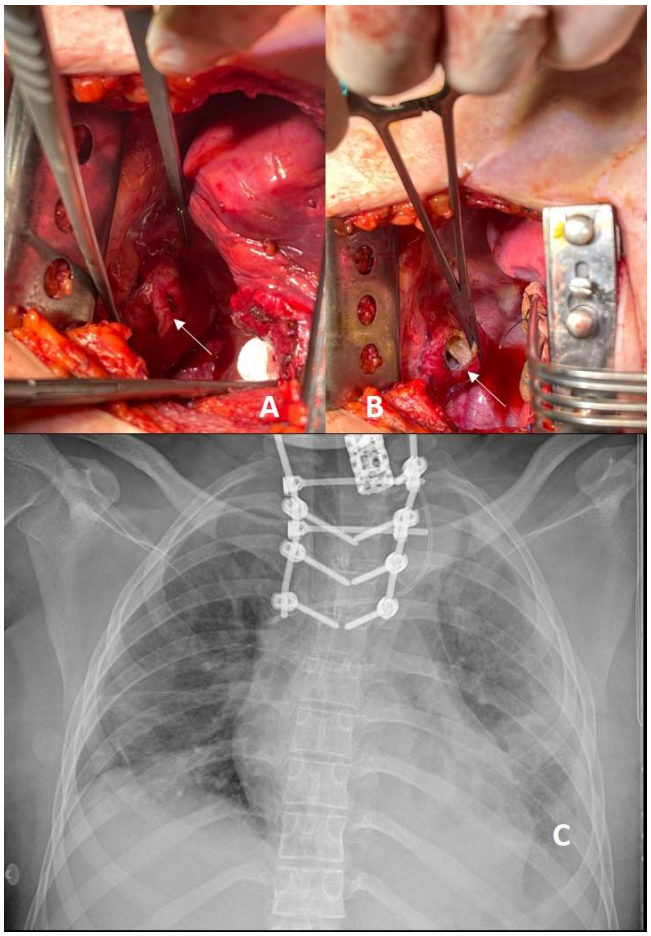

Figure 3: Intraoperative views of the cyst and cyst cavity (A,B); postoperative chest radiograph (C).

Subsequently, at one-month intervals, the patient underwent right thoracotomy to remove multiple cystic lesions extending from the right posterior mediastinum into the spinal canal, followed by left thoracotomy to excise cystic lesions located in the left posterior mediastinum with spinal canal extension. Cystotomy was performed, the germinative membranes were completely excised in both

hemithoraces, and the cyst cavities were irrigated with povidone-iodine solution (Figure 3).

Postoperatively, the patient showed gradual improvement in paraparesis, with increased ease of lower-extremity movement. She is currently continuing physical therapy and rehabilitation.

DISCUSSION

Hydatid disease, caused by Echinococcus species, is one of the oldest known parasitic infections. It is most commonly encountered in Mediterranean countries, South America, the Middle East, New Zealand, and Australia. In Türkiye, the incidence is approximately 20 per million populations. The liver is affected in 60–70% of cases, followed by the lungs as the second most common site. Although hydatid disease may involve nearly any organ except hair, teeth, and nails, intrathoracic extrapulmonary involvement remains rare (3,4).

Osseous involvement accounts for approximately 0.5–2.5% of all hydatid disease cases, with nearly half of these cases affecting the vertebral column. Vertebral hydatid cysts represent only 0.2–1% of all hydatid disease cases. The thoracic spine is most frequently involved, followed by the lumbosacral and cervical regions. Vertebral hydatid cysts typically cause destruction of the vertebral body and compression of the spinal canal or nerve roots, leading to slowly progressive neurologic symptoms. Paraparesis, paraplegia, and quadriplegia are among the most severe clinical manifestations and are often attributed to delayed diagnosis (2,5,6).

In our case, the hydatid cyst was located at the rarely involved cervicothoracic junction, extended into the posterior mediastinum, and caused significant vertebral destruction. In central nervous system hydatid disease, serologic tests may not always be diagnostic due to the blood-brain barrier, making imaging studies crucial for preoperative diagnosis. In endemic regions, hydatid disease should be considered in the differential diagnosis of vertebral destruction. MRI is the most effective imaging modality for demonstrating spinal canal compression and lesion extension along the vertebral column and is essential for surgical planning. The presence of multivesicular, grape-like cystic structures on MRI is highly characteristic of hydatid disease (7). In our patient, MRI and CT revealed multiple grape-like cystic lesions causing destruction of the C7–T1 vertebrae and extending into the spinal canal and mediastinum.

The main goals of surgical treatment in vertebral hydatid disease are decompression of the spinal cord, stabilization of the destroyed vertebral column, and complete excision of the cyst whenever feasible. Surgery is generally performed in two stages. The first stage involves posterior decompression of the neural elements, removal of destroyed bone, and stabilization. The second stage consists of anterior debridement of the vertebral body, complete cyst excision, and, if necessary, total vertebrectomy with reconstruction using iliac or fibular grafts. Due to extensive disease spread, complete excision is not always possible, and recurrence is common (8). In our case, a staged approach was applied as recommended in the literature, beginning with posterior decompression followed by anterior removal of residual cystic tissue. The postoperative improvement in paraparesis highlights the importance of early surgical decompression in achieving neurologic recovery.

CONCLUSION

Hydatid cysts may involve any tissue in the body, may radiologically mimic neoplastic lesions, and are primarily treated by surgical excision. Early surgical intervention is crucial for achieving rapid neurologic recovery and preventing permanent deficits.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - M.K., M.U., İ.E.S., K.İ.T., B.Ç.S.; Planning and Design - M.K., M.U., İ.E.S., K.İ.T., B.Ç.S.; Supervision - M.K., M.U., İ.E.S., K.İ.T., B.Ç.S.; Funding - M.K., M.U., İ.E.S., K.İ.T., B.Ç.S.; Materials - M.K., M.U., İ.E.S., K.İ.T., B.Ç.S.; Data Collection and/or Processing - M.K., M.U., İ.E.S., K.İ.T., B.Ç.S.; Analysis and/or Interpretation - M.K., İ.E.S., K.İ.T.; Literature Review - M.K., B.Ç.S.; Writing - M.K.; Critical Review - M.K., M.U., İ.E.S., K.İ.T., B.Ç.S.

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Primary Pulmonary Rhabdomyosarcoma Diagnosed from an Expectorated Tumor Bud

Ekspektore Edilen Tümör Parçasından Tanı Konulan Primer Pulmoner Rabdomyosarkom

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Abstract

Rhabdomyosarcoma is the most common soft tissue sarcoma. It shows skeletal muscle differentiation and mainly affects children. Thoracic involvement is rare, and pulmonary involvement is exceptional. We report the case of a 49-year-old patient who was asymptomatic except for grade II dyspnea of 40 days' duration and presented to the emergency department after the spontaneous expectoration of a tumor bud during a coughing episode. Chest radiography revealed an opacified left hemithorax with signs of volume loss. Thoracic CT showed a left pulmonary tumor measuring 103 × 100 × 115 mm and invading the left main bronchus, the left upper lobar pulmonary artery, and both left pulmonary veins. Histopathologic examination of the expectorated tumor fragment revealed proliferation of round cells with massive necrosis. Immunohistochemical analysis supported the diagnosis of pulmonary botryoid rhabdomyosarcoma. We present this case along with a review of the literature on pulmonary involvement in adult rhabdomyosarcoma.

Keywords: *Rhabdomyosarcoma, Pulmonary Neoplasms, Spontaneous Tumor Expulsion.*

Öz

Rabdomiyosarkom (RMS), en sık görülen yumuşak doku sarkomudur. Normal iskelet kası dokusundan gelişir ve çoğunlukla çocukları etkiler. Toraks lokalizasyonu nadirdir ve akciğer lokalizasyonu ise çok daha nadir bir durumdur. Öksürme sırasında tümör parçasının kendiliğinden dışarı atılması sonucu acil servise başvuran, 40 gündür devam eden II. derece nefes darlığı dışında asemptomatik olan 49 yaşındaki bir hastayı sunuyoruz. Akciğer röntgeninde sol hemitoraksta opak görünüm ve traksiyon bulguları görüldü. Toraks BT'sinde sol ana bronşu, sol üst pulmoner arteri ve her iki sol pulmoner veni invaze eden, bir tümör görüldü. Balgamla atılan tümör parçasının histopatolojik incelemesinde, yaygın nekrozlu yuvarlak hücre proliferasyonu vardı. İmmünohistokimyasal inceleme, akciğer Botryoid rabdomyosarkomu tanısını destekledi. Bu gözlemimiz doğrultusunda, erişkin rhabdomyosarkomunun akciğer lokalizasyonuna ilişkin literatürün gözden geçilmesini amaçladık.

Anahtar Kelimeler: *Rabdomiyosarkom, Akciğer maligniteleri, spontan tümör ekspulsiyonu.*

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Rhabdomyosarcoma (RMS) is a malignant tumor with skeletal muscle differentiation. It is the most common soft tissue sarcoma in children but remains exceedingly rare in adults, accounting for approximately 1% of adult malignancies (1). Primary pulmonary RMS is an exceptional entity, with very few cases described in the literature, most of which have been reported in pediatric populations (2). Its pathogenesis remains unclear, as the lungs do not contain native striated muscle tissue; current hypotheses suggest an origin from primitive mesenchymal cells capable of skeletal muscle differentiation (1-8).

Clinically, primary pulmonary rhabdomyosarcoma often presents with nonspecific respiratory symptoms such as cough, dyspnea, or chest pain. Imaging typically shows a pulmonary mass with possible invasion of adjacent structures, findings that are indistinguishable from those of other malignant lung tumors (1). Therefore, definitive diagnosis relies on histopathologic and immunohistochemical analysis demonstrating skeletal muscle differentiation markers such as desmin, myogenin, MyoD1, and myoglobin, which remain central to modern diagnostic criteria and WHO classification updates (8,9).

We report an unusual case of primary pulmonary RMS in an adult man, uniquely characterized by spontaneous expectoration of tumor tissue, leading to histopathologic diagnosis. Such a mode of presentation is extremely rare and has only been sporadically described in the literature (1-10).

CASE

A 49-year-old man with no medical history and no history of smoking, alcohol use, or substance use presented to the emergency department with dyspnea of one and a half months' duration, without other respiratory symptoms but with significant weight loss and profound asthenia. The night before his emergency department visit, the patient developed a dry cough, after which he expectorated tissue-like material, followed by relief of all symptoms. The expelled fragment was preserved and later sent for pathologic examination (Figure 1).

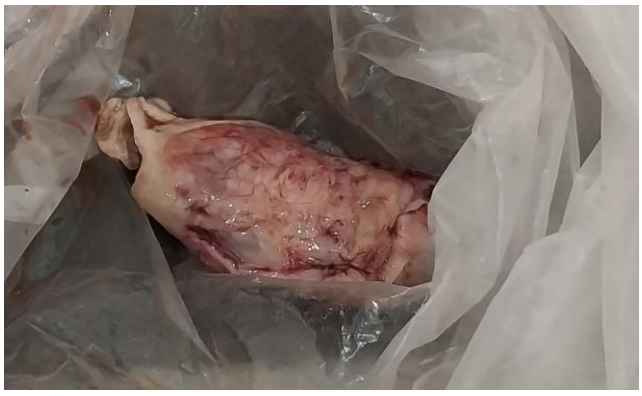

Figure 1: Expectorated tumor bud.

Physical examination revealed a conscious patient with preserved functional autonomy, normal heart and respiratory rates, and an oxygen saturation of 96%. Pulmonary examination revealed dullness to percussion, absent breath sounds, and absent vocal fremitus over the left hemithorax, consistent with a pleural effusion syndrome. The remainder of the physical examination, including lymph node examination, was normal.

Chest radiography showed an opacified left hemithorax with leftward tracheal deviation (Figure 2). CT showed a left pulmonary tumor measuring 103 × 100 × 115 mm, invading the left main bronchus, left upper lobar pulmonary artery, and both left pulmonary veins (Figure 3).

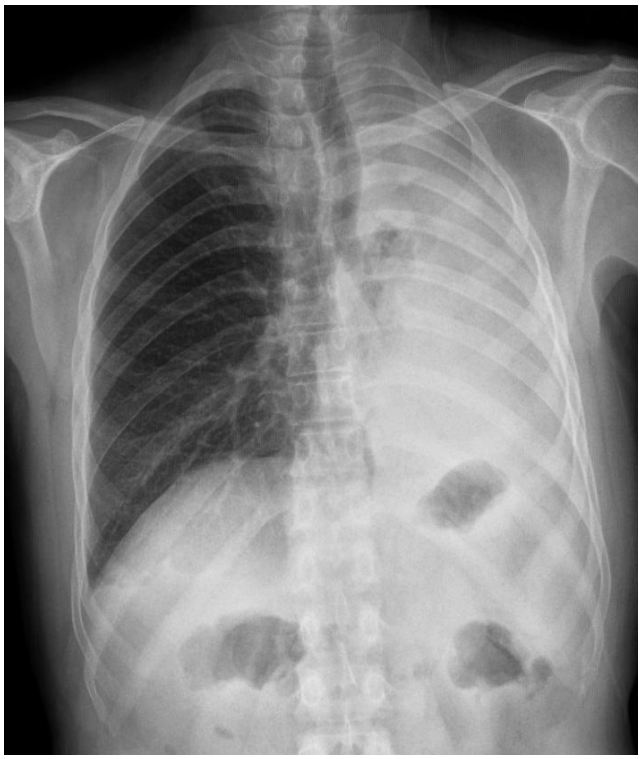

Figure 2: Chest radiograph showing an opacified left hemithorax.

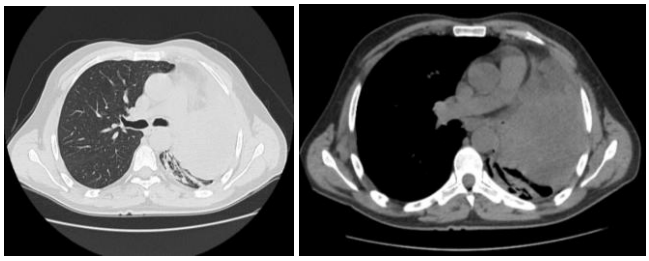

Figure 3: Chest CT scans showing tumor extension.

Histopathologic examination of the expectorated tumor revealed proliferation of round cells with massive necrosis and atypical spindle cell areas (Figure 4A). Immunohistochemical analysis showed strong cytoplasmic positivity for desmin (Figure 4B) and moderate to strong nuclear labeling for myogenin (Figure 4C), with a Ki-67 index estimat-

ed at 30%, supporting the diagnosis of pulmonary rhabdomyosarcoma.

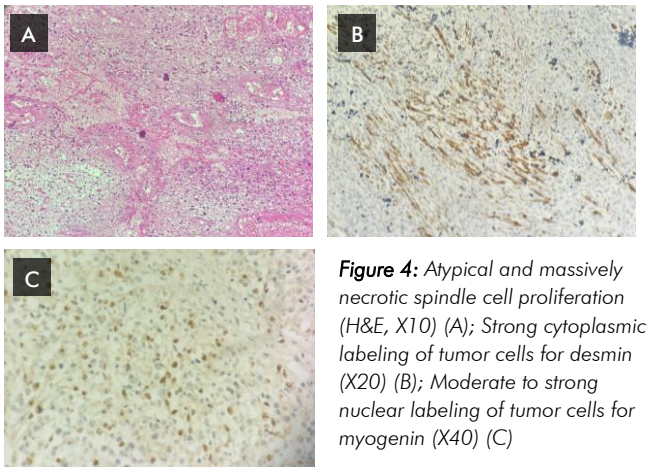

Figure 4: Atypical and massively necrotic spindle cell proliferation (H&E, X10) (A); Strong cytoplasmic labeling of tumor cells for desmin (X20) (B); Moderate to strong nuclear labeling of tumor cells for myogenin (X40) (C)

Because histopathologic examination of the expectorated tumor was conclusive, bronchoscopy was deemed unnecessary. CT scans of the head, neck, abdomen, and pelvis were performed for staging and showed no evidence of distant metastasis.

The patient was then referred to the oncology department, where the diagnosis of high-risk embryonal rhabdomyosarcoma was established. An implantable port was placed, and chemotherapy was initiated with three cycles of ifosfamide, vincristine, and actinomycin. After the first three cycles, clinical reevaluation showed persistent asthenia and decreased functional status (performance status [PS] 4 vs. PS 0 initially). Unfortunately, the patient died before radiologic reassessment could be performed to evaluate treatment response.

DISCUSSION

Rhabdomyosarcoma is a malignant skeletal muscle tumor and represents the most common type of soft tissue sarcoma in children and adolescents, accounting for approximately 7% of cancers in this population and only 1% of cancers in adults (1). No data are currently available regarding pulmonary rhabdomyosarcoma in Morocco.

Rhabdomyosarcoma typically develops in the head and neck, genitourinary tract, and retroperitoneum, making pulmonary rhabdomyosarcoma extremely rare. Fewer than several dozen cases of primary pulmonary RMS have been reported overall, most of them in pediatric populations, with adult cases remaining exceptional (1,2).

Symptoms associated with pulmonary tumors depend on tumor location, with cough and dyspnea being predominant, especially when the lesion involves the bronchial tree, as in our patient. Chest pain may also occur, particularly if the tumor is located peripherally. The absence of symptoms in the early stages often leads to diagnosis at later stages, when the tumor invades adjacent structures, such as the proximal airways or pleura, or when metastasis to extrapulmonary sites occurs (1).

The definitive diagnosis of primary pulmonary rhabdomyosarcoma is histopathologic, with two specific features being critical: cross-striations in the cytoplasm of tumor cells and evidence of striated muscle cell differentiation. However, these features may be absent in many poorly differentiated variants of rhabdomyosarcoma, emphasizing the importance of immunohistochemistry for a more reliable diagnosis (3). Common immunohistochemical markers include vimentin, desmin, myoglobin, MyoD1, and myogenin, which remain central to modern diagnostic criteria and WHO classification updates (9,10).

Rhabdomyosarcoma can be subdivided into four subtypes: embryonal, botryoid, alveolar, and pleomorphic (8). The embryonal subtype is the most common, whereas the pleomorphic subtype is associated with a worse prognosis in adults (10). In our patient, histopathologic analysis favored a diagnosis of botryoid rhabdomyosarcoma, a subtype of the embryonal category.

The therapeutic strategy for adult rhabdomyosarcoma typically involves a combination of surgery, chemotherapy, and radiotherapy (4). However, many patients, including our patient, are ineligible for surgery at the time of diagnosis. The VAC regimen (vincristine, dactinomycin, and cyclophosphamide), with or without radiation therapy, is regarded as the gold standard for rhabdomyosarcoma (5). Nevertheless, its efficacy in adults remains less well established than in pediatric populations. More recent therapeutic approaches have explored intensified chemotherapy regimens and targeted strategies, although no standardized adult-specific protocol has demonstrated clear superiority (1-10).

Recent clinical studies investigating the antitumor activity and toxicity of topotecan, both alone and in combination with conventional therapy, in patients with metastatic rhabdomyosarcoma have yielded promising results, reporting an overall response rate of 46% (complete response: 4%; partial response: 42%) (6).

Compared with children and adolescents with rhabdomyosarcoma, adults with this condition experience significantly worse outcomes, with a 5-year overall survival rate of 27% ± 1.4%, compared with 61% ± 1.4% in the pediatric population (7). Contemporary series confirm these findings, reporting survival rates ranging from 20% to 40% in adults, partly explained by delayed diagnosis, more frequent tumors in unfavorable sites, and distinct tumor biology (8-10).

Our patient's case was particularly unusual due to the spontaneous expectoration of tumor tissue, an extremely

rare mode of discovery that has only been sporadically described in the literature. This highlights the possibility of endobronchial tumor necrosis and fragmentation as a revealing mechanism in large centrally located lesions (8).

CONCLUSION

Primary pulmonary rhabdomyosarcoma in adults is an exceptionally rare and aggressive malignancy that is frequently diagnosed at advanced stages due to nonspecific clinical manifestations. This case illustrates an unusual presentation characterized by the spontaneous expectoration of a tumor bud, highlighting the need to maintain a broad differential diagnosis in patients with unexplained respiratory symptoms and atypical imaging findings. Histopathologic and immunohistochemical evaluations are crucial for achieving an accurate diagnosis, particularly in poorly differentiated tumors. Given the rarity of this condition and the limited therapeutic consensus in adults, each documented case contributes valuable insights to the existing literature. Early identification, multidisciplinary management, and ongoing documentation of such atypical presentations are essential to improve understanding, facilitate earlier diagnosis, and ultimately enhance patient outcomes.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - S.A.M., A.M.B., L.S., A.B., E.M., M.S., B.A., M.C.B., B.A., I.S., M.K.A.; Planning and Design - S.A.M., A.M.B., L.S., A.B., E.M., M.S., B.A., M.C.B., B.A., I.S., M.K.A.; Supervision - S.A.M., A.M.B., L.S., A.B., E.M., M.S., B.A., M.C.B., B.A., I.S., M.K.A.; Funding - S.A.M., A.M.B., L.S., A.B., E.M., M.S., B.A., M.C.B., B.A., I.S., M.K.A.; Materials - S.A.M., A.M.B., B.A., M.K.A., I.S., B.A.; Data Collection and/or Processing - S.A.M., A.M.B., B.A., M.K.A.; Analysis and/or Interpretation - S.A.M., A.M.B., B.A., M.K.A.; Literature Review - S.A.M., A.M.B., L.S., A.B., E.M., M.S., B.A., M.C.B., B.A., I.S., M.K.A.; Writing - S.A.M., A.M.B., B.A., M.K.A.; Critical Review - S.A.M., A.M.B., L.S., A.B., E.M., M.S., B.A., M.C.B., B.A., I.S., M.K.A.

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RESPIRATORY CASE REPORTS

An Uncommon Case of Primary Pulmonary High-Grade B-Cell Lymphoma

Nadir Bir Olgu: Yüksek Dereceli B Hücreli Primer Akciğer Lenfoması

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Abstract

Lymphoproliferative lung diseases may occur secondary to Hodgkin lymphoma (HL) or non-Hodgkin lymphoma (NHL) through hematogenous spread or invasion of the hila from mediastinal lymph nodes. In rare cases, they may present as primary pulmonary lymphoma (PPL), defined as a clonal lymphoid proliferation limited to the lungs and bronchi, with no extrapulmonary disease at diagnosis or during the subsequent three months of follow-up. The most frequent subtype is mucosa-associated lymphoid tissue (MALT) lymphoma, which arises from pulmonary mucosal lymphoid tissue. PPL typically progresses slowly and may be discovered incidentally on imaging. Radiologic findings may resemble pneumonia or lung cancer; therefore, PPL should be considered when pneumonia is unresponsive to treatment. In this case report, we describe a patient presenting with dyspnea, a central right lung lesion, mediastinal lymphadenopathy, and multiple bilateral pulmonary nodules. As bronchoscopic evaluation was non-diagnostic, transthoracic biopsy confirmed the diagnosis of PPL.

Keywords: *Non-Hodgkin lymphoma, primary pulmonary lymphoma, pneumonia with delayed resolution.*

Öz

Akciğerlerde lenfoproliferatif hastalıklar; Hodgkin Lenfoma (HL) ve Non-Hodgkin Lenfoma (NHL)'nın hematojen yayılım veya nodal lenfomaların mediastinal, hiler lenf nodlarına invazyonu ile sekonder olarak görülebilir. Nadiren, tanı anında veya takipten sonraki 3 ay içinde ekstrapulmoner hastalık olmadan akciğerler ve bronşlar ile sınırlı klonal lenfoid proflifersayon olarak tanımlanan primer pulmoner lenfoma (PPL) şeklinde görülebilir. En sık pulmoner mukozal lenfoid dokudan köken alan "mucosa-associated lymphoid tissue" (MALT) lenfoma görülür. Genellikle, sessiz ve yavaş seyirli bir hastalık olan PPL, tesadüfi olarak çekilen radyografilerde saptanmaktadır. Radyolojik olarak pnömoniyi, akciğer kanserlerini taklit edebilen, özellikle rezolüsyonu geciken pnömonilerin ayırıcı tanısında unutulmaması gereken bir hastalıktır. Bu yazıda, nefes darlığı ile başvuran ve sağ akciğer santral yerleşimli lezyon, mediastinal lenfadenopati ve bilateral multiple parankimal nodülleri saptanan bronkoskopik olarak tanımlanamayan, transtorasik biyopsi ile PPL tanısı alan bir hasta sunulmuştur.

Anahtar Kelimeler: *Non-Hodgkin lenfoma, primer pulmoner lenfoma, rezolüsyonu gecikmiş pnömoni.*

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Primary pulmonary lymphoma (PPL) is a distinctly rare entity within the spectrum of extranodal lymphoid neoplasms, accounting for approximately 0.3% of all primary lung malignancies. A definitive diagnosis requires the exclusion of extrapulmonary involvement through comprehensive clinical, radiologic, and histopathologic assessment at presentation and over a follow-up period of at least three months. It is characterized by a clonal malignant lymphoproliferative process localized to one or both lungs and/or the bronchial tree. Although 40% of patients are asymptomatic, symptomatic patients may present with chest pain, cough, dyspnea, and hemoptysis (1). By contrast, pulmonary involvement secondary to systemic lymphoma is more common. Pulmonary involvement in the form of hematogenous spread or direct invasion is seen in 38% of Hodgkin lymphoma (HL) cases and 24% of non-Hodgkin lymphoma (NHL) cases.

Radiological findings include mass lesions, nodular infiltrations, and consolidations with delayed resolution; cavitary lesions may also be seen in diffuse large B-cell lymphoma (DLBCL) (2).

PPL is extremely rare and may present asymptomatically or with nonspecific respiratory symptoms. Consequently, it should be considered in the differential diagnosis of various pathologic conditions.

This case is presented in the context of the existing literature to highlight the clinical and diagnostic features of primary pulmonary DLBCL, a notably rare entity.

CASE

A 76-year-old woman with a history of essential hypertension, diabetes mellitus, and previous tuberculosis presented with dyspnea. She was regularly taking metformin, acetylsalicylic acid, olmesartan, linagliptin, pantoprazole, spironolactone, and hydrochlorothiazide. She had no history of smoking. There were no specific findings on physical examination. Laboratory tests showed an elevated LDH level of 445. A chest computed tomography (CT) scan was performed because her chest radiograph showed nodular infiltrations in the right hilar region and lung parenchyma. The chest CT scan showed multiple bilateral nodules (Figures 1a and 1b). Positron emission tomography/computed tomography (PET/CT), performed with a preliminary diagnosis of a primary lung malignancy with metastases, showed a central mass lesion in the right lung. Pathologic FDG uptake was observed in the hilar (SUVmax: 21.0) and mediastinal (SUVmax: 20.0) lymph nodes and in multiple parenchymal nodules (SUVmax: 15.0) in both lungs (Figure 2).

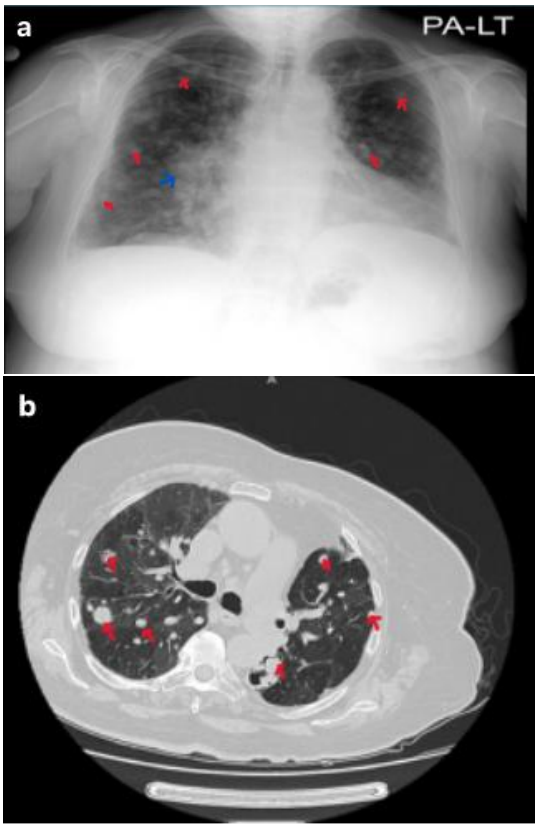

Figure 1: Right hilar enlargement and nodular infiltrative opacities in both lungs on the patient's PA chest radiograph (a); parenchymal nodules on chest CT image (b).

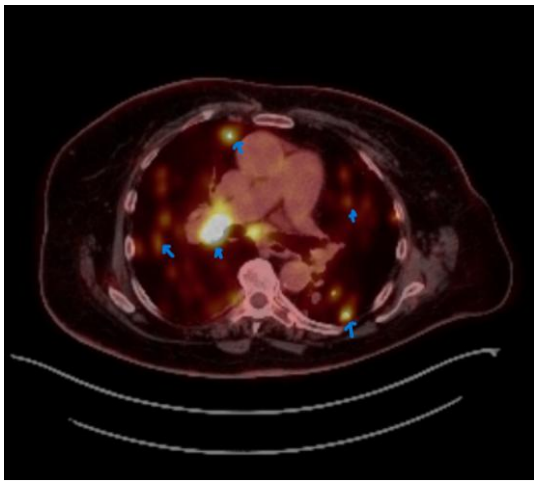

Figure 2: PET/CT image showing a centrally located mass in the right lung, multiple hypermetabolic nodules in both lungs, and hypermetabolic mediastinal lymph nodes.

Flexible bronchoscopy revealed no abnormal findings. A tru-cut biopsy was performed on one of the larger peripheral nodules in the right lung (TTNAB). Pathology revealed CD20 positivity, and the lesion was reported as "high-grade B-cell lymphoma" (Figure 3). The patient was evaluated by the hematology department and staged as Stage 4S. Chemotherapy with R-mini-CHOP—rituximab in combination with reduced-dose cyclophosphamide, doxorubicin (previously known as hydroxydaunorubicin),

vincristine (marketed as Oncovin), and prednisone—was planned because of the patient's deteriorating general condition and comorbidities. The patient's treatment and follow-up were continued by the hematology clinic.

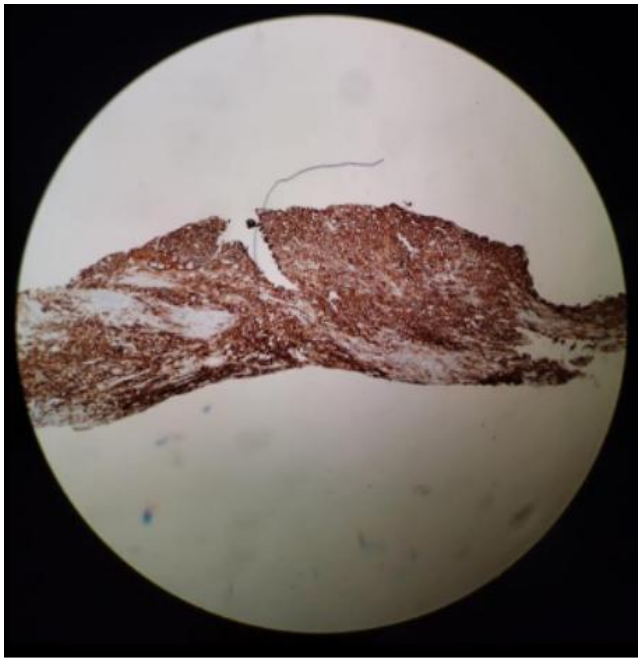

Figure 3: Immunohistochemical study showing strong CD20 positivity (×100).

Informed consent was obtained from the patient for the case report.

DISCUSSION

Primary pulmonary lymphoma (PPL) is a rare tumor, accounting for only 0.5% of all lung tumors (3). The incidence of PPL peaks in two age groups: first in the fifth decade and again in the late sixth and seventh decades (4). Our case was a 76-year-old woman. Surface markers play an important role in lymphoma typing. In B-cell lymphomas, the presence of CD5 and CD10 markers is significant. Our case showed strong CD20 positivity and was diagnosed as high-grade B-cell PPL.

In the differential diagnosis of primary pulmonary lymphoma, conditions such as pneumonia, lung cancer, metastasis, atelectasis, and pulmonary sequestration should be considered. Histopathologic evaluation is essential for a definitive diagnosis (3). In primary pulmonary lymphoma, the contribution of bronchoscopy to diagnosis is limited because there is usually no endobronchial lesion. Rarely, the diagnosis can be made by cytogenetic methods using bronchoalveolar lavage fluid (5). Therefore, VATS or open surgical intervention is usually required (3). In our case, cytologic evaluation of the specimens obtained by flexible bronchoscopy was benign, and the diagnosis was made by TTNAB.

The histopathologic type of the tumor is the most important prognostic factor in PPL cases. PPL can be classified into three subtypes. In 70% of PPL cases, the tumor is a low-grade B-cell MALT lymphoma arising from lymphoid tissue in the bronchial mucosa. The second subtype is diffuse large B-cell lymphoma (DLBCL), which occurs in 12% of cases. In half of DLBCL cases, an underlying MALT lymphoma develops into a high-grade lymphoma. Finally, lymphomatoid granulomatosis (LG) is a lymphoproliferative disease caused by Epstein-Barr virus (EBV)-infected large B cells that begins around the vessels and leads to tissue damage (6).

Poor prognostic factors associated with the disease include age ≥60 years, elevated serum lactate dehydrogenase (LDH) levels, an Eastern Cooperative Oncology Group (ECOG) performance status of 2–4, advanced-stage disease (Stage III/IV), and the presence of extranodal involvement (7,8). Treatment strategies are guided by the patient's prognostic profile and overall clinical condition.

The treatment of primary pulmonary lymphoma is controversial. Complete surgical resection is preferred in localized cases. Due to its typically indolent course, active surveillance with clinical follow-up is recommended over surgical intervention. While combined chemotherapy is preferred in cases of progression, recurrence, or widespread involvement, radiotherapy is rarely used (3,9). Our patient was evaluated by the hematology department and initiated on rituximab in combination with a reduced-dose CHOP regimen—cyclophosphamide, doxorubicin (previously known as hydroxydaunorubicin), vincristine (marketed as Oncovin), and prednisone—referred to as R-mini-CHOP.

CONCLUSION

Despite its rarity, primary pulmonary lymphoma should be considered as a potential diagnosis in patients with multiple nodular pulmonary lesions.

Chemotherapy-related lung involvement is a serious complication observed during treatment and can significantly impact the quality of life of patients undergoing.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - K.U.E., N.Ö., İ.S.K., S.Ö., C.Y., S.Ö., Ü.Ş.; Planning and Design - K.U.E., N.Ö., İ.S.K., S.Ö., C.Y., S.Ö., Ü.Ş.; Supervision - K.U.E., N.Ö., İ.S.K., S.Ö., C.Y., S.Ö., Ü.Ş.; Funding - N.Ö., K.U.E., İ.S.K., S.Ö.; Materials -K.U.E., N.Ö., C.Y., S.Ö.; Data Collection and/or

Processing - N.Ö., K.U.E., İ.S.K., S.Ö.; Analysis and/or Interpretation - N.Ö. İ.S.K.; Literature Review - K.U.E.; Writing - K.U.E., N.Ö.; Critical Review - S.Ö., Ü.Ş.

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RESPIRATORY CASE REPORTS

Fatal Disseminated Mucormycosis Following Exertional Heat Stroke: A Case Report and Diagnostic Challenge

Egzersizle Oluşan Sıcak Çarpması Sonrası Ölümcül Yaygın Mukormikoz: Olgu Sunumu ve Tanısal Zorluk

Shengquan Wei

Abstract

Mucormycosis is an uncommon but life-threatening fungal infection typically seen in immunocompromised hosts. It has rarely been reported following exertional heat stroke (EHS). We describe a fatal case of rhino-orbital-cerebral and pulmonary mucormycosis in a 59-year-old man recovering from EHS. Despite initial stabilization, he developed necrotic facial lesions and respiratory failure. Mucor spp were isolated from tissue and bronchoalveolar lavage (BAL) cultures. He died on day 14 despite liposomal amphotericin B therapy. This rare case of mucormycosis following EHS highlights the potential role of heat-induced immune dysfunction in predisposing patients to opportunistic fungal infections. Clinicians should maintain a high index of suspicion for mucormycosis in EHS patients with unexplained fever or tissue necrosis.

Keywords: *Exertional Heat Stroke (EHS), Multi-Organ Failure, Mucormycosis.*

Öz

Mukormikoz, bağışıklık sistemi baskılanmış konaklarda tipik olarak görülen ve nadiren yaşamı tehdit eden bir mantar enfeksiyonudur. Egzersizle oluşan sıcak çarpması (ESÇ) sonrası nadiren bildirilmiştir. Elli dokuz yaşındaki, ESÇ'den kurtulan bir erkekte gelişen ölümcül rinomikso-orbital-serebral ve pulmoner mukormikoz olgusu anlatılmaktadır. Başlangıçta stabilizasyon sağlandıktan sonra, hastada nekrotik yüz lezyonları ve solunum yetmezliği gelişti. Doku ve BAL kültürleri ile Mucor spp. teyit edildi. Lipozomal amfoterisin B tedavisine rağmen hasta 14. günde öldü. ESÇ sonrası gelişen bu nadir bildirilen mukormikoz olgusu, ısı kaynaklı bağışıklık baskılanması hasarının fırsatçı mantar enfeksiyonlarına eğilim yaratma potansiyelini vurgulamaktadır. Klinisyenler, açıklanamayan ateş veya doku nekrozu olan ESÇ hastalarında mukormikoz için yüksek şüphe duymalıdırlar.

Anahtar Kelimeler: *Egzersizle oluşan sıcak çarpması (ESÇ), çoklu organ yetmezliği, mukormikoz.*

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Exertional heat stroke (EHS) is a medical emergency characterized by central nervous system (CNS) dysfunction and multi-organ failure following exposure to high ambient temperatures during physical exertion (1,2). It induces a systemic inflammatory response and can cause significant immune dysregulation, thereby increasing susceptibility to infections. Mucormycosis is an aggressive angioinvasive fungal infection caused by organisms of the order Mucorales. Although it is typically seen in patients with diabetes or immunocompromised individuals (3,4), only a few cases of EHS-associated mucormycosis have been documented to date. We report a case of fatal rhino-orbital-cerebral and pulmonary mucormycosis complicating the recovery phase of EHS.

CASE

A 59-year-old male agricultural worker engaged in heavy physical labor, with no significant medical history (including diabetes mellitus, hypertension, or immunosuppressive conditions), was found unconscious after working in a high-temperature environment. On admission to the emergency department (day 0), he was comatose (Glasgow Coma Scale score of 3 [GCS 3]) with a core temperature >42°C, tachypnea, and tachycardia. Initial laboratory tests revealed metabolic acidosis, acute kidney injury, elevated creatine kinase (623 U/L), elevated transaminases (AST 154 U/L), coagulopathy (PT 21.3 s, platelets 91 $\times$ 10^9/L), and an HbA1c level of 5.4%. Initial chest radiography showed bilateral pulmonary infiltrates (Figure 1). A diagnosis of EHS with multiple organ dysfunction syndrome (MODS; respiratory, hepatic, renal, and hematologic involvement) and aspiration pneumonia was made.

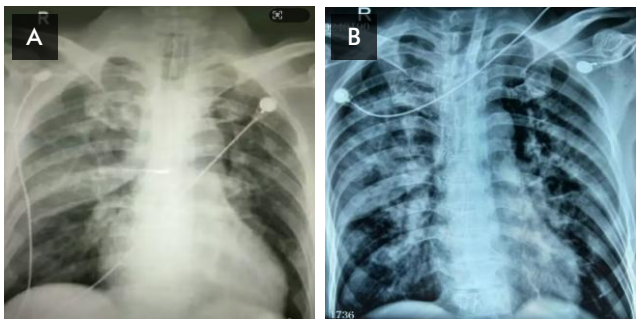

Figure 1: *Chest radiographs demonstrating bilateral pulmonary infiltrates: admission radiograph (A) and radiograph on hospital day 7 (B).*

He was intubated, cooled, and received supportive care, including vasopressors, renal replacement therapy, and multiple transfusions of fresh frozen plasma and platelets for worsening disseminated intravascular coagulation (DIC). His neurologic status gradually improved, and he was extubated on day 7.

However, from day 8 onward, he developed intermittent high-grade fevers (maximum temperature, 39.4°C). Physical examination revealed well-demarcated black necrotic eschars involving the nasal bridge, upper lip, hard palate, and oral mucosa (Figure 2).

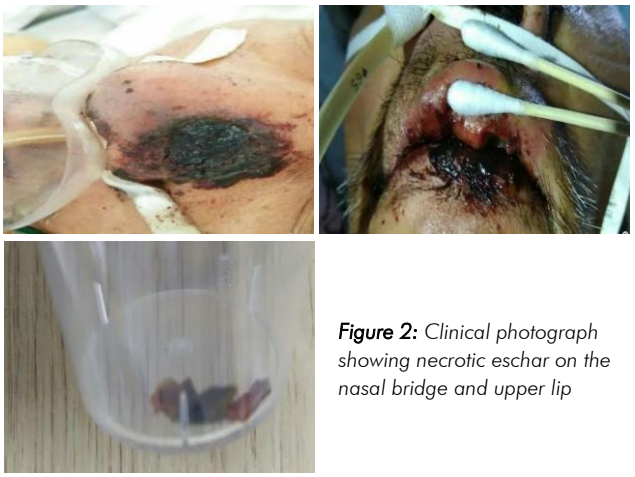

Figure 2: *Clinical photograph showing necrotic eschar on the nasal bridge and upper lip*

An urgent otorhinolaryngology consultation was obtained. Bronchoalveolar lavage (BAL) was performed. Bronchoscopy showed necrotic material and blackish secretions in the airways. The BAL fluid appeared dark and sedimented, suggestive of hemorrhagic and inflammatory material (Figure 3). A biopsy of the necrotic facial lesion was performed, and direct microscopic analysis of the BAL fluid with potassium hydroxide (KOH) wet mount was conducted. Microscopy of nasal scrapings and BAL fluid revealed broad, ribbon-like, pauciseptate hyphae consistent with mucormycetes.

Figure 3: *Bronchoalveolar lavage (BAL) fluid specimen showing a hemorrhagic appearance.*

Culture confirmed *Mucor* species (*Rhizopus oryzae*, as shown in Figure 4). Given that surgical debridement represents the definitive therapy for mucormycosis, urgent multidisciplinary consultation was undertaken. Objective contraindications were identified: refractory hemodynamic instability despite maximal vasopressor support, a SOFA score of 14, and severe thrombocytopenia (platelets 18 $\times$ 10^9/L). After careful deliberation among the family, anesthesiologists, and surgeons regarding the prohibitive operative risks versus the limited anticipated survival benefit, the decision was made to forgo surgical intervention and pursue medical management alone.

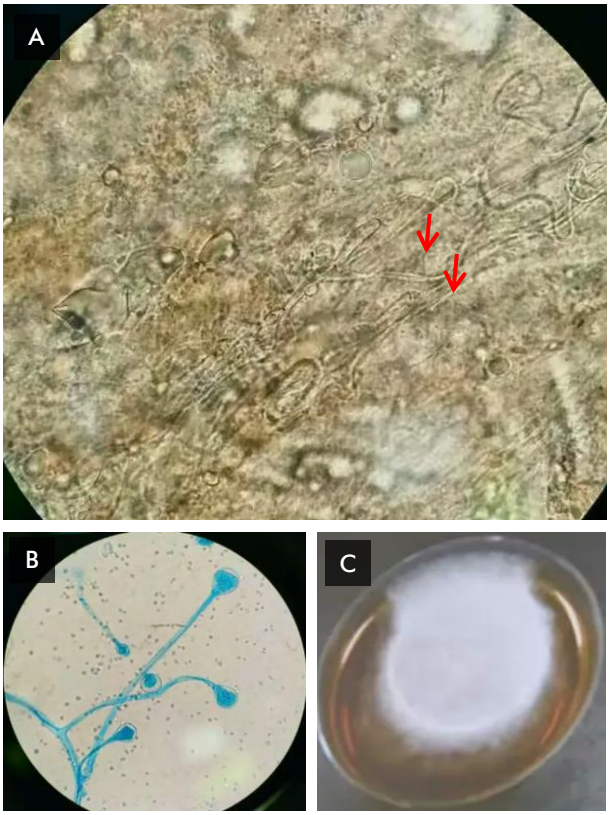

Figure 4: Microscopy and culture demonstrating features of Mucorales: nasal tissue showing broad, pauciseptate hyphae (KOH wet mount, 40X) (A); BAL fluid showing hyphae (cotton blue stain, 40X) (B); and Sabouraud dextrose agar culture after 5 days showing fluffy colonies of Rhizopus oryzae (C).

Despite the immediate initiation of intravenous liposomal amphotericin B (5 mg/kg/day, gradually titrated upward) and thymopolypeptides (10 mg IM once daily), the infection progressed rapidly. Follow-up imaging suggested rhinocerebral involvement and worsening pulmonary disease. He developed recurrent septic shock with worsening MODS. His condition deteriorated irreversibly, and he succumbed to the infection on day 14 after his family elected compassionate care because of the poor prognosis (Table 1).

DISCUSSION

This report describes a rare but devastating case of disseminated mucormycosis complicating recovery from EHS. A prior case report described gastric mucormycosis after heat stroke that was successfully treated with timely endoscopic diagnosis and surgical debridement (5). By contrast, our patient developed rapid rhino-orbital-cerebral and pulmonary dissemination, with limited opportunity for localized surgical control, underscoring how the site of infection and timing of diagnosis may affect outcome (Table 2).

Severe EHS may create a milieu of immune dysfunction that could lower host resistance to opportunistic fungi. EHS is known to produce hyperthermia-mediated tissue injury, systemic inflammation, and endothelial damage, followed by a compensatory anti-inflammatory phase that some investigators have termed "immunoparalysis," characterized by lymphopenia and reduced cellular immune function. Concurrent disruption of endothelial and mucosal barriers might theoretically facilitate angioinvasion by Mucorales organisms.

In our patient, we observed persistent lymphopenia (absolute lymphocyte count [ALC] consistently $<0.6 \times 10^9$/L) and elevated IL-10 levels, which are surrogate markers that have been associated with immune dysregulation in other critical illness contexts. However, it is crucial to emphasize that these findings represent correlations rather than definitive evidence of causation. The temporal sequence, with lesions appearing on day 8, is consistent with a post-EHS opportunistic infection, but we cannot

Table 1: Clinical Timeline, Laboratory Data, and Immunological Markers (Corticosteroids were not administered during the treatment course)

Day	Key clinical events/interventions	Key labs/immune markers (value, day)	Imaging / pathology	Antimicrobials / other interventions	Remarks
0	Collapse after ~5 h work core temp >42□C; intubation & cooling initiated	CK 623 U/L; Cr 312 μmol/L PCT >100 ng/mL ALC 0.5,HbA1c 5.4%	CXR: diffuse infiltrates	Intubation, vasopressor (NE), CRRT started	Initial Dx: EHS + MODS
1	–	IL-6 1842 pg/mL; IL-10 456 pg/mL	–	FFP/PLT for DIC	
3	–	ALC 0.4; PCT 86 ng/mL	–	–	
7	Extubated	ALC 0.6	–	—	
8	New fever; facial pain; black eschar on nasal bridge/upper lip/hard palate	ALC 0.5→0.3next days	CT chest: cavitation	Surgical consult requested	First suspicion of invasive fungal infection
9	Bronchoscopy: necrotic debris & black secretions; BAL obtained; nasal biopsy	KOH wet mount: broad aseptate hyphae (Mucorales)	Histopath: angioinvasion (pending)	Liposomal Amphotericin B started (5 mg/kg/day)	Bronchoscope had no photo function (no endoscopic images)
10	Rapid deterioration; refractory shock	IL-10 215 pg/mL IL-6 812 pg/mL ALC 0.3	Culture later grew Mucor sp.	Ongoing amphotericin B; surgical debridement considered but not feasible	CNS involvement suspected
14	Death	SOFA up to 16	—	Support withdrawn per family wishes	

Abbreviations: SOFA (Sequential Organ Failure Assessment), IL-10 (Interleukin-10), KOH (Potassium Hydroxide,), ALC (Absolute Lymphocyte Count), PCT (Procalcitonin), CNS (Central Nervous System), CK (Creatine Kinase), IL-6 (Interleukin-6), Cr (Creatinine)

Table 2: Comparative analysis of reported cases of mucormycosis following exertional heat stroke (EHS)

Reference	Age/Sex	Type of Heat Stroke	Underlying Conditions	Site of Mucormycosis	Time to Diagnosis (Days post-EHS)	Key Diagnostic Modalities	Antifungal Therapy	Surgical Intervention	Outcome	Key Features & Immunological Markers
Current case	59-year-old male	EHS (agricultural work, >5h, core temp >42□C)	No significant history; HbA1c 5.4%; no diabetes or immunosuppression	Disseminated: rhino-orbital-cerebral + pulmonary	Day 8 (lesion onset) → Day 9 (confirmed)	Facial biopsy, BAL (KOH/culture), CT chest	Liposomal amphotericin B (5 mg/kg/day) + Thymopolypeptides (10 mg IM daily)	Not feasible (SOFA 14, refractory shock, platelets 18□10⁹/L)	Death (Day 14)	Persistent lymphopenia (ALC <0.6□10⁹/L), elevated IL-10 (peak 456 pg/mL), inverted IL-6/IL-10 ratio; no traditional risk factors
Marco et al., 2023	53-year-old male	EHS (warehouse work, axillary temp 41□C)	Well-controlled schizophrenia (quetiapine); no immunosuppression	Gastric (localized)	Day 12 (fever onset) → Day ~19 (endoscopic biopsy)	Gastroscopy with biopsy, CT abdomen, Culture (Rhizopus microsporus)	Liposomal amphotericin B (10 mg/kg/day) + Isavuconazole → Posaconazole (maintenance)	Laparoscopic radical gastrectomy (converted to open due to contained perforation)	Survived (discharged after 2 months)	Transient hyperglycemia from steroids/glucose; mucosal barrier disruption from intestinal ischemia; prompt diagnosis enabled surgery
Literature summary	(n=2 reported cases)	Both exertional	No traditional immunosuppression	Variable (gastric vs. disseminated)	8-19 days	Endoscopic/tissue biopsy cornerstone	Amphotericin B-based regimen standard	Surgery feasible in localized disease	Mortality 50% (1/2)	Immunoparalysis hypothesized but not directly measured in either case; resource limitations prevented HLA-DR testing

(Gastric mucormycosis presenting as fever of unknown origin in an immunocompetent host after heatstroke, References(5))

exclude the possibility that other unmeasured factors, such as occult environmental exposure, subtle genetic predisposition, or the synergistic effects of multi-organ dysfunction itself, may have contributed. The available immune markers are, at best, suggestive and cannot prove that immunoparalysis directly caused this infection.

EHS induces hyperthermia-driven tissue injury, overwhelming systemic inflammation, and endothelial damage. The subsequent compensatory anti-inflammatory response may culminate in immunoparalysis, characterized by lymphopenia, impaired antigen presentation, and reduced cellular immune function. Concurrent disruption of endothelial and mucosal barriers may facilitate angioinvasion by Mucorales (6-8). The angioinvasive properties of Mucorales species play a crucial role, as these organisms secrete proteases and elastases that directly damage the vascular endothelium, initiating thrombosis and tissue necrosis (9).

In our patient, persistent lymphopenia (absolute lymphocyte count consistently $<0.6 \times 10^9$/L), markedly elevated anti-inflammatory cytokine IL-10 levels (peak, 456 pg/mL), and dynamic IL-6/IL-10 profiles were consistent with profound post-EHS immune dysregulation. Although monocyte HLA-DR expression and functional T-cell assays were not available (see Limitations), the clinical course and available immune markers strongly suggest clinically significant immunosuppression that likely predisposed the patient to invasive mucormycosis.

Recognition of immunoparalysis after critical illness has led to increasing interest in immune-restorative strategies. Reported approaches include cytokine-based therapies (e.g., interferon-γ to enhance macrophage and monocyte function), colony-stimulating factors (granulocyte-macrophage colony-stimulating factor [GM-CSF] and granulocyte colony-stimulating factor [G-CSF]) to support myeloid recovery, interleukin-7 to restore lymphocyte counts and function, and immune checkpoint modulation in selected settings. Thymic peptides, such as thymosin alpha-1 and related formulations, have also been used as immunomodulatory adjuncts in sepsis and some invasive infections (10), although high-quality evidence specifically for mucormycosis remains limited.

In this case, we administered thymopolypeptides (10 mg intramuscularly once daily) as an immune-supportive intervention based on clinical judgment and local availability, recognizing the low level and indirect nature of the supporting evidence. At present, no immune-restorative therapy has been conclusively shown to improve outcomes in disseminated mucormycosis, and such interventions should be individualized and, when possible, investigated within research protocols.

Early recognition of invasive fungal infection and prompt combined medical-surgical management are central to improving outcomes in mucormycosis. Liposomal amphotericin B remains the first-line antifungal agent, with posaconazole or isavuconazole typically reserved as salvage or step-down therapy. However, in disseminated disease with extensive angioinvasion, antifungal therapy alone is often insufficient when surgical debridement is not feasible. In our patient, severe hemodynamic instability and extensive disseminated involvement precluded aggressive surgical intervention, which likely contributed to the poor outcome.

Limitations

This report has several limitations. First, bronchoscopic photographic documentation was unavailable. The fiberoptic bronchoscope used (Olympus BF-XT40) is a legacy model lacking integrated digital image capture. Furthermore, the external display monitor malfunctioned and could not be repaired because replacement components for this discontinued system are no longer commercially available. Instead, we provide gross BAL fluid images and microbiologic/pathologic findings (Figures 3 and 4), and include a photograph of the bronchoscope as Supplementary Figure illustrate this technical constraint. Second, comprehensive immune functional testing was incomplete. Monocyte HLA-DR expression and detailed T-cell functional assays were not performed because of local resource limitations and patient financial constraints. Consequently, we relied on serial lymphocyte counts and cytokine profiles as surrogate indicators of immune status, and we have clearly stated which assays were not available. Third, this is a single case report, and a causal relationship between EHS and mucormycosis cannot be definitively established. Nonetheless, the temporal association, biological plausibility, and supporting literature suggest that post-EHS immune dysregulation may have contributed to the development of disseminated mucormycosis, warranting heightened clinical awareness and further study.

Supplementary Figure: *The fiberoptic bronchoscope employed (Olympus BF XT40) is a legacy model lacking integrated digital image capture. Furthermore, the external display monitor malfunctioned and could not be repaired, as replacement components for this discontinued system are no longer commercially available.*

CONCLUSION

This case illustrates that survivors of exertional heat stroke (EHS) may develop profound immune dysregulation that predisposes them to opportunistic angioinvasive fungal infections such as mucormycosis, even in the absence of traditional risk factors. Clinicians should maintain a high index of suspicion for invasive fungal disease in EHS patients who develop new tissue necrosis, persistent fever, or otherwise unexplained clinical deterioration, and should promptly perform tissue sampling and targeted fungal diagnostics. Careful assessment of immune status and, when appropriate, multidisciplinary discussion regarding the feasibility of surgical debridement and the potential role of adjunctive immune-restorative strategies are essential in managing these complex cases.

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Heartfelt thanks to Director Li Haiying of Microbiological Testing for her efforts in the microbiological testing of this case.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - S.W.; Planning and Design - S.W.; Supervision - S.W.; Funding - S.W.; Materials - S.W.; Data Collection and/or Processing - S.W.; Analysis and/or Interpretation - S.W.; Literature Review - S.W.; Writing - S.W.; Critical Review - S.W.

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RESPIRATORY CASE REPORTS

Pulmonary *Mycobacterium simiae* Infection in an Immunocompetent Patient

İmmünkompetan Hastada Pulmoner Mycobacterium Simiae İnfeksiyonu

Sena Melis Sert[1], Emine Afşin[2]

Abstract

Mycobacterium simiae is a slow-growing nontuberculous mycobacterial species with intrinsic resistance to conventional antituberculosis agents and is capable of causing pulmonary and/or extrapulmonary infections. The incidence and severity of disease are increased particularly in immunosuppressed individuals. In this young incarcerated patient, during the etiologic evaluation of pneumonia, sputum smears were negative for acid-fast bacilli; however, M. simiae was isolated from two separate sputum mycobacterial cultures. The patient demonstrated clinical and radiologic improvement with nonspecific antibiotic therapy.

Keywords: *Mycobacterium simiae, immunocompetent, nontuberculous mycobacteria.*

Öz

Mycobacterium simiae; geleneksel anti-tüberküloz ajanlara dirençli, yavaş büyüyen ve pulmoner ve/veya ekstrapulmoner enfeksiyonlara neden olabilen bir tüberküloz dışı mikobakteri türüdür. Özellikle immunsuprese bireylerde hastalığın görülme sıklığı ve şiddeti artmaktadır. Genç mahkum hastamızda pnömoni etkenleri araştırılırken balgamda asidorezistan basil negatif ancak iki ayrı balgam mikobakteri kültüründe M. simiae üremesi saptandı. Hastada nonspesifik antibiyotik tedavisi ile klinik ve radyolojik yanıt alındı.

Anahtar Kelimeler: *Mikobakteri simiae, immünkompetan, tüberküloz dışı mikobakteri.*

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Nontuberculous mycobacteria (NTM) comprise more than 190 species and subspecies, some of which can cause disease in individuals of all ages and may involve both pulmonary and extrapulmonary sites (1). Population-based studies have documented a steady increase in the prevalence of NTM since 2000 (2). Clinically, the most frequently isolated NTM pathogens include the slowly growing Mycobacterium avium complex, which encompasses multiple subspecies, and the slow-growing *M. abscessus* complex. *M. kansasii, M. simiae*, and *M. fortuitum* represent other less commonly isolated species (3). *M. simiae* infections can lead to severe clinical manifestations, particularly in immunocompromised individuals, and may rarely be detected in immunocompetent patients. However, isolation of *M. simiae* from respiratory specimens does not always indicate true infection. Here, we present a case of an immunocompetent patient in whom *M. simiae* was isolated during the etiologic evaluation of cavitary pneumonia (4).

CASE

A 22-year-old incarcerated male patient presented with a two-month history of cough, purulent sputum, loss of appetite, and 5-kg weight loss. He reported having experienced fever one week before presentation, which did not recur. He denied dyspnea, night sweats, or hemoptysis. The patient had no known comorbidities but had a 7-pack-year smoking history. He had been incarcerated for approximately one year and had no history of tuberculosis (TB), no contact with anyone with TB, and no history of illicit drug use. He did not appear cachectic. On physical examination, oxygen saturation was 98% on room air, and breath sounds were normal. A chest radiograph obtained at an outside facility three weeks earlier demonstrated a peripheral cavitary lesion with surrounding increased density in the lower zone of the left lung (Figure 1A and B).

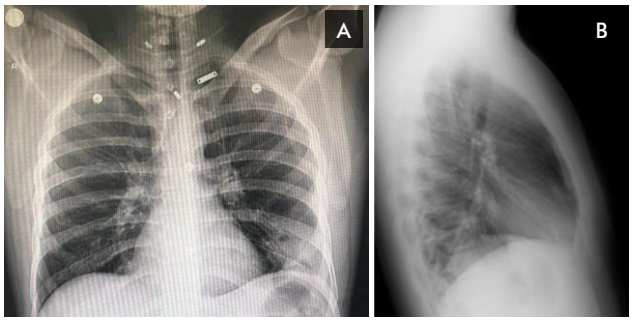

Figure 1: *Chest radiograph showing a cavitary opacity in the left lower peripheral lung zone (A,B)*

Thoracic computed tomography (CT) revealed pneumonic consolidation in the superior segment of the left lower lobe, broadly abutting the pleura and containing cystic cavitation. In addition, bilateral, predominantly right middle lobe, ground-glass opacities and a subtle tree-in-bud pattern were noted (Figure 2). CRP was 4.2 mg/L, the white blood cell count was 7.9 K/μL, renal and hepatic function tests were within normal limits, and HBsAg, anti-HCV, and anti-HIV tests were negative.

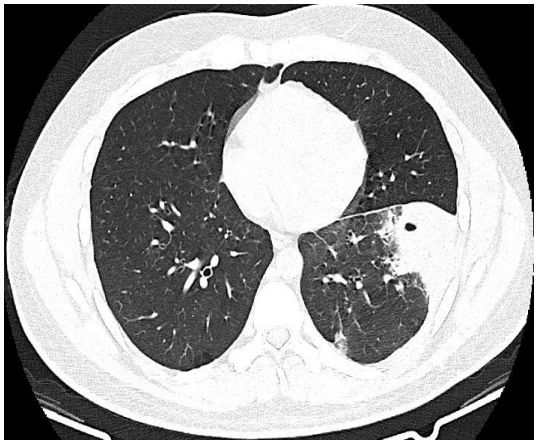

Figure 2: *Chest computed tomography showing a consolidative area with cavitation in the superior segment of the left lower lobe*

Sputum samples were obtained for acid-fast bacilli (AFB) smear, mycobacterial culture, and routine bacterial culture, and the patient was initiated on a cephalosporin-class antibiotic. At the one-week follow-up, two AFB smears were negative, and routine sputum culture showed no growth. Mycobacterial culture, however, revealed growth of NTM, with species identification pending. The patient's clinical symptoms improved, laboratory values remained normal, and follow-up chest radiography showed resolution of the cavitary lesion, with a residual linear density compared with the previous radiograph (Figure 3).

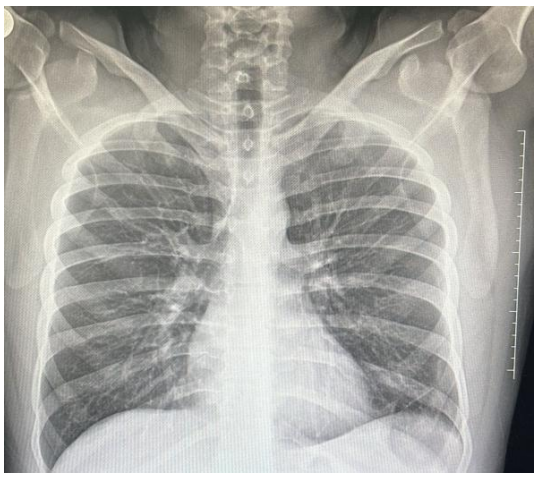

Figure 3: *Chest radiograph showing a linear density in the left lower lung zone*

Repeat thoracic CT demonstrated near-complete resolution of the cavitary lesion and surrounding consolidation in the left lung (Figure 4). Given the clinical and radiologic improvement, the patient was followed without NTM-specific antimicrobial therapy. Species identification from

the mycobacterial cultures later confirmed the isolate as *Mycobacterium simiae*. The patient remained asymptomatic during the two-month follow-up period, and no growth was observed in the follow-up mycobacterial cultures.

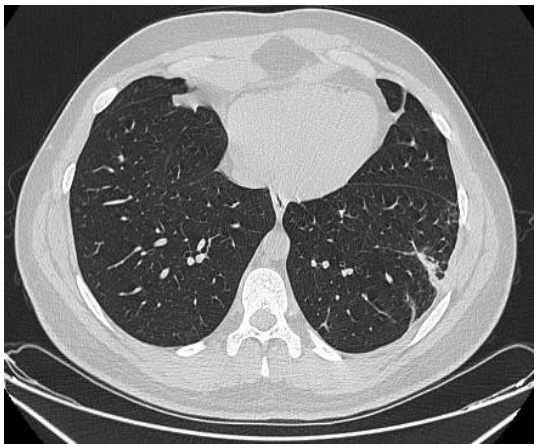

Figure 4: *Chest computed tomography showing linear opacity in the left lower lobe*

DISCUSSION

Mycobacterium simiae is an NTM species that can cause pulmonary infections in many countries worldwide (5). In Türkiye, between 2014 and 2023, 66 of 171 NTM isolates obtained at the Dicle University Hospital Mycobacteriology Laboratory were identified at the species level; the most frequently isolated species were *M. simiae* (28.78%), Mycobacterium avium complex (21.21%), and M. abscessus complex (13.63%) (6). This organism has been isolated not only from human and animal specimens but also from various environmental reservoirs, such as shower water, spring water, and tap water (7). Given that our patient was incarcerated, environmental conditions may have contributed to the acquisition of this isolate. Previous reports indicate that *M. simiae* may lead to a broad spectrum of clinical presentations, ranging from asymptomatic infection to disseminated disease with a potentially fatal course (6). Our patient presented with pneumonia and showed clinical improvement with non-specific antibiotic therapy. Immunocompromised individuals, patients with underlying medical conditions, and patients with a history of TB are the groups most susceptible to *M. simiae* infections (8). The favorable clinical response may be partly explained by the patient's young age and absence of major risk factors. There is also evidence in the literature that, although rare, *M. simiae* infection can occur in immunocompetent patients and may not always require additional antimicrobial therapy (9).

In the study by Song et al. (10), bronchiectasis was observed in 98% of cases and cavitation in 36%. Previous reports have noted that CT findings are generally described for NTM infections overall rather than as species-specific features. In our case, cavitation accompanied by consolidation was observed. The optimal management of *M. simiae* infections remains controversial, and no standardized treatment protocol currently exists (11,12). These infections are characterized by widespread resistance to first-line antituberculosis drugs; therefore, treatment regimens differ from classical tuberculosis protocols and may include agents such as moxifloxacin, clarithromycin, and trimethoprim-sulfamethoxazole (13). In our case, treatment for presumed NTM infection was deferred until species identification was available. Once *M. simiae* was confirmed, the patient was managed with observation because of both the absence of a standardized regimen and his favorable clinical condition. Based on our patient's course, treatment may be deferred in asymptomatic or mildly symptomatic cases.

CONCLUSION

M. simiae should be considered a potential pathogen in pulmonary diseases, including infections in immunocompetent individuals. Its potential role in a wide spectrum of clinical manifestations across various hosts should be recognized, and treatment decisions should be individualized based on the patient's clinical context.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - S.M.S., E.A.; Planning and Design - S.M.S., E.A.; Supervision - E.A., S.M.S.; Funding - S.M.S., E.A.; Materials - S.M.S.; Data Collection and/or Processing - S.M.S., E.A.; Analysis and/or Interpretation - S.M.S., E.A.; Literature Review - S.M.S.; Writing - S.M.S., E.A.; Critical Review - S.M.S., E.A.

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Endobronchial Tuberculosis: A Case Report

Endobronşial Tüberküloz: Olgu sunumu

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Abstract

Endobronchial tuberculosis (EBTB) is a rare form of tuberculosis. Although its clinical, laboratory, microbiologic, and treatment characteristics are similar to those of other forms of tuberculosis, EBTB differs in that bronchoscopy plays a central role in diagnosis and, particularly in untreated cases, it may lead to serious airway complications such as bronchial stenosis. This case report describes a 57-year-old female patient with risk factors for lung cancer who was diagnosed with EBTB during advanced diagnostic evaluation. The case is presented to draw attention to the diagnosis, treatment, and management of this rare form of tuberculosis.

Keywords: *Endobronchial Tuberculosis, Bronchoscopy, Tuberculosis.*

Öz

Endobronşial Tüberküloza (EBTB) tüberkülozun oldukça nadir görülen bir formudur. Klinik, laboratuvar, mikrobiyolojik ve tedavi özellikleri Tüberküloz Hastalığının diğer formları ile benzerlik gösterse de, tanıda bronkoskopik yöntemlerin öncelikli olması ve başta tedavisiz olgularda olmak üzere görülen komplikasyonları arasında bronşial stenoz gibi ciddi bir havayolu hastalığına yol açabilmesi nedeni ile tüberkülozun diğer formlarından ayrışır. Bu olgu sunumunda Akciğer Kanseri açısından risk faktörlerine sahip olan 57 yaşında bir kadın olgu ileri tetkikler sırasında EBTB tanısı alması literatür eşliğinde incelenmiştir. Olgu nadir görülen EBTB'nin tanı, tedavi ve yönetimine dikkat çekmek için sunulmuştur.

Anahtar Kelimeler: *Endobronşial Tüberküloz, Bronkoskopi, Tüberküloz.*

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Tuberculosis (TB) is a chronic infectious disease caused by *Mycobacterium tuberculosis* and remains among the leading causes of morbidity and mortality worldwide. According to estimates in the Tuberculosis Report published by the World Health Organization (WHO) in 2025, millions of new cases are diagnosed each year (1).

Endobronchial tuberculosis (EBTB) is a distinct and relatively rare form of pulmonary TB. EBTB is defined as involvement of the tracheobronchial tree supported by microbiologic and histopathologic evidence. The reported incidence of EBTB is not clearly known, largely because bronchoscopy is not performed in every patient with TB. Although the clinical presentation of EBTB often includes nonspecific symptoms such as cough, wheezing, hemoptysis, or dyspnea, it may also mimic asthma, pneumonia, or malignancy. Radiologic findings may be negative, indeterminate, or misleading, which further contributes to diagnostic difficulty. Bronchoscopic, histopathologic, and microbiologic evaluations play an important role in preventing bronchial stenosis, a serious complication of EBTB (2,3).

The purpose of presenting this case is to emphasize the diagnostic challenges associated with EBTB and to highlight the importance of considering this disease in the differential diagnosis, even when the clinical and radiologic findings of endobronchial lesions suggest malignancy.

CASE

A 57-year-old female patient presented to a health center with dry cough, fatigue, loss of appetite, and weight loss lasting for 3 months. She underwent posteroanterior chest radiography (PA-CXR), followed by chest computed tomography (CT) and positron emission tomography-computed tomography (PET-CT), and was referred to our center for further evaluation. Physical examination revealed that the patient was in fair general condition and was alert, cooperative, and oriented. Auscultation revealed rales in the right lower zone. The remainder of the physical examination was unremarkable. The patient's medical history included chronic obstructive pulmonary disease (COPD), hypertension, hyperlipidemia, and coronary artery disease. She had been receiving inhaled therapy consisting of a long-acting beta-2 agonist and an inhaled corticosteroid for COPD for 2 years. The patient had a 35-pack-year smoking history. She reported that her father had died of lung cancer and her brother had died of malignant melanoma.

Evaluation of the patient's PA-CXR revealed a nonhomogeneous opacity in the right lower zone (Figure 1). Chest CT obtained at an outside center revealed areas of consolidation in the right lung, particularly prominent in the basal segments of the lower lobe (Figure 2).

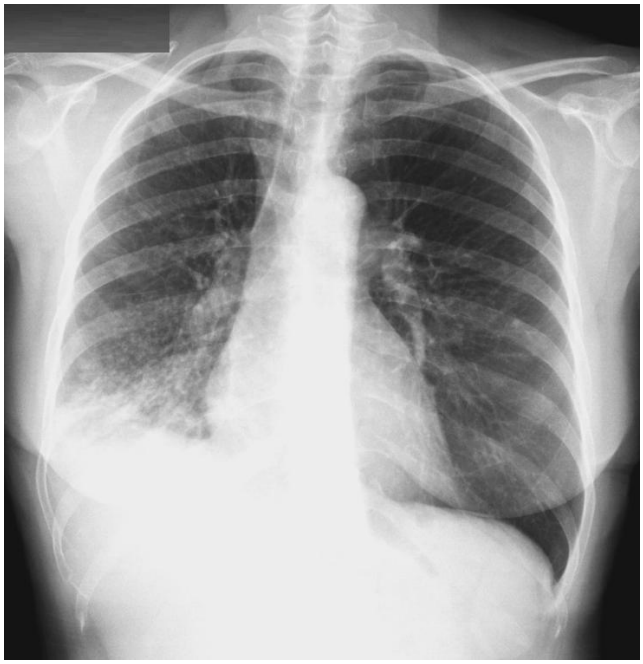

Figure 1: *Increased nonhomogeneous opacity in the right lower zone and volume loss in the right lung*

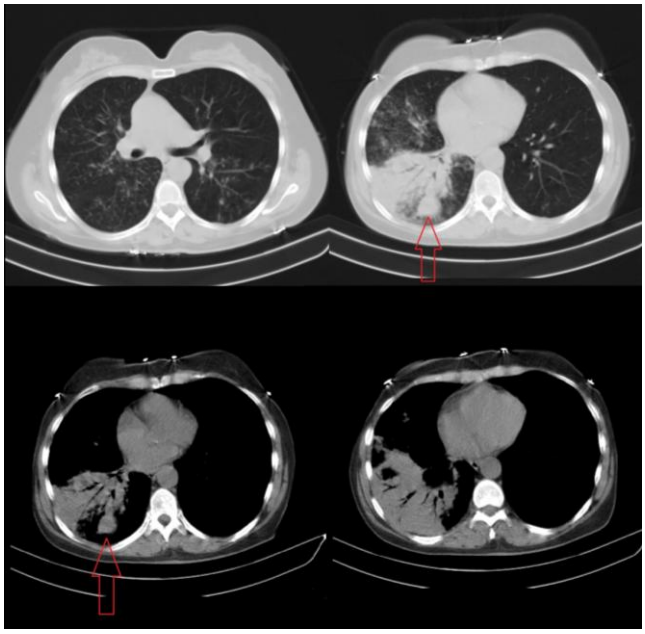

Figure 2: *Increased millimetric nodular densities along the bronchovascular branches in the posterior upper lobes of both lungs, more prominent on the right; areas of infiltration with air bronchograms in the right lower lobe; and a 2.5-cm nodule adjacent to the infiltration in the posterobasal segment of the right lung, indicated by the red arrow*

PET-CT showed areas of FDG uptake. Although the findings were suggestive of an infectious process, malignancy was also considered in the differential diagnosis because of the existing risk factors (Figure 3). Laboratory tests showed no significant pathologic findings other than elevated erythrocyte sedimentation rate (ESR) and serum C-reactive protein (CRP) levels (Table 1).

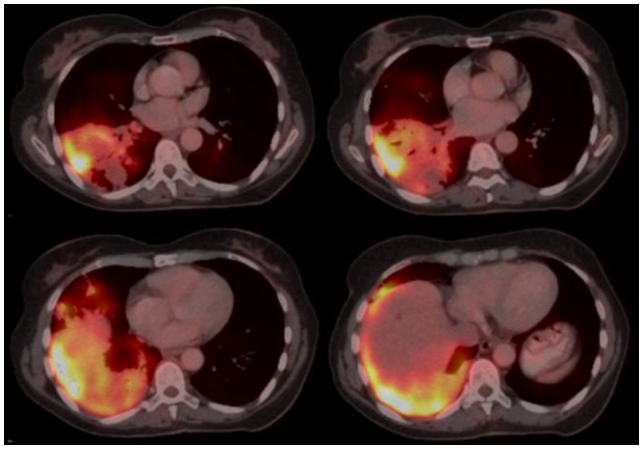

Figure 3: Areas of increased reticulonodular density accompanied by mild FDG uptake in the apicoposterior segment of the right upper lobe, superior segment of the right lower lobe, and left lower lobe (SUVmax = 2), with a large consolidation area showing intense FDG uptake in the right lower lobe (SUVmax = 12.1). Adjacent to the consolidation area, a 2.5-cm nodular lesion with mild FDG uptake is observed in the postero-basal region of the right lower lobe (SUVmax = 2.6)

Fiberoptic bronchoscopy (FOB) was planned to rule out possible malignancy. During FOB, a white membranous lesion narrowing the lumen and adhering to the carina was observed at the entrance of the right lower lobe, and multiple forceps biopsies were obtained from the lesion (Figure 4).

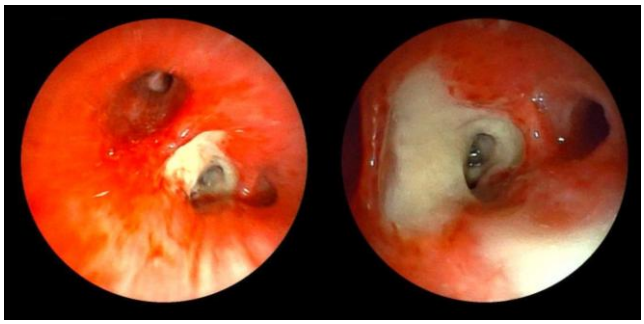

Figure 4: Fiberoptic bronchoscopy showing a white membranous lesion narrowing the lumen and adhering to the carina at the entrance of the right lower lobe

In addition, bronchial lavage was performed from the right lower lobe. Microbiologic examination of the bronchial lavage samples revealed acid-fast bacilli (AFB) positivity. Histopathologic examination showed necrotic tissue covered with respiratory epithelium, with inflammatory cells present within the necrotic areas (Figure 5).

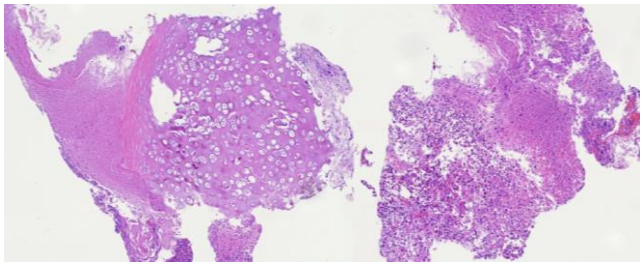

Figure 5: Caseous necrosis adjacent to the bronchial cartilage, with epithelioid histiocytes within the inflammatory cell infiltrate (H&E, X50)

Table 1: The patient's laboratory test results

Parameter	Result	Reference	Parameter	Result	Reference
WBC (10³/μL)	5,6	4.0–10.0	**CRP** (mg/L)	**44**	< 5
Hemoglobin (g/dL)	10,8	12.0–16.0	**INR**	0,96	0.8–1.2
PLT (10³/μL)	318	150–400	**ALT** (U/L)	13	7–35
Urea (mg/dL)	35	15–45	**AST** (U/L)	18	10–35
Kreatinin (mg/dL)	0,64	0.5–1.1	**ESR** (mm/saat)	**56**	0–20

WBC: White Blood Cell Count. CRP: C-Reactive Protein. INR: International Normalized Ratio. PLT: Platelet Count. ALT: Alanine Aminotransferase. AST: Aspartate Aminotransferase. ESR: Erythrocyte Sedimentation Rate.

Based on the current clinical, radiologic, bronchoscopic, and microbiologic findings, the patient was diagnosed with pulmonary tuberculosis with endobronchial involvement and was started on a four-drug antituberculosis regimen consisting of isoniazid, rifampicin, ethambutol, and pyrazinamide. In the second month of treatment, marked improvement in symptoms, regression of radiographic findings on PA-CXR, and regression of the endobronchial lesion on follow-up FOB were observed (Figure 6). Written informed consent was obtained from the patient.

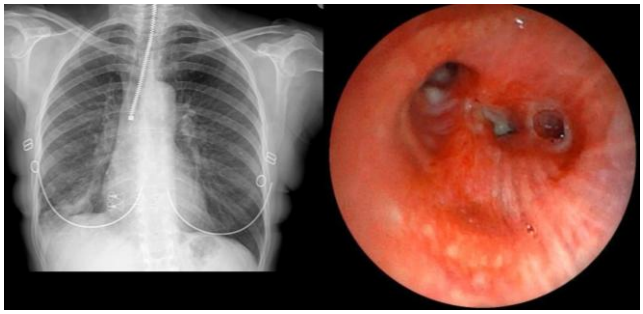

Figure 6: Follow-up chest radiograph and fiberoptic bronchoscopy obtained in the second month of treatment showing regression

DISCUSSION

This case report presents a 57-year-old woman at increased risk for lung cancer due to a history of heavy smoking and a family history of lung cancer in a first-degree relative, who was diagnosed with EBTB, a rare form of TB, during advanced evaluation including FOB.

The tuberculosis bacillus is a slow-growing obligate aerobic bacterium. Individuals first exposed to the TB bacillus during childhood may either develop primary TB disease or harbor dormant bacilli in the apical regions of the lungs, where ventilation is greater. Therefore, the form of TB seen in adults later in life, known as post-primary TB, is often cavitary TB (4). EBTB is a rare form of TB, and the diagnosis is often made using bronchoscopic methods. Although it is less common than other forms of TB, there

is no clear consensus on its prevalence. Studies report a wide prevalence range of 5% to 38%. One possible reason for this wide range may be the greater use of microbiologic methods in TB diagnosis and the more limited use of bronchoscopic methods (5).

EBTB, which is more commonly seen in women in their 20s and 30s, has also been reported to have a second peak in older patients (2). Our case was consistent with the literature in terms of age and sex. The clinical presentation of EBTB is usually nonspecific, with symptoms such as cough, dyspnea, wheezing, and hemoptysis. These findings may be confused with asthma, pneumonia, and especially malignancy. Radiologic findings may include consolidation, atelectasis, and an endobronchial mass-like appearance; in some cases, FDG uptake on PET-CT may also raise suspicion of malignancy. This demonstrates that evaluations based solely on radiologic methods may be insufficient for diagnosis (6,7). Our patient was diagnosed with EBTB during evaluation for suspected lung cancer due to her high-risk status. A review of the literature shows that although FOB biopsy is diagnostically important in EBTB, the role of histopathologic examination may sometimes be limited compared with microbiologic methods. This may be due to the high degree of necrosis in the samples obtained, nonspecific inflammation, or results that only rule out malignancy. For this reason, microbiologic methods, particularly M. tuberculosis culture and PCR testing in bronchial lavage samples, are critical for definitive diagnosis (5,8).

The most important feature distinguishing EBTB from other forms of TB, despite a similar treatment approach, is the risk of bronchial stenosis. This is the most significant complication of EBTB and may develop in some patients despite appropriate antituberculosis treatment and steroid therapy. Although rare, bronchial stenosis that develops during treatment may be irreversible, requiring bronchoscopic or surgical intervention to maintain airway patency. Early diagnostic investigation and timely initiation of antituberculosis therapy are critical for preventing irreversible complications such as bronchial stenosis (9,10). Therefore, EBTB should always be considered in the differential diagnosis of endobronchial lesions mimicking malignancy.

Another notable aspect of our case is the patient's history of heavy smoking, diagnosis of COPD, and the LABA+ICS therapy she had been receiving for this condition. ICSs, in particular, may suppress the cellular immune response, adversely affect alveolar macrophages, reduce local defense mechanisms in the bronchial mucosa, and potentially weaken granuloma integrity through TNF-α inhibition. Studies have reported that ICSs may increase the risk of pneumonia, nontuberculous mycobacterial infections, and reactivation of latent tuberculosis, with these effects being small but significant, particularly with high-dose and long-term use (11). Ikeda et al. (12) recently presented a case exhibiting clinical and radiologic features very similar to our case. In that report, EBTB diagnosed by bronchoscopy was thought to have been triggered by long-term ICS use and presented with atypical epidemiologic features, anatomic involvement, and atypical radiologic findings. The authors highlighted the possibility of EBTB in patients receiving long-term ICS therapy, even when the clinical findings are largely atypical for tuberculosis.

CONCLUSION

In our case, despite clinical and radiologic findings mimicking malignancy, EBTB was diagnosed based on bronchoscopic and microbiologic evaluation. This case emphasizes the importance of bronchoscopic and microbiologic examinations in the evaluation of endobronchial lesions. The primary goal of treatment in EBTB is eradication of the tuberculosis bacillus. Careful follow-up for possible bronchial stenosis is also essential.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - N.A.A., C.D., S.Ö.; Planning and Design - N.A.A., C.D., S.Ö.; Supervision - N.A.A., C.D., S.Ö.; Funding - N.A.A., C.D., S.Ö.; Materials - N.A.A., C.D., S.Ö.; Data Collection and/or Processing - N.A.A., C.D., S.Ö.; Analysis and/or Interpretation - N.A.A., C.D., S.Ö.; Literature Review - N.A.A., C.D., S.Ö.; Writing - N.A.A., C.D.; Critical Review - C.D.

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A Case of Talcosis Complicated by Progressive Massive Fibrosis

Progresif Masif Fibrozisin Eşlik Ettiği Bir Talkozis Olgusu

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Abstract

Talcosis is a fibrotic lung disease that results from the inhalation of talc. It can occur in various occupational settings, such as mining and industrial work. In talcosis, the typical finding on high-resolution computed tomography (HRCT) is diffuse centrilobular micronodules. A patient with no history of smoking who worked in the lamination department of an automotive glass factory presented with shortness of breath. The patient's chest radiograph showed increased bilateral reticulonodular densities and large opacities in the upper zones of both lungs. Based on a comprehensive evaluation of the patient's occupational history, clinical presentation, and imaging findings, talc pneumoconiosis was considered the most likely diagnosis. In progressive fibrotic occupational lung diseases such as this, for which there is generally no specific treatment, the usual approach is to control the factors that contribute to disease development.

Keywords: *Pneumoconiosis, Pulmonary Fibroses, Pulmonary talcosis, Progressive Massive Fibrosiss.*

Öz

Talkozis, talk mineralinin solunması ile meydana gelen fibrotik bir akciğer hastalığıdır. Madencilik ve sanayi gibi birçok iş kolunda meydana gelebilmektedir. Talkoziste Yüksek Çözünürlüklü Bilgisayarlı Tomografi'de beklenen bulgu, genellikle yaygın olarak dağılmış sentrilobüler mikronodüllerdir. Otocam sanayinde laminasyon bölümünde çalışan daha önce sigara içme öyküsü olmayan hasta, nefes darlığı yakınması ile başvurdu. Hastanın akciğer grafisinde bilateral retikülonodüler dansite artışı, her iki akciğerde üst zonlarda büyük opasiteler görüldü. Hasta klinik ve radyolojik bulgular ile birlikte ayrıntılı meslek öyküsü değerlendirildiğinde talk pnömokonyozu düşünüldü. Tedavisi genellikle olmayan ilerleyici fibrotik mesleki akciğer hastalıklarında genel yaklaşım, hastalığın oluşumuna etki eden faktörlerin kontrol edilmesidir.

Anahtar Kelimeler: *Pnömokonyoz, Akciğer fibrozisleri, Pulmoner talkozis, Progresif Masif Fibrozis.*

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Talcosis is a lung disease caused by the inhalation of talc and is closely associated with a history of occupational exposure. Inhalational exposure to talc is a significant occupational and environmental risk that can occur in various industries, including mining, secondary industrial processes, and the production of consumer products. Workers in various sectors, including rubber, paper, textiles, ceramics, pharmaceuticals, and cosmetics, may be exposed during the production and processing of talc-containing materials. Furthermore, the use of finished products, such as cosmetic talcum powder, has also been identified as a potential source of inhalational exposure (1).

The causes of pulmonary talcosis can be categorized into different forms: pure talcosis, talc inhalation with silica (talcosilicosis), talc inhalation with asbestos fibers (talcoasbestosis), and intravenous talc injection, typically associated with substance use (1-4). Talc causes non-necrotizing granulomatous inflammation, which leads to progressive fibrosis (2,3). Radiographic findings may show diffuse nodulation and reticulation, similar to those of asbestosis; however, the lung apices and costophrenic sinuses are generally spared. As with other forms of pneumoconiosis, the coalescence of nodules can lead to large opacities resembling progressive massive fibrosis. Some patients also develop hilar lymphadenopathy (2).

On High-Resolution Computed Tomography, the most common finding is diffuse centrilobular micronodules. Additional findings may include septal and subpleural lines, ground-glass opacities, and large opacities with areas of high attenuation due to talc accumulation. Patients may also present with emphysematous changes, pleural plaques, and lymph node enlargement. The imaging findings of talcoasbestosis and talcosilicosis are generally similar to those of asbestosis and silicosis, respectively (3,4).

CASE

A 50-year-old male patient presented to the outpatient clinic with shortness of breath. His occupational history revealed that he had worked in a glass factory for 6 years (2002–2008), where his duties included placing bus windows into a machine in an enclosed space, pouring talc into the machine, holding the glass while it was being coated with talc, and manually brushing the windows with talc powder. Substantial amounts of talcum powder were present in the work environment. The patient had no previous history of smoking and was found to have stridor and expiratory rhonchi on respiratory examination. He had no prior history of tuberculosis.

Patient's oxygen saturation was 96% on room air. Pulmonary function tests (PFTs) showed a forced expiratory volume in 1 second (FEV1) of 18% predicted (0.61 L), a forced vital capacity (FVC) of 38% predicted (1.57 L), and an FEV1/FVC ratio of 39%. Despite the absence of a smoking history, the patient's PFT results indicated a severe mixed obstructive and restrictive defect. Although stridor was noted during physical examination and is not considered a typical manifestation of talcosis, comprehensive evaluation revealed no acute or chronic pathologic process causing bronchial airway obstruction. The patient's chest radiograph showed bilateral reticulonodular infiltrates and large opacities in the upper zones of both lungs (Figure 1).

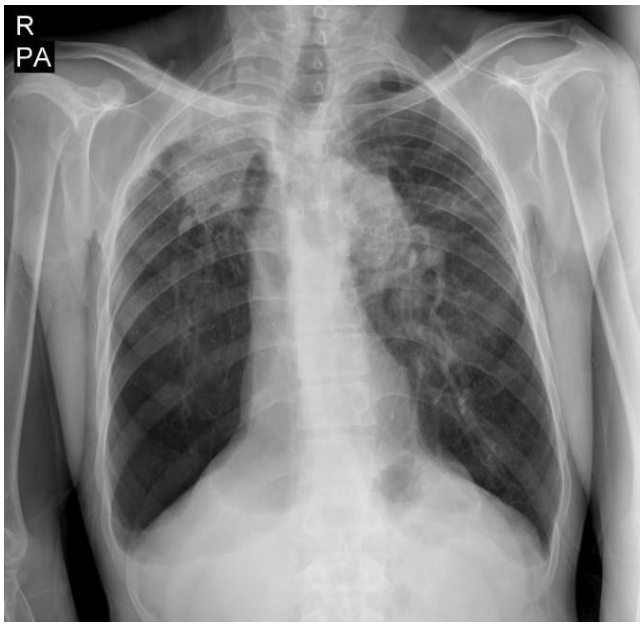

Figure 1: Increased bilateral reticulonodular densities and large opacities in the upper zones of both lungs.

A High-resolution computed tomography (HRCT) of the lungs was performed and revealed several key findings. The HRCT findings included enlarged and partially calcified lymph nodes in the mediastinal and hilar regions. HRCT demonstrated emphysematous changes, with micronodules and reticular opacities showing upper lung predominance. Consolidated areas containing calcifications were identified in the right upper zone and the perihilar area of the left mid-lung zone (Figure 2).

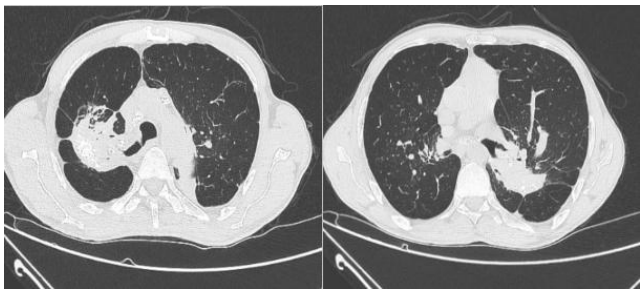

Figure 2: HRCT showing consolidated areas containing calcifications in the right (A) and left (B) perihilar regions.

Based on clinical and radiologic evaluations, the patient was diagnosed with talcosis. Pneumoconiosis was reported according to the International Labour Organization (ILO) classification, with a score of q/t 1/2 + C. The patient, who had no history of smoking, was considered to have occupational chronic obstructive pulmonary disease (COPD). The CT findings of talc pneumoconiosis are nonspecific and can also be seen in asbestosis and silicosis. Although this combination of findings may be seen in patients with mixed dust exposure, talc pneumoconiosis should be considered in patients with a history of occupational exposure who present with diffuse centrilobular nodules, large dense opacities, dense lymph nodes, and pleural plaques.

DISCUSSION

Talc is used in many sectors as a raw material in the manufacture of products such as paints, paper, textiles, and ceramic tiles (2). Talcosis, which develops after excessive inhalation of talc, is usually asymptomatic. When symptomatic, it presents with nonspecific symptoms such as progressive shortness of breath and cough. Late complications include chronic respiratory failure, emphysema, pulmonary arterial hypertension, and cor pulmonale (1). High-resolution computed tomography provides superior diagnostic efficacy in differentiating talc-induced lesions from other interstitial lung diseases. Exposure to talc leads to pulmonary parenchymal fibrosis, and progressive massive fibrosis may develop depending on the extent of fibrosis (1,4).

Rather than being based solely on clinical evaluation, the diagnosis of pneumoconiosis requires a thorough occupational exposure assessment, including a detailed occupational history and an evaluation of workplace hygiene and engineering controls (5). In several occupational lung diseases, histopathologic confirmation is not mandatory when a consistent occupational history is accompanied by typical chest imaging findings (6).

The severe mixed obstructive and restrictive pattern on the patient's pulmonary function tests was attributed to pneumoconiosis. Both small and large airway obstruction are expected findings in patients with pneumoconiosis and progressive massive fibrosis (PMF), with a higher prevalence among those with PMF (7,8). In a study evaluating patients with pneumoconiosis, Sarı et al. (9) found that FEV1 and FVC values were significantly lower in the group with PMF than in the group without PMF. Additionally, COPD was more frequent in the PMF group. Furthermore, a study by Pınar et al. (10) evaluating the prevalence of COPD in patients with pneumoconiosis reported that the frequency of COPD was correlated with the size of PMF. The loss of respiratory function in our patient with inorganic dust exposure and PMF, together with category C opacities on radiography, was consistent with the literature. Recent studies suggest that exposure to inorganic dust may lead to chronic obstructive pulmonary disease (11).

CONCLUSION

Although talc is known to cause pneumoconiosis, reports on this condition remain limited in the literature. Furthermore, in the automotive industry, where talc use is not widespread, a history of occupational talc exposure and a compatible imaging pattern should prompt consideration of talcosis. A detailed occupational exposure history and clinical awareness of occupational diseases are crucial for the diagnosis of pneumoconiosis. This case illustrates a rare presentation of talcosis manifesting as PMF and emphasizes the importance of recognizing occupational exposure as a key contributing factor. Due to the irreversible decline in lung function, the patient was referred to a lung transplantation center. Unfortunately, recent literature indicates that no clinically validated effective treatments are available to prevent PMF (12,13). The general approach to occupational diseases is to control the factors that contribute to disease development.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - G.E.A., A.K., G.S., C.Ş.; Planning and Design - G.E.A., A.K., G.S., C.Ş.; Supervision - G.E.A., A.K., G.S., C.Ş.; Funding - G.E.A., A.K.; Materials - G.E.A., A.K., G.S.; Data Collection and/or Processing - G.E.A., A.K., G.S.; Analysis and/or Interpretation - G.E.A., A.K., G.S.; Literature Review - G.E.A.; Writing - G.E.A.; Critical Review - G.E.A., A.K., G.S., C.Ş.

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Re-evaluation of the Role of Corticosteroids in Extrapulmonary ARDS Following Peptic Ulcer Perforation

Peptik Ülser Perforasyonunu Takiben Ekstra Pulmoner ARDS'de Kortikosteroidlerin Rolünün Yeniden Değerlendirilmesi

Veysel Garani Soylu[1], İbrahim Kurtoğlu[2], Gökçe Kızılkale Kayıkcı[3]

Abstract

It is a common belief among clinicians that corticosteroid therapy is an etiologic factor in peptic ulcer disease and its complications. However, this belief is based largely on older studies, and many clinicians avoid using these agents in such patients. Nevertheless, complex clinical situations may arise in which both indications for and concerns about corticosteroid use coexist. Although corticosteroids are often considered risky or relatively contraindicated in patients with peptic ulcers, they may be indicated in the treatment of acute respiratory distress syndrome (ARDS). Here, we report a case of extrapulmonary ARDS secondary to peptic ulcer perforation that was successfully treated with corticosteroids. The patient received methylprednisolone at a dose of 1 mg/kg/day and was successfully treated and discharged without complications. Because of the widespread belief that corticosteroid therapy is contraindicated in peptic ulcer disease, few recent studies have addressed this issue. This case may renew interest in the presumed ulcerogenic effects of corticosteroids in peptic ulcer disease and its complications, thereby encouraging further research.

Keywords: *Steroid, Perforated Peptic Ulcer, ARDS.*

Öz

Klinisyenler arasında kortikosteroid tedavisinin peptik ülser hastalığı ve komplikasyonlarında etiyolojik bir faktör olduğuna dair yaygın bir inanış vardır. Ancak bu inanış eski çalışmalara dayanmaktadır ve birçok klinisyen bu tür hastalarda bu ajanları kullanmaktan kaçınmaktadır. Öte yandan, kullanımları için hem endikasyonların hem de kontrendikasyonların bir arada bulunduğu karmaşık klinik durumlar ortaya çıkabilir. Steroidler peptik ülserler için kontrendike olmasına rağmen, akut solunum sıkıntısı sendromu (ARDS) için endikedir. Burada, kortikosteroidler ile başarılı bir şekilde tedavi edilen peptik ülser perforasyonuna bağlı ekstrapulmoner ARDS olgusunu bildiriyoruz. Hasta, günde 1 mg/kg dozunda metilprednizolon aldı ve herhangi bir komplikasyon olmadan başarılı bir şekilde tedavi edilerek taburcu edildi. Kortikosteroid tedavisinin peptik ülser hastalığında kontrendike olduğuna dair yaygın inanış nedeni ile, bu alanda son zamanlarda çok az çalışma yapılmıştır. Bu olgu, steroidlerin peptik ülser ve komplikasyonlarındaki ülserojenik etkilerine olan ilgiyi artırabilir ve yeni araştırmaları teşvik edebilir.

Anahtar Kelimeler: *Steroid, Perfore peptik ülser, ARDS.*

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Synthetic corticosteroids, such as methylprednisolone and hydrocortisone, exert their clinical effects by mimicking the actions of natural glucocorticoids. Glucocorticoids are potent anti-inflammatory agents that primarily act by binding to cytoplasmic glucocorticoid receptors (1).

Steroids reduce gastric mucus and bicarbonate secretion, thereby weakening gastric mucosal defenses. In addition, steroids disrupt angiogenesis and epithelial repair mechanisms in experimental ulcers (2,3). As a result of these effects, the gastric mucosa may become more sensitive to gastric acid and more prone to peptic ulcer-related complications. For these reasons, the administration of steroid therapy in patients with peptic ulcers and related complications remains a controversial and concerning issue for clinicians.

Acute respiratory distress syndrome (ARDS) is characterized by hypoxic respiratory failure associated with dysregulated inflammation (4). Because of their anti-inflammatory effects, steroids are recommended in the early stage of ARDS treatment. Peritonitis following peptic ulcer perforation is an abnormal inflammatory response driven by cytokines such as tumor necrosis factor (TNF), interleukin-1, and interferon (5). This inflammatory response may also lead to the development of extrapulmonary ARDS (6).

To our knowledge, no data are available on steroid administration for complications following peptic ulcer perforation in clinical situations where both indications and contraindications coexist. In this case, we present the administration of steroids in a patient who developed extrapulmonary ARDS secondary to peptic ulcer perforation, despite the potential concerns associated with steroid use in peptic ulcer disease. Written informed consent was obtained from the patient.

CASE

A 27-year-old male patient presented to the emergency department with abdominal pain of three days' duration. He had no comorbidities or history of medication use. He reported intermittent cramping pain in the upper-middle abdominal region. On physical examination, diffuse abdominal guarding was noted. His vital signs were as follows: SpO_2 was 88% on room air, heart rate was 122 bpm, and blood pressure was 140/82 mmHg. Laboratory findings included a leukocyte count of 25 $\times$ $10^3/\mu L$, hemoglobin of 12.2 g/dL, platelet count of 293 $\times$ $10^3/\mu L$, creatinine of 1.45 mg/dL, alanine aminotransferase (ALT) of 15 U/L, aspartate aminotransferase (AST) of 30 U/L, C-reactive protein (CRP) of 498 mg/L, and an INR of 1.74. Arterial blood gas analysis showed a pH of 7.21, PCO_2 of 58.8 mmHg, PO_2 of 38 mmHg, and lactate level of 4.1 mmol/L. The PaO_2/FiO_2 ratio was 190, indicating moderate ARDS. Upright abdominal radiography, posteroanterior chest radiography (PA-CXR), and abdominal/thoracic computed tomography (CT) were performed. Upright abdominal radiography showed free air under both diaphragms. Abdominal CT demonstrated diffuse free fluid and free intraperitoneal air. Thoracic CT showed patchy, predominantly consolidated peribronchovascular areas and widespread acinar infiltrates in both lungs, more prominent in the right upper lobe (Figure 1).

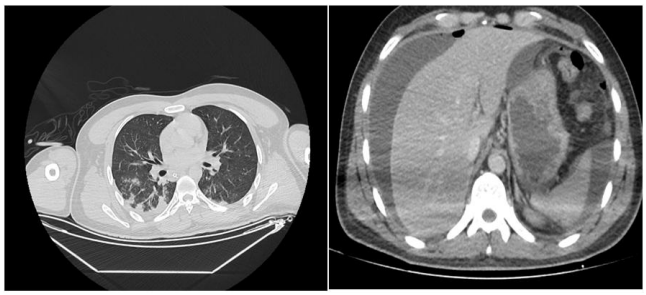

Figure 1: Abdominal and thoracic CT images of the patient on admission to the emergency department

The patient was taken to surgery with a diagnosis of acute abdomen due to free perforation. A 0.8 $\times$ 0.5 cm perforated area was observed on the anterior surface of the stomach, and modified Graham omentopexy was performed. The patient was orotracheally intubated for hypoxic respiratory failure and transferred to the intensive care unit (ICU). After admission to the ICU, antibiotic therapy (meropenem 1 g IV three times daily), sedation (midazolam 5 mg/h IV), proton pump inhibitor therapy (lansoprazole 40 mg/day IV), and fluid resuscitation were initiated. The patient had severe ARDS, with a PO_2 of 80 mmHg despite receiving 100% FiO_2, and lung-protective ventilation was started.

On postoperative day 1, PA-CXR showed diffuse bilateral opacification (Figure 2). Despite lung-protective ventilation, the patient remained in severe ARDS; therefore, he was placed in the prone position for 16 hours, and methylprednisolone 1 mg/kg/day IV was added to the treatment. After 72 hours of follow-up, with a FiO_2 of 60% and a PO_2 of 112 mmHg, the patient no longer required prone positioning, and ventilation was changed from a lung-protective strategy to a conventional strategy. Daily PA-CXRs were obtained (Figure 2).

By day 7 in the ICU, the patient was extubated and switched to high-flow nasal oxygen (HFNO) support. On day 12, he was transferred from the ICU to the general ward. Methylprednisolone therapy at 1 mg/kg/day was continued for 10 days. After follow-up in the ward, the patient was discharged in good condition.

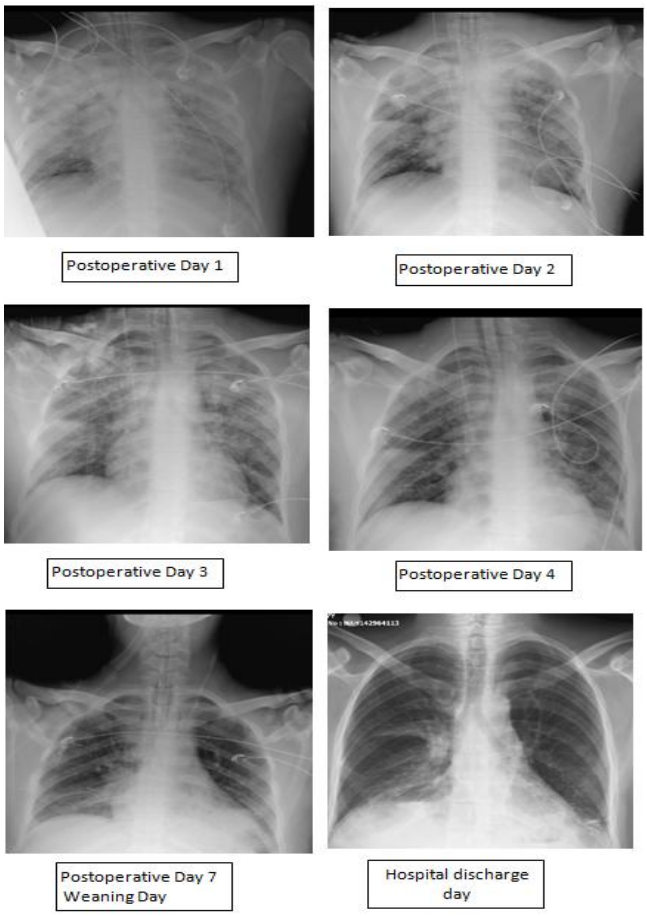

Figure 2: Daily PA chest radiographs obtained during ICU follow-up

DISCUSSION

The histologic features of early-stage ARDS include interstitial and alveolar edema, infiltration of neutrophils, macrophages, and red blood cells into the alveolar spaces, and injury to the alveolar epithelium and endothelium (7). In ARDS, alveolar macrophages release proinflammatory cytokines, including neutrophil chemoattractants, which trigger the activation and migration of neutrophils into the interstitial and alveolar spaces. These activated neutrophils release proinflammatory molecules that maintain and amplify the inflammatory response (8). In peritonitis secondary to peptic ulcer perforation, dysregulated release of inflammatory cytokines may lead to extrapulmonary ARDS (5). In patients with peptic ulcer perforation, sepsis may be the underlying cause of extrapulmonary ARDS and is the most common etiology of extrapulmonary ARDS (9). Our patient developed extrapulmonary ARDS secondary to delayed peptic ulcer perforation.

Steroids are potent anti-inflammatory agents that primarily act by binding to cytoplasmic glucocorticoid receptors and modulating fibrotic pathways by inhibiting fibroblast proliferation and decreasing collagen accumulation (10,11). Because of these effects, steroid therapy is recommended in the early phase of ARDS (12). Although steroids are thought to play a role in the etiology of peptic ulcers, early low-dose steroids are recommended as an adjunct to ARDS treatment alongside other therapies such as lung-protective ventilation, prone positioning, and surgical source control in cases of abdominal sepsis, because of their synergistic effect on treatment (13). In ARDS, low-dose steroids have been associated with better outcomes than high-dose steroids in terms of mortality, infection, and ventilator-free days (14). It has been reported that steroid therapy is effective when given within 14 days after ARDS diagnosis and continued for more than 7 days (15). In our patient, steroid therapy was started on the first day of ARDS and continued for 10 days.

Peptic ulcer disease results from mucosal damage caused by hydrochloric acid (HCl) and pepsin, most commonly in the stomach and duodenum, although it may occur in any part of the digestive system. In peptic ulcer disease, mortality occurs secondary to major ulcer complications, such as bleeding, perforation, and obstruction. The use of nonsteroidal anti-inflammatory drugs (NSAIDs) and Helicobacter pylori infection are the most important risk factors for peptic ulcer disease (16). The effect of corticosteroids on the development of peptic ulcers is a long-standing concept, based mainly on evidence from experimental and pharmacologic studies, and is still widely accepted by clinicians. A study conducted in the Czech Republic showed that 82% of physicians believed that steroids are ulcerogenic (17). Although experimental studies suggest that steroids may cause ulcers by impairing gastric mucus and bicarbonate secretion, the actual incidence of steroid-induced ulcers in clinical practice is relatively low. Studies suggesting that steroids are ulcerogenic are relatively old. The last large analysis was conducted in 1994, and no extensive studies have been undertaken since then. Because of the widely accepted belief among clinicians that steroids are ulcerogenic, new studies in this field remain limited (18,19). In our clinic, there was also concern about using steroids in patients with peptic ulcer disease and its complications. In our case, competing indications were present, a risk-benefit assessment was performed, and low-dose steroid therapy (1 mg/kg/day) was administered. No complications were observed.

CONCLUSION

This case does not allow definitive conclusions regarding corticosteroid use in patients with peptic ulcer disease and related complications. However, it suggests that in carefully selected patients with competing indications, corticosteroid therapy may be considered after individualized risk-benefit assessment. Further studies are needed to clarify the safety and efficacy of corticosteroid therapy in this clinical context.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - V.G.S., İ.K., G.K.K.; Planning and Design - V.G.S., İ.K., G.K.K.; Supervision - V.G.S., İ.K., G.K.K.; Funding -; Materials -; Data Collection and/or Processing - V.G.S.; Analysis and/or Interpretation -; Literature Review - V.G.S., G.K.K.; Writing - V.G.S., İ.K., G.K.K.; Critical Review - V.G.S.

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Anomalous Systemic Arterial Supply to the Left Lower Lobe: Evaluation with Multimodality Imaging and Conservative Management

Sol Akciğer Alt Lobun Anormal Sistemik Arteriyel Beslenmesi: Multimodal Görüntüleme Temelli Değerlendirme ve Konservatif Tedavi Yaklaşımı

Adil Aytaç, Bahar Yanık

Abstract

Anomalous systemic arterial supply to the left lower lobe is a rare congenital pulmonary vascular anomaly in which normally developed lung parenchyma with intact bronchial communication receives blood directly from a systemic artery rather than from the pulmonary arterial circulation. We report the case of a 35-year-old man who presented with mild hemoptysis and palpitations. CT and MR angiography revealed a 10-mm aberrant systemic artery arising from the descending thoracic aorta and supplying the basal segments of the left lower lobe, with normal pulmonary venous drainage and no dysplastic parenchyma. In the absence of pulmonary hypertension or a significant shunt, conservative management with follow-up was chosen. During one year of follow-up, the patient remained asymptomatic without radiologic progression. This case underscores the critical role of multidetector CT and MR angiography in accurately differentiating this entity from pulmonary sequestration and highlights that noninvasive diagnosis can support safe conservative management in appropriately selected patients.

Keywords: *Anomalous Systemic Arterial Supply, Pulmonary Sequestration, CT Angiography.*

Öz

Sol akciğer alt lobunun anormal sistemik arteriyel beslenmesi, bronşiyal bağlantısı korunmuş ve normal gelişim gösteren akciğer parankiminin pulmoner arter yerine doğrudan sistemik bir arterden kan aldığı, nadir görülen konjenital bir pulmoner vasküler anomalidir. Bu makalede, hafif hemoptizi ve çarpıntı yakınmaları ile başvuran 35 yaşındaki bir erkek olgu sunulmaktadır. Bilgisayarlı tomografi ve manyetik rezonans anjiyografi incelemeleri, inen torasik aortadan köken alan ve sol alt lobun bazal segmentlerini besleyen 10 mm çapında aberan bir sistemik arter ortaya koymuş; pulmoner venöz drenajın normal olduğu ve parankimde herhangi bir displazi bulunmadığı gösterilmiştir. Pulmoner hipertansiyon veya belirgin bir şant akımının saptanmaması nedeni ile konservatif izlem tercih edilmiştir. Bir yıllık takip süresince hasta asemptomatik seyretmiş ve radyolojik olarak progresyon izlenmemiştir. Bu olgu, multidetektör BT ve MR anjiyografinin söz konusu anomalinin pulmoner sekestrasyondan doğru şekilde ayırt edilmesindeki kritik önemini vurgulamakta; noninvazif tanının uygun seçilmiş hastalarda güvenli konservatif yönetimi mümkün kıldığını göstermektedir.

Anahtar Kelimeler: *Anormal Sistemik Arteriyel Beslenme, Pulmoner Sekestrasyon, BT Anjiyografi.*

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Anomalous systemic arterial supply to the left lower lobe is a congenital pulmonary vascular anomaly in which normally developed lung parenchyma with preserved bronchial communication receives high-pressure systemic blood flow directly from a systemic artery—most commonly the descending thoracic aorta—rather than from the pulmonary arterial circulation, while venous drainage usually occurs through the normal pulmonary veins (1). It is thought to arise from the persistence of primitive systemic-pulmonary arterial connections that fail to regress during the dual-phase vascular development of the fetal lung (2).

This anomaly was first described by Pryce in 1946 and has since been referred to in the literature by various terms, including anomalous systemic arterial supply to the lung, isolated systemic arterialization, and the nonsequestrated type (3-5). The clinical presentation of this anomaly varies widely, ranging from asymptomatic cases to recurrent infections, hemoptysis, and, in some instances, the development of pulmonary hypertension (6). The true incidence remains unclear, but the anomaly is considered much rarer than pulmonary sequestration and is most often reported as isolated case reports or small case series (3-7). Because systemic arterial pressure is transmitted to the pulmonary circulation, unrecognized cases may lead to serious complications, and misdiagnosis as pulmonary sequestration can result in inappropriate management (8). Although it most frequently occurs as an isolated anomaly, coexistence with other congenital abnormalities—such as partial anomalous pulmonary venous return, bronchopulmonary foregut malformations, congenital cardiac defects, or diaphragmatic anomalies—has also been described (9).

The aim of this report is to present a rare vascular lung anomaly characterized by anomalous systemic arterial supply to the left lower lobe, emphasize the diagnostic value of noninvasive imaging techniques, highlight the importance of differentiating it from pulmonary sequestration, and demonstrate that conservative management may be a safe and appropriate option in selected patients (10).

CASE

A 35-year-old man presented to our cardiology department with a three-month history of fatigue and palpitations accompanied by intermittent mild hemoptysis. He denied dyspnea, cough, or other respiratory symptoms. The patient had no history of chronic disease and did not smoke, consume alcohol, or use any medications. Physical examination was unremarkable. Except for mild hypertriglyceridemia (194 mg/dL), comprehensive laboratory studies, including hematologic, renal, hepatic, and thyroid function tests, were within normal limits. Electrocardiography (ECG) findings were normal. Transthoracic echocardiography revealed a small atrial septal defect (ASD) measuring approximately 3 mm in the interatrial septum. To further characterize the defect, assess right heart chamber volume and function, quantify shunt flow, and evaluate potential surgical or percutaneous management options, cardiac MRI and MR angiography were performed. Phase-contrast MRI-based flow quantification demonstrated pulmonary (Qp) and systemic (Qs) flow rates of 5.04 L/min and 4.76 L/min, respectively, corresponding to a Qp/Qs ratio of 1.06 and a shunt fraction of approximately 6%. These findings indicated no hemodynamically significant shunt and were consistent with normal right heart volumes. Dynamic contrast-enhanced MR angiography demonstrated a 10-mm aberrant systemic artery arising from the descending thoracic aorta and extending toward the left lower lobe, accompanied by mild ectasia of the inferior pulmonary vein (Figure 1).

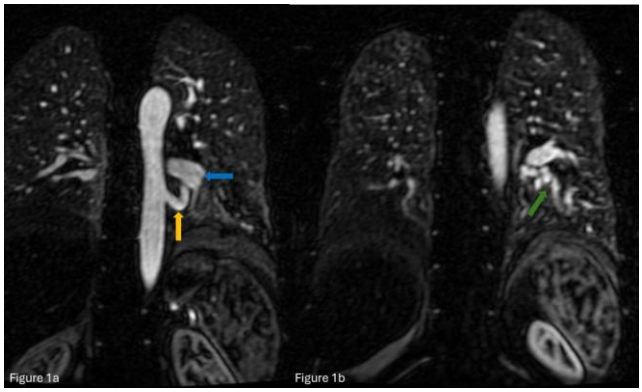

Figure 1: Contrast-enhanced MR angiography images in the coronal plane. The yellow arrow indicates an aberrant arterial vessel originating from the descending thoracic aorta, while the blue arrow demonstrates the ectatic inferior pulmonary vein (a); the green arrow highlights the dilated segmental arterial branches formed by the aberrant artery within the left lower lobe parenchyma (b).

To obtain higher spatial resolution, evaluate the vascular wall and pulmonary parenchyma in greater detail, and more precisely map pulmonary venous drainage using three-dimensional (3D) reformatted images for potential surgical or endovascular planning, thoracic CT angiography was subsequently performed. CT angiography confirmed a 10-mm aberrant systemic artery originating from the descending thoracic aorta at the level of the T8 vertebra and branching into segmental arteries supplying the left lower lobe. The left inferior pulmonary vein was mildly ectatic, and the bronchial tree was normal. Importantly, no dysplastic or cystic parenchymal changes suggestive of pulmonary sequestration were identified. Patchy ground-glass opacities suggestive of increased pulmonary perfusion were observed in the anteromedial basal, lateral basal, and posterior basal segments of the left lower lobe (Figure 2).

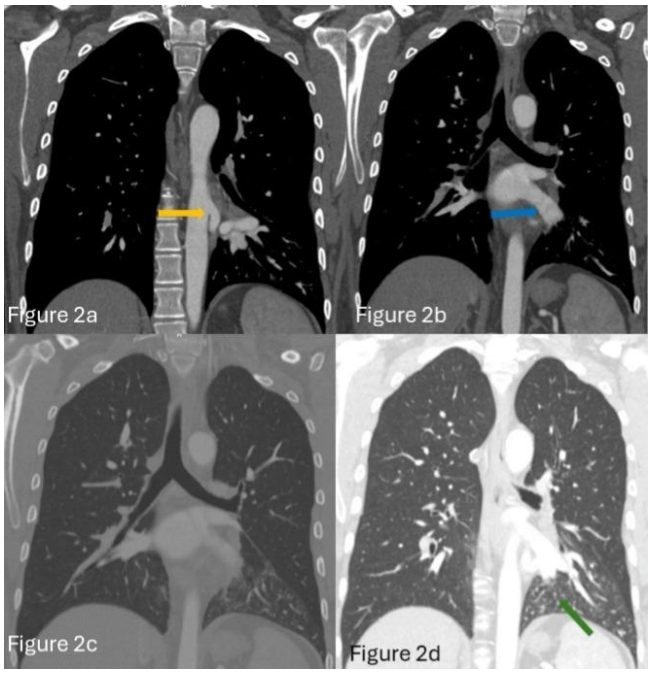

Figure 2: Coronal CT images. Mediastinal window image showing an aberrant arterial vessel originating from the descending thoracic aorta (yellow arrow) (a); mediastinal window image demonstrating the ectatic inferior pulmonary vein (blue arrow) (b); mediastinal window image depicting normal bronchial distribution (c); lung window image showing ground-glass opacities in the left lower lobe secondary to hyperperfusion (green arrow) (d).

Volume-rendered (VR) and maximum intensity projection (MIP) images demonstrated an aberrant systemic artery originating from the descending thoracic aorta and supplying the basal segments of the left lower lobe (Figure 3).

Figure 4 illustrates the imaging features of the left lower lobe, including patchy ground-glass opacities, a mildly ectatic inferior pulmonary vein, the normal course of the left lower lobe bronchus, and the origin and course of the anomalous systemic arterial branch.

Based on these imaging findings, a diagnosis of anomalous systemic arterial supply to the left lower lobe was established. As the patient had no evidence of pulmonary hypertension or a significant shunt, and right heart volumes were within normal limits, conservative management was preferred over surgical or endovascular intervention. During one year of follow-up with evaluations every three months, the patient remained clinically stable without new symptoms or radiologic progression.

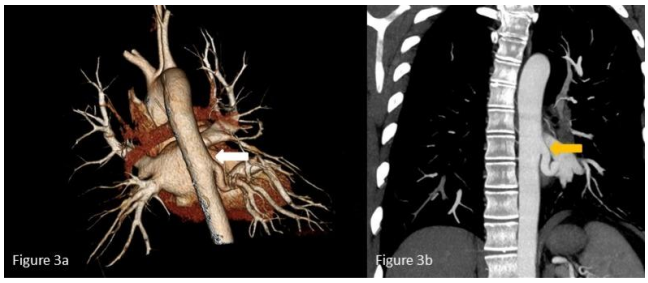

Figure 3: Volume-rendered image showing the aberrant systemic arterial structure originating from the descending thoracic aorta (white arrow) (a); coronal maximum intensity projection (MIP) image demonstrating the same anomalous systemic artery supplying the basal segments of the left lower lobe (yellow arrow) (b).

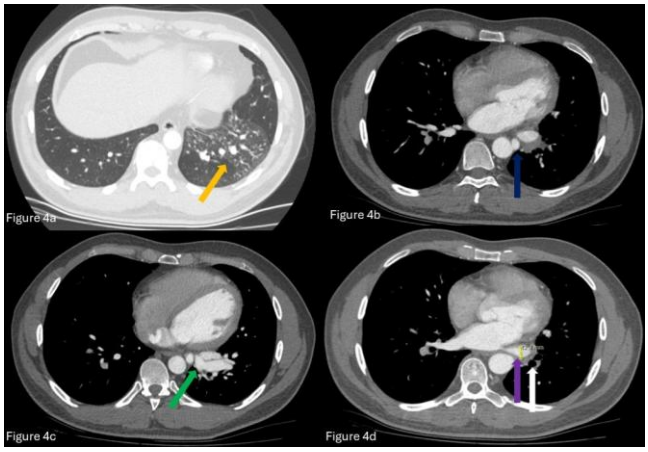

Figure 4: Axial thoracic CT images demonstrating the imaging features of anomalous systemic arterial supply to the left lower lobe. Axial lung window image showing patchy ground-glass opacities in the left lower lobe, indicating abnormal parenchymal hyperperfusion (yellow arrow) (a); axial mediastinal window image demonstrating the origin of an aberrant systemic arterial branch arising from the descending thoracic aorta (dark blue arrow) (b); axial mediastinal window image depicting the course of the dilated anomalous systemic arterial branch supplying the left lower lobe (c); axial mediastinal window image showing a mildly ectatic inferior pulmonary vein (purple arrow) and the normally coursing lower lobe bronchial branch (white arrow) (d).

DISCUSSION

A total of 57 cases of anomalous systemic arterial supply to the left lower lobe have been reported to date, most of which involved the basal segments and received arterial supply from the descending thoracic aorta (3,11). This anatomic pattern is consistent with the findings of Yamanaka et al. (11), whose 15-patient series demonstrated uniform involvement of the basal segments, with aberrant arteries arising exclusively from the descending thoracic aorta. Yan et al. (12) evaluated 23 patients and reported that hemoptysis was the most frequent presenting symptom, occurring in approximately one-third of cases, while all patients exhibited a large-caliber systemic artery arising from the descending thoracic aorta. Wu et al. (10) emphasized the diagnostic superiority of three-dimensional CT angiography, demonstrating its ability to clearly depict the origin, course, and branching pattern of the aberrant systemic artery and to guide preoperative

planning. Most reports in the Western literature consist of isolated case reports, which consistently demonstrate preserved bronchial communication, normal pulmonary parenchyma, and venous drainage via the pulmonary veins (1-5,13,14). These findings suggest that systemic arterialization can occur in the absence of pulmonary sequestration and underscore the diagnostic importance of CT angiography (14).

Imaging plays a central role in the diagnosis of anomalous systemic arterial supply to the left lower lobe (10). Multidetector CT angiography (MDCTA) is considered the gold standard, as it provides high spatial resolution for delineating the origin, course, and caliber of the aberrant artery, as well as pulmonary venous drainage patterns (10-12). Three-dimensional reformatted and multiplanar reconstruction images clearly demonstrate the relationship between the aberrant artery and the adjacent parenchyma, offering valuable anatomic information for surgical or endovascular planning (14,15). Cardiac MRI and contrast-enhanced MR angiography are valuable complementary modalities, especially in young or asymptomatic patients, as they allow noninvasive evaluation of right heart volumes, shunt quantification (Qp/Qs), and potential volume overload without radiation exposure (12).

The main differential diagnosis includes pulmonary sequestration, pulmonary arteriovenous malformation, and bronchopulmonary foregut malformations. Compared with pulmonary sequestration, anomalous systemic arterial supply to the left lower lobe demonstrates normal bronchial distribution, absence of cystic or dysplastic parenchyma, a dilated left inferior pulmonary vein, and an enlarged anomalous systemic artery arising from the descending thoracic aorta. Ground-glass opacities may reflect increased perfusion rather than infection or fibrosis, which are typical findings in pulmonary sequestration (1,5). Pulmonary arteriovenous malformations differ by demonstrating direct arteriovenous communication with early venous filling on contrast-enhanced computed tomography, whereas anomalous systemic arterial supply maintains an intervening capillary bed between the arterial and venous systems (9). Bronchopulmonary foregut malformations may mimic vascular anomalies but usually demonstrate fistulous communication with the esophagus or stomach and are often associated with gastrointestinal symptoms (8). A comprehensive evaluation of the systemic arterial origin, bronchial communication, and pulmonary venous drainage pattern is therefore essential for accurate differential diagnosis.

Inferior pulmonary vein ectasia constitutes an important ancillary imaging feature in anomalous systemic arterial supply to the left lower lobe and reflects chronic exposure of the pulmonary venous system to high-pressure systemic arterial inflow. In their CT-based comparative study, Qin et al. (16) demonstrated that dilatation of the inferior pulmonary vein is significantly more frequent in anomalous systemic arterial supply than in pulmonary sequestration, in which venous drainage is often abnormal or systemic and parenchymal dysplasia is typically present. This venous finding therefore supports preserved pulmonary venous return and aids in distinguishing these two entities. Similarly, Miyake et al. (17) described characteristic CT features of systemic arterial supply to normal basal lung segments and emphasized that enlargement of the pulmonary veins represents a secondary hemodynamic adaptation rather than a primary venous anomaly. Furthermore, Do et al. (18) highlighted that careful assessment of pulmonary venous caliber and drainage patterns on multidetector CT is essential for accurate diagnosis of systemic arterial supply to the lung and for avoiding misclassification as pulmonary sequestration. Collectively, these observations indicate that inferior pulmonary vein ectasia, when evaluated together with arterial anatomy and preserved bronchial structure, provides valuable diagnostic support in anomalous systemic arterial supply to the left lower lobe.

Therapeutic management depends on the patient's symptoms, hemodynamic status, and the presence of complications. Surgical resection and endovascular embolization are the most commonly employed approaches in symptomatic cases. Yamanaka et al. (11) treated all symptomatic patients surgically and reported complete clinical recovery. Sun et al. (13) highlighted coil or vascular plug embolization as a safe and effective alternative to surgery, especially in patients with smaller aberrant arteries or those unsuitable for operative management. Recently, conservative management has gained attention for asymptomatic or mildly symptomatic patients (9,14). This strategy is generally preferred when the systemic artery is of small caliber, pulmonary venous drainage is normal, and no significant shunt is present (7,9). Overall, current evidence supports surgical or endovascular intervention for symptomatic patients and careful clinical and radiologic follow-up for asymptomatic individuals as an appropriate management approach (7,9,10).

This case illustrates a mildly symptomatic presentation of anomalous systemic arterial supply to the left lower lobe and demonstrates that detailed anatomic assessment can be reliably achieved using noninvasive imaging methods. The patient's stable course without intervention supports the view that conservative management may be a safe option in appropriately selected individuals. In asymptomatic or mildly symptomatic patients, including those with minimal hemoptysis, conservative follow-up with regular clinical and radiologic monitoring appears reasonable.

Several limitations of this case report should be acknowledged. First, as the findings are based on a single patient, the results cannot be generalized to all individuals with this rare anomaly, given the potential for significant anatomic and physiologic variations. Second, although the patient remained clinically and radiologically stable during one year of follow-up, this period may not be sufficient to fully assess the long-term risks of conservative management, such as the late development of pulmonary hypertension or high-output heart failure. Third, the conservative management strategy was not compared with surgical or endovascular treatment, precluding a direct assessment of relative efficacy in this specific case. Larger, multicenter prospective studies with extended surveillance are necessary to establish more robust, evidence-based criteria for selecting candidates for conservative versus interventional management.

In conclusion, anomalous systemic arterial supply to the left lower lobe is a rare vascular anomaly that may be misidentified as pulmonary sequestration but can be accurately differentiated through detailed radiologic evaluation. CT angiography remains the primary modality for assessing the origin, course, and venous drainage of the aberrant vessel. Careful interpretation of noninvasive imaging findings helps prevent unnecessary surgical intervention and supports safe conservative management in suitable patients.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - A.A., B.Y.K.; Planning and Design - A.A., B.Y.K.; Supervision - A.A., B.Y.K.; Funding -; Materials - A.A., B.Y.K.; Data Collection and/or Processing - A.A.; Analysis and/or Interpretation - A.A.; Literature Review - A.A.; Writing - A.A., B.Y.K.; Critical Review - A.A., B.Y.K.

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