

A Rare Co-Existence of Aspergillosis and Pulmonary Hydatid Cyst in Immunocompetant Adults: Two Rare Case Reports and Review of the Literature

İmmünkompetan Yetişkinlerde Nadir Bir Aspergillozis ve Pulmoner Kist Hidatik Birlikteliği: Nadir İki Olgu Sunumu ve Literatürün Gözden Geçirilmesi

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ABSTRACT *Aspergillus* is a saprophytic fungus usually colonizing in pre-existing structural or infectious lung cavities. Mixed infection of *Aspergillus* and hydatid cyst in immunocompetant patients is rarely seen. We report two cases in 69 and 53 years old with mixed infection of *Aspergillus* and ruptured hydatid cyst in lung with review of the literature. Patient 1 presented with chest pain, cough and hemoptysis. Radiology revealed a cystic lesion in right upper lung. Patient 2 presented with cough and his radiology showed bilateral cystic lung lesions. Both patients underwent cystectomy and capping procedures. Herein we aimed to underline the importance of careful histopathologic evaluation of hydatid cystectomy specimens in terms of *Aspergillus* colonization to avoid from possible fatal complications of this fungus, by pathologists.

Key Words: *Aspergillus*; coinfection; echinococcosis, pulmonary

ÖZET *Aspergillus*, sıklıkla önceden var olan yapısal veya enfeksiyöz akciğer kavitelerinde kolonize çoğalan saprofitik mantardır. *Aspergillus* ve kist hidatiğin mikst enfeksiyonu immünkompetan bireylerde oldukça nadirdir. Bu çalışmada, rüptüre kist hidatik üzerinde *Aspergillus* kolonizasyonu gösteren 69 ve 53 yaşlarında iki olgu sunulmuş ve literatürde yayınlanmış olgular gözden geçirilmiştir. Olgu 1, göğüs ağrısı, öksürük ve hemoptizi şikâyetleri ile başvurmuş ve radyolojik olarak sağ akciğer üst lobda kist saptanmıştır. Olgu 2, öksürük şikâyeti ile başvurmuş ve radyolojik olarak bilateral akciğerlerinde kistik lezyonlar saptanmıştır. Her iki olguya kistektomi ve kapitonaj cerrahi prosedürü uygulanmıştır. Kist hidatik ameliyat materyallerinin patoloğlar tarafından histopatolojik incelenmesinde, *Aspergillus* kolonizasyonu yönünden dikkatle değerlendirilmesi, hastaları olası ölümcül komplikasyonlardan korumak adına önem taşımaktadır.

Anahtar Kelimeler: *Aspergillus*; koenfeksiyon; ekinokokkozis, pulmoner

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Hydatic cyst is a zoonotic disease, caused by *Echinococcus granulosus*, which be endemically seen in Turkey as well as in other Mediterranean countries.¹ The infection is usually asymptomatic, and the patients may become symptomatic by spontaneous rupture of pulmonary hydatid cysts to bronchial branches and pleural spaces or as a result of trauma. Aspergilloma, caused by *Aspergillus* species, usually complicates preexisting cavities such as structural lung defects, post-tuberculosis cavity, sarcoidosis, bronchial cycts, bullae and neoplasms.² Mixed infection of non-operated hydatid cyst and *Aspergillus* is very rare in the literature.

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There are 14 cases without a predisposing factor in the literature to our knowledge. It is important to recognize this coexistence, to avoid the patients from possible fatal complications depending on their immunocompetency. The pathologists should search this fungus, while evaluating hydatid cystectomy specimens because of invasion potential to surrounding lung parenchyma especially in patients with immune or systemic disorders. Herein, we report two immunocompetent patients with mixed infection of *Aspergillus* and ruptured hydatid cyst in lung with clinical, radiologic and histopathologic findings.

CASE REPORTS

CASE REPORT 1

A 69 year-old female referred to Chest Diseases Department of our institute with chest pain, cough and hemoptysis. She was immunocompetent and did not give any operation or systemic disease history. Chest X-ray graphics revealed cystic lesion in the right upper lobe. The patient underwent cystectomy and capping procedure. Histopathologically, laminated ectocyst wall samples were observed. Ectocyst wall was infiltrated by septate hyphae (Figures 1,2). The hyphae were branched with narrow angulations resembling *Aspergillus*. Histochemically, these hyphae were visualized better, by periodic-acid-schiff (PAS) and Grocott's

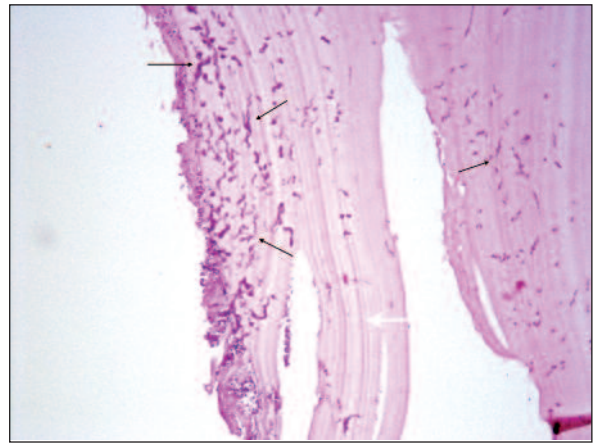


FIGURE 1: Laminated wall of the hydatid cyst (white arrow) is infiltrated by fungal pathogens (black arrows) (Hematoxylin and Eosin, x100).

methenamine silver (GMS) (Figure 3). The case was diagnosed as “mixed-infection of hydatid cyst and *Aspergillus*”. She did not receive any antiparasitic or antifungal therapy, because she did not have any predisposing factor. The patient is doing well following 14 months after the operation.

CASE REPORT 2

A 53 year-old male referred to Chest Diseases Department of our institute with cough complaint. He did not have a systemic disease or any operation history. Computed tomography revealed bilateral cystic lesions in the lung. In the posterior-basal segment of the right lung, five cm diameter cystic le-

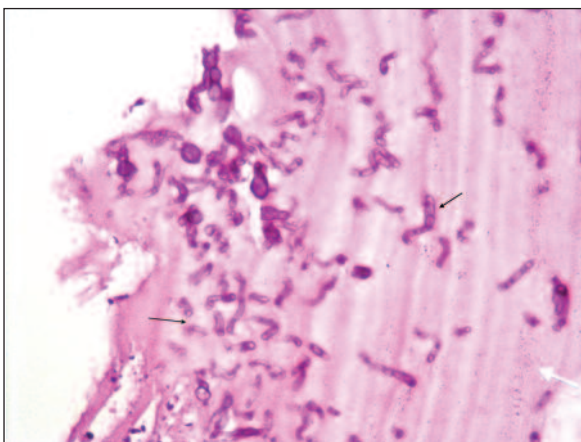


FIGURE 2: Septate and narrow angulated hyphae similar with *Aspergillus* (black arrow) are colonized on the laminated wall of hydatid cyst (white arrow) (Hematoxylin and Eosin, x400).

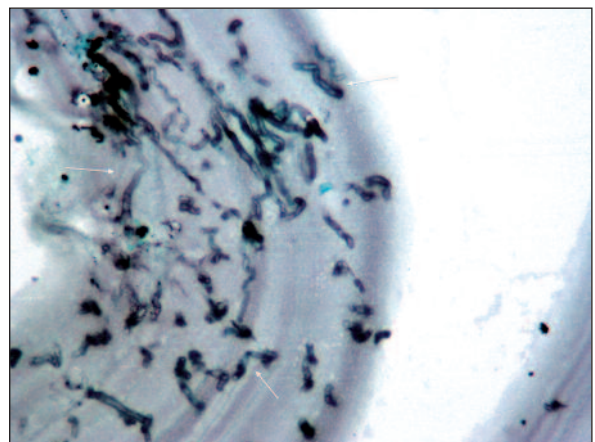


FIGURE 3: Hyphae are detailed better by Grocott's methenamine silver (GMS) (Histochemistry, x400).

sion showed air loculations and pleural thickening nearby. In the apical segment of the left lung, four cm diameter lesion with similar characteristics and additional satellite nodules around this lesion were present. All these radiologic findings were compatible with bilateral ruptured pulmonary hydatid cyst. The patient underwent bilateral thoracotomy with cystectomy and capping. Light microscopic evaluation revealed typical laminated ectocyst wall samples infiltrated by fungal hyphae consistent with *Aspergillus*. Histochemically, PAS and GMS were performed. The case was reported as “mixed-infection of hydatid cyst and *Aspergillus*”. He did not receive antiparasitic or antifungal therapy, because he did not have any predisposing factor and postoperative radiologic and clinical findings were did not indicate an invasive infection. He is doing well following nine months after the operation.

Unfortunately, we were not able to identify the species of *Aspergillus* by a reliable method in both cases.

DISCUSSION

Daily exposure to airborne *Aspergillus* is an inescapable fact of life. Therefore, either structural pulmonary or immunological deficiency allows this rapidly growing filamentous fungus to infect localized areas of lung.³ Pulmonary aspergilloma usually complicates post-tuberculosis cavity. In a study, this ratio was reported as 69% of a series of 89 cases of aspergilloma.⁴ Ruptured hydatid cyst may become infected with bacteria or saprophytic or invasive fungi. The incidence of *Aspergillus* colonization in hydatid cyst has variable ratios like 33% and 2% by Vaideswar et al. and Kocer et al., respectively.^{1,5} This difference may be associated with the limited number of the coexistence of hydatid cyst and *Aspergillus*. To our knowledge, 22 cases were reported in the literature.^{1,2,6-17} Demographic and clinical features of the reported cases were summarized in Table 1.

Five out of the 22 reported cases were detected after echinococcal cystectomy operation.^{5,6,8} This is an expectable finding hence *Aspergillus* likes to col-

onize in post-infection cavities. The rest 17 cases were mixed infections without an operation history as in our two cases (Table 1). The pathogenesis of this coexistence is unknown. Yet, it has been proposed that cyst cavity needs to be in contact with the airways. Some authors suggest that fungi can invade outer aspect of laminated membrane and microorganisms settle into the cavity.^{5,7} All cases reported in the literature were immunocompetent, except for three cases with diabetes mellitus (DM).^{7,8,10} DM is generally a predisposing factor for pulmonary mycotic infections. Additionally, two of these three cases had previously treated tuberculosis history, which was a predisposing factor.^{8,10} Our both cases were immunocompetent and did not have any systemic disease (including DM) or pulmonary operation history, in consistent with 14 cases without any defined risk factors as also presented in Table 1. Ages of the 14 cases were ranged between 15-56 year-old, and twelve of these 14 cases were localized in right lung (Table 1). Our cases were 69 and 53 year-old and both were localized in right lung (Patient 2 was bilateral).

Surgery is preferred in pulmonary hydatid cyst treatment. To avoid from potentially secondary colonization, obliteration of residual cavity (capping) must be added after endocystectomy.⁸ Although co-infection has been established as an incidental finding in most patients, a high index of suspicion is needed to predict the superimposed *Aspergillus* hence they can cause invasive diseases and fatal progress especially in immunosuppressed patients. Both cases reported in this study were immunocompetent and were incidentally determined in hydatid cystectomy specimens. The patients did not allow any additional antifungal therapy and both were healthy after operation. Early diagnosis and treatment with antifungal drugs is important to prevent life-threatening complications in especially immunosuppressed patients.

Definitive diagnosis would be by histopathology in incidental and asymptomatic patients. Ruptured hydatid cysts are particularly under risk of this coexistence. Histochemical stains like PAS and GMS are inexpensive and easy techniques to visualize *Aspergillus*. In summary, adequate and correct sam-

TABLE 1: Clinical findings of the reported cases in the literature.

Reference	Age (Year)	Gender	Immun deficiency or systemic disease	Localization	Pre-existing pulmonary operation	Surgery	Symptoms
Aydemir et al. ⁶ (2006)	32	F	None	RUL	H/CY	CY	Cough
Gupta et al. ⁷ (2006)	53	M	DM	RUL	None	N/A	Cough, fever, hemoptysis
Vasquez et al. ⁸ (2008)	1. 41	1. F	1. None	1. LUL	1. H/CY	1. lobectomy	1. Hemoptysis, cough, fever
	2. 42	2. M	2. None	2. RL	2. H/CY	2. RP	2. Cough, bloody sputum
	3. 63	3. F	3. DM+previous tbc	3. RL	3. None	3. CY+CP	3. Cough, bloody sputum
Monzoor et al. ⁹ (2008)	40	M	None	Apical segment of RLL	None	W/R	Fever, sweating
Bal et al. ² (2008)	52	F	None	RML	None	CY	Chest pain, cough, hemoptysis, shortness of breath
Kocer et al. ¹ (2008)	1. N/A	1. N/A	1. None	1. RUL	1. None	1. CY+CP	1. Cough, hemoptysis
	2. N/A	2. N/A	2. None	2. N/A	2. None	2. CY+CP	2. Chest pain, fever
Vaideswaar et al. ⁵ (2009)	1. 36	1. M	1. None	1. RLL	1. H/CY	1. CY+CP	1. Cough, hemoptysis, chest pain
	2. 25	2. M	2. None	2. RUL	2. H/CY	2. RUL lobectomy	2. Chest pain, hemoptysis
Nabi et al. ¹⁰ (2009)	50	M	DM+ previous tbc	RLL	None	W/R	Hemoptysis
M'saad et al. ¹¹ (2010)	1. 52	1. F	1. None	1. RUL	1. None	1. W/R	1. Hemoptysis, chest pain, fever
	2. 56	2. M	2. None	2. RLL	2. None	2. RLL lobectomy	2. Hemoptysis
Garcia et al. ¹² (2010)	1. N/A	1. N/A	1. N/A	1. N/A	1. None	1. N/A	1. Asymptomatic
	2. N/A	2. N/A	2. N/A	2. N/A	2. None	2. N/A	2. Asymptomatic
	3. N/A	3. N/A	3. N/A	3. N/A	3. None	3. N/A	3. Asymptomatic
Mahmood et al. ¹³ (2011)	27	M	None	Lingular lobectomy	None	LL	Fever, cough
Gürel et al. ¹⁴ (2011)	36	M	None	LLL	None	CY	Hemoptysis
Pan et al. ¹⁵ (2013)	15	M	None	RLL	None	W/R	Cough, hemoptysis
Agarwal et al. ¹⁶ (2013)	45	F	None	RLL	None	CY	Cough
Tiwari et al. ¹⁷ (2013)	42	M	None	Apical segment of RLL	None	Lobectomy	Cough, hemoptysis
Present report	1. 69	1. F	1. None	1. RUL	1. None	1. CY+CP	1. Cough, hemoptysis
	2. 53	2. M	2. None	2. RLL, LLL	2. None	2. CY+CP	2. Cough

N/A: Not available; CY: Cystectomy; CP: Cappitonage; H/C: Hydatid cystectomy; W/R: Wedge resection; RUL: Right upper lobe; LUL: Left upper lobe; RL: Right lobe; RLL: Right lower lobe; LLL: Left lower lobe; RP: Right pneumonectomy; DM: Diabetes mellitus, F: Female; M: Male; tbc: Tuberculosis.

pling and keeping this mixed infection in mind while evaluating pulmonary ruptured hydatid cystectomy specimens is important for management.

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