ISSN 1023-9855

胸腔醫學

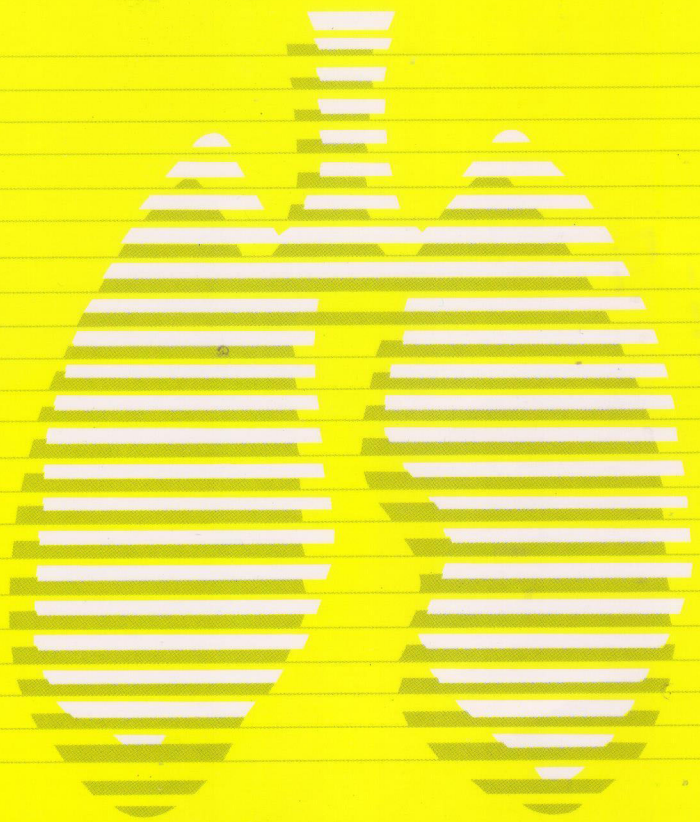
Thoracic Medicine

The Official Journal of Taiwan Society of Pulmonary and Critical Care Medicine

Vol.26 No.2 Apr. 2011

第二十六卷 第二期

中華民國一〇〇年四月



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ISSN 1023-9855

中華民國一〇〇年四月　第二十六卷　第二期

ISSN 1023－9855

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原著

病例報告

中華民國一〇〇年四月　第二十六卷　第二期

ISSN 1023－9855

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胸腔醫學

Thoracic Medicine

The Official Journal of Taiwan Society of Pulmonary and Critical Care Medicine

Orginial Articles

Case Reports

Case Fatality Rate of Tuberculosis Patients in a Community Hospital in Taiwan, 2003-2008

Yun-Hsiang Chan, Chin-Hui Yang*, Kuan-Jung Chen

Background: This study aimed to identify the possible risk factors associated with the fatality rate of tuberculosis (TB) patients in a community hospital in Taiwan.

Methods: This is a retrospective review of 223 bacteriology-confirmed TB cases in the Ren-Ai Branch of Taipei City Hospital, Taiwan from 2003 to 2008. Demographic and clinical data, treatment outcomes, and HIV testing information were collected through medical records. Cox regression analysis was used to determine independent risk factors for death.

Results: Over one-half of the patients were aged >65 years. The overall fatality rate was 29.6% (66 cases), of which only 21.2% were TB-related. DM was the most frequent co-morbidity, but liver cirrhosis had the highest fatality. Old age and liver cirrhosis were independent risk factors for fatality in TB patients. Only 28 patients (12.6%) had HIV testing and 3 new HIV infections were identified.

Conclusions: The high proportion of co-morbidity among the elderly TB patients increased the risk of fatality. Around 80% of deaths were non-TB-related. Improved clinical management and prevention strategies for high-risk populations are important to reduce TB fatality. The frequency of HIV testing among TB patients is relatively low and more aggressive promotion is necessary. ***(Thorac Med 2011; 26: 62-72)***

Key words: tuberculosis, fatality, HIV testing, liver cirrhosis, elderly

Introduction

Tuberculosis (TB) is a major public health problem worldwide [1]. The World Health Organization (WHO) estimates that 9.27 million new cases and 13.7 million prevalent cases of TB occurred in 2007 globally [2]. Although the incidence rate has declined gradually, to <1% per year, TB remains 1 of the most common communicable diseases in developing countries. Persistently among the top 10 leading causes of death globally, TB was responsible for 1.8 million deaths in 2007. There is higher mortality among TB patients: it is nearly 6.1-10 times that of the general population [3-4].

For years, TB has had the highest incidence and the most deaths of all communicable diseases in Taiwan. According to the Taiwan Tu-

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berculosis Control Report, there were 16,093 newly reported cases of TB in 2006, and the incidence rate was 67.4 per 100,000 population, which ranked in the median level globally [5-6]. In addition, TB has been the leading killer among all communicable diseases in Taiwan for many years. In 1947, the TB mortality rate in Taiwan was 294 deaths per 100,000 population. It has declined annually since then and was as low as 3.6 per 100,000 population in 2006 [5, 7]. With 832 fatalities, TB still accounted for 0.6% of all deaths in 2006 and ranked 13th as a leading cause of death. All-cause mortality among pulmonary TB patients in Taiwan was 8.7 times greater than that of the age- and sex-matched general population [8]. According to the Taiwan Centers for Disease Control's (CDC) TB Control Report, the overall treatment success rate was 69.2% in the 2005 cohort after a 12-month follow-up, with a high fatality rate of 19.6% [5].

Several risk factors, like age [9-12], human immuno-deficiency virus (HIV) infection [13-14], diabetes mellitus (DM) [15-16], liver cirrhosis [17-18], and renal failure [8, 19-20], are important in predicting the outcome of TB treatment. Although Taiwan has managed to reduce its TB fatality rate, with a 16% reduction in 2006 compared to the previous year, it is still higher than in many Western countries. A possible reason may be the aging population in Taiwan, and that over half of the TB patients and 80% of the TB fatality cases are those aged >65 years. Compared with Japan (11% fatality rate) and Singapore (14% fatality rate), where the demographic structures are similar to those in Taiwan [21], the fatality rate in Taiwan is still higher. The present study aimed to identify possible risk factors associated with fatality in patients diagnosed with and treated for TB in a community hospital in Taiwan during the period 2003-2008.

Materials and Methods

This study was conducted at the Ren-Ai Branch of Taipei City Hospital, which is a 600-bed community hospital providing full-service, and total health care services to more than 500,000 residents in the Da-An District of Taipei City. The hospital's institutional review board approved the study. However, written or inform consent from the participants was not obtained due to the retrospective design. Nonetheless, the results excluded all personal identities to protect personal confidentiality.

Using the hospital medical records International Classification of Diseases coding system and the Taiwan National Tuberculosis Registry Campaign in the Taiwan CDC, 442 patients with a diagnosis of suspected TB were identified from 1 January 2003 to 31 December 2008. A trained nurse case manager retrospectively reviewed all of the medical and microbiologic records of these patients using a structured questionnaire to obtain clinical information, which was then reviewed by a physician investigator. Bacteriology-confirmed TB patients were included based on the following criteria: culture of *Mycobacterium tuberculosis* (MTB) or acid-fast bacilli present in sputum and excluding non-TB mycobacteria infection or tissue samples from a site other than the lungs. Risk factors for TB were recorded in the medical records, and included co-morbidities such as DM, malignancy, liver cirrhosis, end-stage renal disease (ESRD), HIV infection, and long-term glucosteroid or immuno-suppressive drug use. HIV antibody screening tests during TB diagnosis and treatment were also recorded.

All patients were followed up from the time

of TB diagnosis until death or treatment completion, or 31 December 2009, whichever came first. Treatment outcomes were assessed at the end and categorized according to WHO recommendations. "Treatment success" was defined as "cured" for patients who became sputum culture-negative for MTB during the last month of treatment and on at least 1 previous occasion, or when treatment was completed, for those patients who completed treatment but did not meet the classification criteria for "cured" or "failed". "Death" was defined as a patient who died for any reason during the course of treatment. A physician examined the cause of death based on information from the medical records and death certificates. TB-related death was verified based on the death certificate (TB listed as the cause of death), medical chart review (TB identified as cause of death), and active TB at the time of death (positive AFB smear after last treatment or during treatment if the patient did not complete treatment ≤6 months before death). A patient's death was attributed to TB if 2 or more of these conditions were met.

Statistical analyses were performed using SPSS 14.0 statistical software. Categorical variables were compared using χ^2 tests or Fisher's exact tests, while continuous variables (means) were compared using Student's *t*-tests. Survival analyses included Kaplan-Meier curves and the Cox proportional hazards model for death due to any cause, which were repeated for TB-related deaths using the observation period from TB diagnosis as a reference time. Variables were entered into the models according to their statistical significance in univariate analysis and their biological relevance, using stepwise selection based on maximum partial likelihood estimates ($p>0.10$ for removal and $p<0.05$ for entry of variables). All variables with $p<0.05$ were retained in the final multivariate model. Results were expressed as hazard ratios (HR) with 95% confidence intervals [CI].

Results

The demographic characteristics of the 223 bacteriology-proven TB cases included in this study are shown in Table 1. The case number remained stable during the study period. Their mean age was 65.5 ± 21 years (range, 17-95 years) and 60% of them were over 65 years old. Only 35 cases were covered by the direct observation treatment (DOTS) program implemented by the Taiwan CDC. Among the 177 positive MTB culture results, 87% were pan-susceptible MTB strains, 11.3% had drug resistance, and 1.7% was multi-drug resistant MTB. Elderly patients (≥65 years old) had a higher fatality rate than younger (<65 years) patients (27.6% vs. 12.5%, $p<0.001$). DM was the most common co-morbidity (27% of patients), but liver cirrhosis had the highest case fatality rate, though a majority were non-TB-related.

One hundred fifty-four (69.1%) patients completed the anti-TB therapy and were considered cured; 66 (29.6%) patients died, 2 (0.9%) defaulted, and 1 (0.4%) was still on treatment. The mean duration of anti-TB treatment was 267 days (range, 172-772 days) among the completely treated cases. Nineteen (12.3%) cases were treated for over 1 year with a rifampicin-sparing regimen, most commonly due to drug side effects or drug resistance and extra-pulmonary involvement. The average duration from TB diagnosis to death of the 66 deaths was 67 ± 69 days (range, 0-248 days), and only 16 of the 66 deaths (24.2%) were interpreted as TB-related (Table 1). Twenty-two (33.3%) of the 66 deaths occurred within 14 days after TB

Table 1. Demographic information of confirmed tuberculosis patients (n=223)

	Total (N=223)	Death (N=66)		TB-related death (N=16)	
	n (%)	n (%)	fatality rate[+]	n (%)	fatality rate[+]
TB diagnosis Year					
2003~2004	87 (39.0)	16 (24.2)	18.4%	3 (18.8)	3.4%
2005~2006	64 (28.7)	23 (34.8)	35.9%	7 (43.8)	10.9%
2007~2008	72 (32.3)	27 (40.9)	37.5%	6 (37.4)	8.3%
Age at TB diagnosis, mean {range}years	65.5 {17-95}	77.9 {30-95}		82.8 {58-95}	
< 45	44 (19.7)	1 (1.5)	2.3%	0	0
45~64	44 (19.7)	10 (15.1)	22.7%	1 (6.2)	2.3%
≥ 65	135 (60.5)	55 (83.3)	27.6%	15 (93.8)	11.1%
Sex					
Male	162 (72.6)	51 (77.2)	31.5%	12 (75)	7.4%
Female	61 (27.4)	15 (22.7)	24.6%	4 (25)	6.6%
Localization of TB					
Pulmonary	208 (93.3)	59 (89.4)	28.4%	15 (93.8)	7.2%
Pulmonary + extrapulmonary	4 (1.8)	1 (1.5)	25%	1 (6.2)	25%
Extrapulmonary only	11 (4.9)	6 (9.1)	54.5%	0	0
Bacteriology findings: acid-fast smear					
Negative	76 (34.1)	22 (33.3)	28.9%	6 (37.5)	7.9%
Positive	134 (60.1)	39 (59.1)	29.1%	9 (56.3)	6.7%
Not performed	13 (5.8)	5 (7.6)	38.4%	1 (6.2)	7.7%
Bacteriology findings : *Mycobacteria tuberculosis* culture					
Negative	24 (10.8)	13 (19.7)	54.2%	0	0
Positive	177 (79.4)	47 (71.2)	26.6%	15 (93.8)	8.5%
Not performed	22 (9.9)	6 (9.1)	27.3%	1 (6.2)	4.5%
Comorbidities					
Malignancy	36 (16.1)	22 (33.3)	61.1%	3 (18.8)	8.3%
Diabetes mellitus	60 (26.9)	18 (27.3)	30.0%	4 (25)	6.7%
Liver cirrhosis	12 (5.4)	10 (15.2)	83.3%	2 (12.5)	16.7%
End-stage renal disease.	17 (7.6)	9 (13.6)	52.9%	2 (12.5)	11.8%
Others[#]	2 (0.5)	0 (0)	0%	0	0
No comorbidity	110 (49.3)	18 (27.3)	13.4%	5 (31.3)	4.5%
≥ 1 specific condition	113 (50.7)	48 (72.7)	42.5%	11 (68.7)	9.7%
HIV status					
Negative	25 (11.2)	8 (12.1)	32.0%	1 (6.2)	4%
Positive	9 (4.0)	1 (1.5)	11.1%	0	0
Not performed	189 (84.8)	59 (89.4)	31.2%	15 (93.8)	7.9%

[+] Fatality rate (%) was calculated by total case number divided by death number

[#] One case had systemic lupus erythematosus and the other had rheumatoid arthritis, both on long-term glucosteroid medication

diagnosis, and 10 occurred before TB diagnosis (culture-confirmed post-mortem). Death within 90 days accounted for 68%, and 6 patients died 180 days or more after starting treatment.

The demographic data and combined co-morbidity status were not significantly different between the early fatality cases and other fatalities, and between TB-related or non-TB-related

deaths. Among the non-TB-related deaths, infections (e.g., pneumonia due to pathogens other than MTB, spontaneous bacterial peritonitis, etc.) were the most common cause of death, accounting for over half, followed by other medical conditions such as myocardial infarction and massive gastrointestinal bleeding and malignancy.

Factors associated with TB patients who died from any cause during the observation period after TB diagnosis, were first analyzed. Using univariate analyses, increasing age, negative sputum MTB culture, and patients with concomitant malignancy, COPD, liver cirrhosis and ESRD were associated with increased fatality. Two factors remained significantly associated with death in the multivariate analysis: increased age (adjusted HR [aHR] = 1.6 per 10 years, 95% CI 1.3-2.0) and liver cirrhosis (aHR = 5.8, 95% CI 2.6-12.8) (Table 2; Figures 1). Separate analysis for TB-related deaths revealed similar results, although patients with malignancy also showed an increased risk for TB-related death.

Only 28 (12.6%) TB patients received HIV testing and only 3 were identified as new HIV-infected cases. With the 6 known HIV-infected cases, there were 9 HIV-TB co-infections in all in this cohort. The most common reason for HIV testing was renal failure and pre-hemodialysis examinations (19 cases). Other reasons included regular health exam or being at risk for HIV infection, through behavior such as drug addiction or unsafe sex. The mean age of the patients who received HIV testing was significantly younger than the mean age of those not tested (53 vs. 68 years, $p<0.01$).

Discussion

This study shows a very high mortality rate (up to 29.6%) for TB patients, with old age and co-morbid liver cirrhosis significantly associated with death among this group of patients. The fatality rate in this study was high compared with the Taiwan national data (19.6% of 2005 cohort) or Taipei City's data (16.8% of 2003 cohort) [5,8]. One possible explanation is the significantly older age of the study patients, as 60% of the cases were aged >65 years, com-

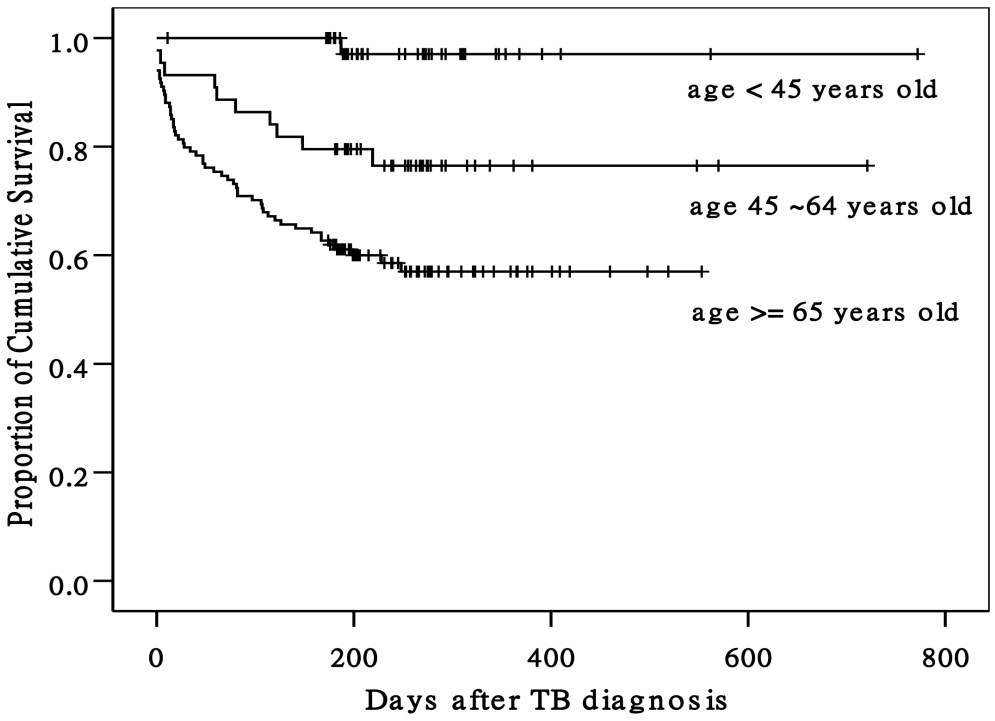
Table 2. Factors associated with death from any cause and due to tuberculosis among TB patients at Ren-Ai Hospital (2003-2008)

Variable	Total	Total deaths			TB-related deaths		
		N	Univariate HR (95% CI)	Adjusted HR (95% CI)	n	Univariate HR (95% CI)	Adjusted HR (95% CI)
Age, per 10 years			1.7 (1.4-2.0)*	1.6 (1.3-2.0)*		1.5 (1.3-1.9)*	1.5 (1.2-1.9)*
Positive MTB culture							
Negative	24	13	1	1	0	1	1
Positive	177	47	0.40 (0.22-0.75)*	0.48 (0.26-0.92)$^{+}$	15	0.26 (0.14-0.49)*	0.32 (0.16-0.63)$^{+}$
Not performed	22	6	0.41 (0.16-1.08)	0.35 (0.13-0.94)$^{+}$	1	0.27 (0.11-0.90)	0.27 (0.09-0.79)$^{+}$
Comorbidities							
Malignancy	36	22	3.1 (1.9-5.2)*		3	4.0 (2.3-7.1)*	1.9 (1.0-3.6)$^{+}$
Liver cirrhosis	12	10	5.4 (2.7-10.6)*	5.8 (2.6-12.8) *	2	6.3 (2.9-14.5)*	6.3 (2.6-15.4) *
ESRD	17	9	2.3 (1.1-4.6)$^{+}$		2	2.4 (1.1-5.4)$^{+}$	

Abbreviations: MTB, *Mycobacteria tuberculosis*; ESRD, end-stage renal disease

*$p<0.001$; $^{+}$ $p<0.05$

(A)



(B)

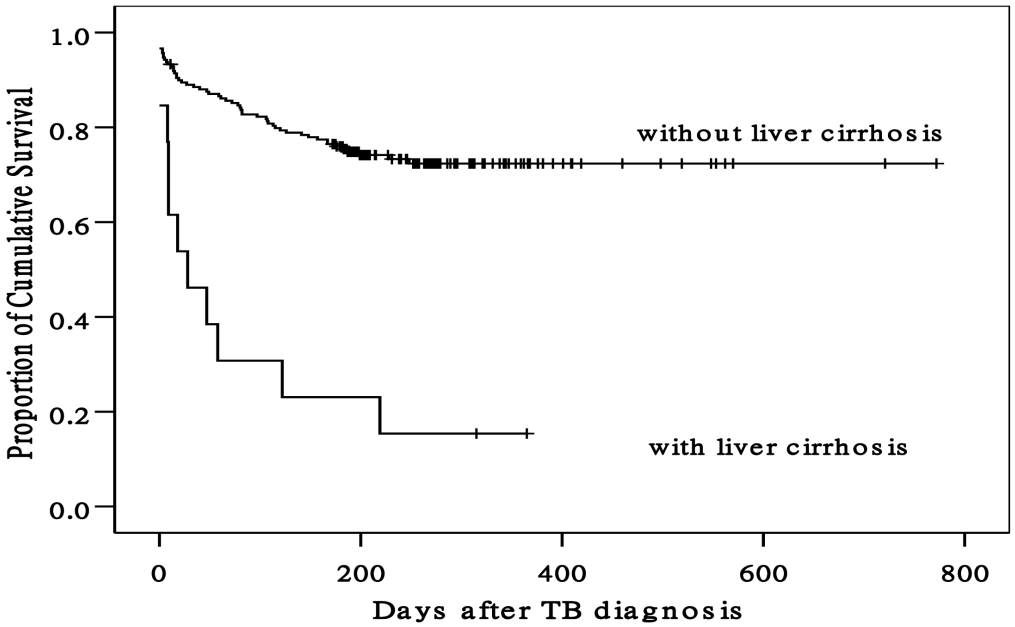


Fig. 1. Kaplan-Meier survival estimate by (A) age group and (B) liver cirrhosis, adjusted by age, gender, sputum acid-fast smear status, and other co-morbidities (includes malignancy and end-stage renal disease).

pared to 50.9% and 46% in the other 2 datasets, respectively. Previous studies have also reported that older age is a risk factor for TB fatality [9-12]. The elderly frequently have atypical clinical presentations and are prone to having a delayed TB diagnosis, which may contribute to the high mortality [12, 22]. Using multivariate analysis, our data confirmed that the risk of death increased 1.7 times per 10-year increase in age among TB patients.

Despite the early fatality (death within 2 weeks after TB diagnosis), there was no difference in age distribution when compared with other fatality cases. Possible causes for early TB fatality include advanced disease, late diagnosis, and delayed treatment [23-24]. The high proportion of concomitant co-morbidities among the study patients may also have contributed to the high fatality rate. Around 80% had been hospitalized and over half had one or more co-morbidities, implying that the study population had a poorer general condition, which resulted in a higher fatality rate. In contrast to the early fatality rate, 6 patients (9.1%) died after 180 days of anti-TB treatment due to non-TB-related causes. These patients are included among TB fatalities, by WHO definition, because of the longer treatment duration due to anti-TB drug side effects.

Several co-morbidities were significantly associated with TB fatality, using univariate analysis, but only liver cirrhosis demonstrated significance in the multivariate model. Liver cirrhosis patients had the highest TB fatality rate (83.3%) in this study, but only 2 were categorized as TB-related deaths. Previous studies have shown that patients with cirrhosis and TB manifest more treatment-related hepatotoxicity, making sustained TB treatment difficult. Nonetheless, there was no significant difference in TB fatality reported [17-18]. Another study of TB treatment outcomes in Taipei showed that hepatic disease is not an independent risk factor for death among TB patients [8]. The divergence may be due to the different severities of the hepatic diseases and liver cirrhosis. According to Sun's study, liver cirrhosis is an independent risk factor associated with hepatitis (OR = 6.0; 95% CI = 1.143-31.951), 3 times higher than in hepatitis B virus infection only [25].

Two-thirds of the liver cirrhosis patients in our study were classified as Child's C class, which means more advanced disease [26]. All died during TB treatment: 2 were TB-related and the other 6 died due to spontaneous bacterial peritonitis (3), pneumonia (2) with sepsis, and massive bleeding (1). Thus, since liver cirrhosis patients with TB have a higher risk of fatality, appropriate and close monitoring should be instituted to prevent deaths.

Dooley's study shows that TB fatality is 6.5 times higher in patients with DM than in non-DM patients [15]. However, other studies do not support this observation [8, 16, 27]. DM was the most common co-morbidity in this study, but it was not significantly associated with death in the multivariate analysis. The small case number and lack of DM severity data limit the interpretation.

In high HIV prevalence areas, TB remains the leading cause of death among HIV-infected patients [13-14]. Nine HIV-TB co-infected patients were identified by this review, but only 1 case (11.1%) died. This fatality rate was not higher than others. In another hospital-based study in Taiwan, the outcome of HIV-infected patients with TB was not inferior to the outcome of those without TB [28]. Several reports have shown that HAART can reduce the fatality rate of HIV/TB co-infected patients [29]. The

Taiwan government offers free anti-retroviral agents for HIV-infected individuals, which may improve the prognosis of TB-HIV co-infected patients. On the other hand, the WHO recommendation of offering HIV tests for all TB patients has not been widely undertaken in Taiwan.

In a previous study by Chiang *et al*., who prospectively screened 378 patients with active pulmonary TB for HIV infection in 1996, only 1 patient (0.26%) was found to be HIV-infected [30]. However, the study was limited by the small sample size and only pulmonary TB patients were selected in the early years, when the prevalence and incidence of HIV was low in Taiwan. Kung's recent hospital-based study found 28.8% of TB patients were offered HIV testing, and the positive testing rate increased from 2.7% in 2001 to 8.5% in 2004 [31]. In the current study, only 13% of TB patients received HIV testing and 3 were identified (11% of cases who received HIV tests) as new HIV-infected cases. Since HIV infection is an important risk factor for TB, the lack of alertness by physicians may cost the patients the opportunity of an early diagnosis of HIV and adequate treatment.

This study has several limitations. First, it was hospital-based. Selection bias is probable and the study cohort may not represent community epidemiology in Taiwan. Second, as a retrospective cohort study, sputum samples were not systematically collected. Thus, some patients lacked smear and/or culture results. Efforts to separate the sputum examination status in the analysis using the multivariate model showed no significance. Third, since HIV is a strong risk factor for fatality among TB patients, HIV status was included in the multivariate model initially. However, more than 85% of the patients with an unknown HIV status before TB diagnosis were not provided with HIV testing; thus, the exact relationship between HIV status and TB fatality remains unknown.

In conclusion, older age and liver cirrhosis are independent risk factors for fatality among TB patients. Complicated co-morbidities among the elderly may increase the risk of death, but a high proportion of deaths among TB patients were attributable to non-TB causes. Improved clinical management and prevention strategies for the high-risk population are important. The frequency of HIV testing among TB patients is low, so strategies to increase this for all TB patients are needed in order to better assess trends in TB and HIV co-infection.

Acknowledgements

Dr. Chen KJ contributed to the conception and design of this study. Dr. Chan collected the clinical data and was involved in the drafting of the article. Dr. Yang analyzed and interpreted the data and did a critical revision of the manuscript. The authors wish to thank Chi-Fang Feng, who helped in linking with the Taiwan National TB Registry to identify the treatment outcomes for patients transferred to other hospitals.

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2003-2008 年臺灣一社區型醫院結核病人死亡之危險因子分析

詹雲翔　楊靖慧*　陳寬榮

前言：分析臺灣一社區型醫院結核病人死亡之危險因子。

方法：此為一回溯性研究，對象為台北市立聯合醫院仁愛院區自 2003 年到 2008 年間 223 位細菌學確診之結核病人，由病歷收集其背景資料、臨床病程、治療結果與HIV檢測情形。使用 Cox 回歸分析方法來決定與結核病死亡相關之危險因子。

結果：超過一半以上之病人年紀超過 65 歲。整體的結核病致死率為 29.6%（66例），其中只有 21.2% 的死亡可直接歸因於結核病相關。糖尿病是最常見的原在性疾病，但是肝硬化病人有最高之結核病致死率。各種可能的預後因子中只有病人的年齡與肝硬化對與死亡預後有明顯相關。只有 28 位病人（21%）有檢查HIV，但從其中發現了 3 個新的 HIV 感染病。

結論：有很高比例的結核病人為年老且合併各種潛在性疾病，因此增加了結核病治療的致死率，但是近八成的死因為非結核相關因素。因此，針對具有高危險因子的病人，增強臨床診治能力以及預防結核病發病的相關政策有助於降低結核病致死率。此外，針對結核病人的 HIV 檢驗普及率很低，需要有更積極的方式來推行此政策。*(胸腔醫學* ***2011; 26: 62-72)***

關鍵詞：結核病，致死率，HIV檢驗，肝硬化，年長者

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Pancreatic Pseudocyst Complicated with Acute Suppurative Mediastinitis and Neck Involvement

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Pancreatic pseudocyst is a complication of acute or chronic pancreatitis. Since 1951, only about 50 cases of mediastinal extension of pancreatic pseudocyst haves been published in the literature. However, pancreatic pseudocyst complicated with ascending mediastinitis is rarely reported. To our knowledge, this is the first case report of a patient suffering from ascending mediastinitis with extension to the neck due to a pancreatic pseudocyst. A 47-year-old male patient with alcohol-related chronic pancreatitis presented fever, dysphagia, and neck swelling for 1 week. Chest X-ray at our emergency department showed mediastinal widening with left pleural effusion. Base on the computed tomography images, pancreatic pseudocyst complicated with ascending mediastinitis and extension to the neck was confirmed. We treated the patient with video-assisted thoracoscopic surgery for mediastinitis, and used percutaneous computed tomography guided drainage for the pancreatic pseudocyst. The patient recovered and has been followed at our out-patient department. ***(Thorac Med 2011; 26: 73-79)***

Key words: pancreatic pseudocyst, acute mediastinitis

Introduction

Mediastinitis is a surgical emergency and carries a high mortality rate especially when recognized late or treated improperly [1]. Most cases of mediastinitis occur after cardiovascular or thoracic surgery [2]. Infrequently, esophageal rupture, tracheobronchial perforation, or descending extension from an oropharyngeal infection may also cause mediastinitis [3]. Reports of ascending mediastinitis due to an intra-abdominal lesion are rarely published in the literature [4]. We report our experience with a patient with ascending mediastinitis extending to the neck, due to a pancreatic pseudocyst, that resolved after percutaneous drainage of the pseudocyst through video-assisted thoracoscopic surgery (VATS) and percutaneous computed tomography (CT)-guided drainage.

Case Report

A 47-year-old male construction worker presented at our outpatient department with neck swelling and tenderness for 1 day. One week before admission, he suffered from el-

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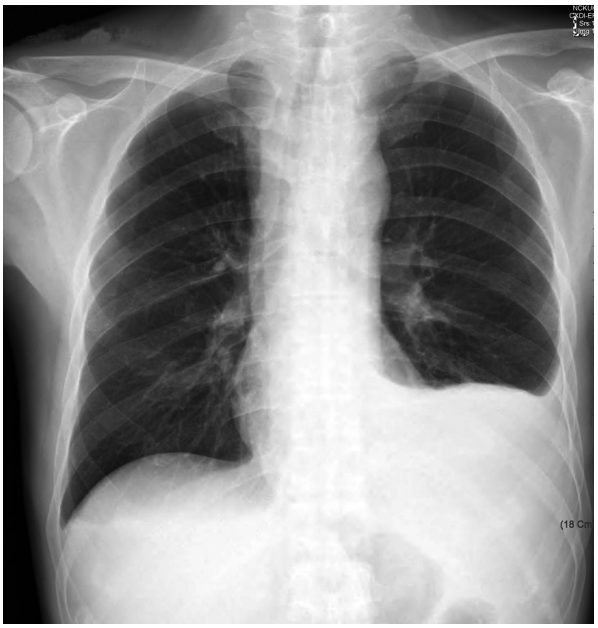


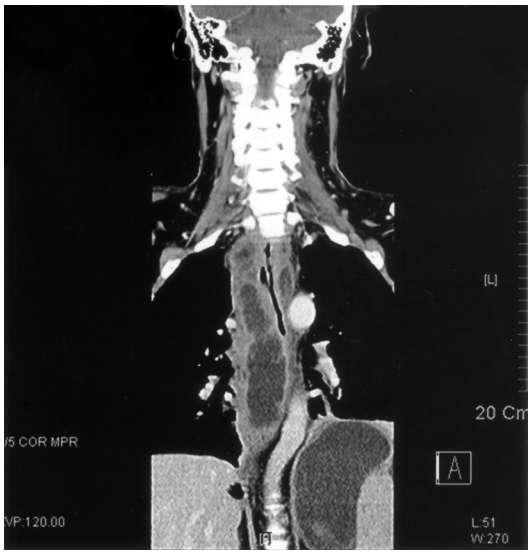
Fig. 1. Mild mediastinal widening and small amount of pleural effusion with an elevated diaphragm on the left side.

evated body temperature (37.5°C) and cough. Odynophagia and dysphagia developed in the subsequent 2 days without abdominal distress. He denied swallowing a foreign body. Mild mediastinal widening and elevated left hemidiaphragm with a small amount of pleural effusion were noted on chest X-ray (Figure 1). The otolaryngologist suspected a deep neck infection because the retropharyngeal space was widened on the neck X-ray, and flexible laryngoscopy revealed narrowing of the pharyngeal lumen. A CT scan of the neck and the mediastinum showed extensive low-density loculated fluid accumulation with peripheral enhancement in the posterior mediastinum, tracheoesophageal space, and both side of the neck (Figure 2a-1, 2a-2). The fluid collections could be traced downward to the abdomen, so we favored pancreatic pseudocysts with extension to the mediastinum through an esophageal hiatus and the neck. The CT image of the abdomen showed no obvious intraluminal calcified lesion or dilation at the pancreatic duct.

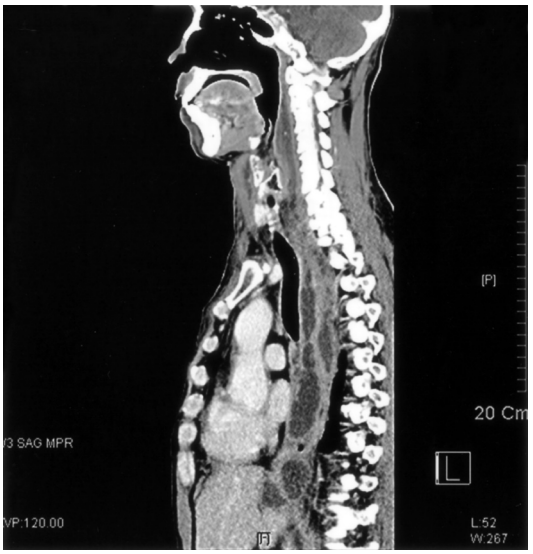
The patient had a 2-year medical history of recurrent alcoholic pancreatitis. He also was a hepatitis C virus carrier. He had been admitted several times because of recurrent pancreatitis. One and a half years ago, a pancreatic pseudocyst was detected in this patient.

At the emergency department, vital signs disclosed body temperature of 36.3°C, blood pressure of 99/78 mmHg, respiratory rate of 20 bpm, and pulse of 97 bpm. His peripheral oxygen saturation by pulse oximetry was 98% in room air. The hemogram showed a white blood count (WBC) of 24,000/μL with neutrophils: 88%. Elevated serum amylase (747 U/L) and lipase (2,276 U/L), and an increased level of C-reactive protein (267.7 mg/L) were reported. Emergency debridement and drainage of the mediastinum, bilateral neck incision and drainage, and percutaneous drainage of the pseudocyst were performed. Panendoscopy at the operation room confirmed no esophageal laceration or perforation. During the VATS procedure, profuse dark red jelly-like contents were found in the wide-opened mediastinal pleura; analysis of the contents showed amylase 34,200 U/L and lipase 253,940 U/L.

After the operation, the patient was transferred to the intensive care unit and the postoperative course was uneventful. The broad-spectrum antibiotic, flomoxef, was used empirically. Cultures of the contents from the mediastinum, neck, pseudocyst, and blood yielded no microorganisms. The pathological findings were suppurative inflammation with fibrosis in the mediastinal tissure. On post operative day 4, extubation was performed smoothly, and his blood biochemistry and hemogram on day 10 showed WBC count, 11,000 /μL; amylase, 93



(a-1)



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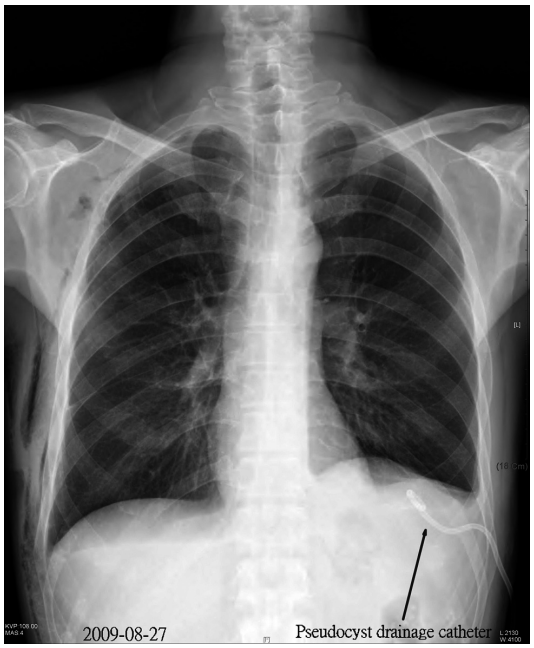

(b)

Fig. 2. Coronal and sagittal view of the mediastinum (2a-1, 2a-2), showing ascending mediastinitis with huge cyst at the left subdiaphragm due to pancreatic pseudocyst; (2b) Resolution of the mediatinal widening and elevated diaphragm after VATS and pseudocyst drainage.

U/L; and lipase, 408 U/L. On day 13, the chest tube was removed. Complete resolution of mediastinal widening and elevated left hemidiaphram was demonstrated in the follow-up chest films.

Unfortunately, the patient developed catheter-related infection after pseudocyst drainage on day 20. Bacterial culture of the contents from the pseudocyst drainage catheter yielded *Enterobacter cloacae* and *Acinetobacter* species. After the administration of piperacillin/tazobactam, his systemic inflammatory response subsided. About 1 month after the operation, the patient was discharged with a catheter in

place for drainage of the pseudocyst. Repeated chest films showed complete resolution of the patient's mediastinal widening and left elevated diaphragm (Figure 2b).

Discussion

Pseudocyst formation is a common complication of both acute and chronic pancreatitis. The complication has an approximately 10% occurrence rate after acute pancreatitis [3-4]. A pseudocyst results from necrosis of parenchymal or peripancreatic tissue with liquefaction and subsequent organization. It can communicate with the pancreatic duct or lead to complete ductal disruption and gross leakage of pancreatic juice. Pseudocysts also occur in around 20% of patients with chronic pancreatitis, often due to alcohol abuse, and are formed following acute exacerbation of pancreatitis or progressive ductal obstruction induced by ductal stricturing or an intra-ductal stone. Up to 40% of pseudocysts resolve spontaneously without intervention [5]. But invasive intervention with pseudocyst drainage becomes necessary because of complications or unrelenting symptoms associated with the pseudocyst. In addition, the traditional dictum of requiring invasive intervention for pseudocysts "longer than 6 cm or more than 6 weeks" is still a relative indicator [6-7]. With regard to drainage options, surgical management is still considered the "gold standard" of pancreatic pseudocyst treatment. However, open surgical drainage was associated with substantial morbidity and mortality (25% and 5%, respectively) [8-10]. Two additional treatment options, percutaneous catheter drainage and endoscopic drainage, have gained popularity. At present, no randomized comparative studies exist concerning which of the options should be offered to the patient as initial therapy. Percutaneous drainage was favored for those patients with normal pancreatic ducts and those with strictures and no duct-cyst communication [11]. But the main complication is drain tract infection, which occurred in one-half of patients [12]. The 2 major techniques of endoscopic drainage are transmural cystenterostomy with stent placement and transpapillary stenting. Transmural cystenterostomy is suggested for the completely obstructed pancreatic duct, and transpapillary stenting was suggested for patients with a communicating pseudocyst. Complications of endoscopic drainage include bleeding, perforation, secondary infection, and stent migration (about 11% to 37%) [13-16]. Therefore, which of these techniques should be offered to the patient as an initial therapy depends on the pancreatic ductal condition, complications of the pseudocysts, and the operator's experience.

Depending on the location and extent of the fluid collection or the presence of infection, complications can occur in up to 40% of cases, as reported in various studies [17]. Mediastinal pseudocyst is a rare complication that is caused by rupture of the pancreatic duct posteriorly into the retroperitoneal space, with pancreatic fluid tracking through the diaphragmatic hiatuses into the mediastinum [18]. Methods of successful management for mediastinal pancreatic pseudocyst have been published in many reports. Several treatment options were reported. Conservative treatment with parenteral nutrition combined with/without somatostatin or its analogue, octreotide, has been shown to improve outcome in patients with mediastinal pancreatic fluid accumulation [19-22]. Radiologic drainage with a percutaneous catheter, endoscopic stenting, external or internal drainage, and surgical

drainage should be considered for the complications or unrelenting symptoms associated with a pseudocyst. One rare complication arising from pancreatic pseudocysts was suppurative mediastinitis, also known as pancreatic enzymatic mediastinitis. This pathogenicity is caused by activated pancreatic enzyme spreading into the mediastinum following disruption either of the pancreatic duct or of the pseudocyst [23]. Most cases of mediastinitis are due to postoperative complication of cardiovascular or some thoracic surgical procedures. Esophageal perforation or descending spread of odontogenic or retropharyngeal infections is another cause of mediastinitis.

The clinical situation of ascending mediastinitis due to pancreatic pseudocyst is similar to the fulminant course of descending necrotizing mediastinitis [24]. Early diagnosis, adequate antibiotics, and early effective surgical drainage can reduce the high mortality rate from 67% to 15.4% [25]. Since Roberts first reported the efficacy of thoracoscopic drainage for descending necrotizing mediastinitis in 1997 [26], less invasive techniques, such as VATS, have become effective options for the treatment of mediastinitis by pleuromediastinal drainage with or without cervical drainage [24-25]. However, the optimal surgical approach for mediastinitis is still controversial. Our case is the first report in the literature of ascending mediastinitis with extension to the neck due to a pancreatic pseudocyst. To our knowledge, only 1 case report of mediastinitis arising from a pancreatic pseudocyst that was successfully treated by thoracoscopic drainage alone has been published. According to the previous methods of treatment for descending necrotizing mediastinitis from Cho and Roberts [25-26], and the results of surgery and percutaneous catheter drainage for sterile and infected pseudocysts are similar [27]. Therefore, in this case, we performed bilateral neck incision with drainage, mediastinal debridement with drainage by VATS, and percutaneous CT-guided drainage for the pancreatic pseudocyst. After the operation, the mediastinal widening has improved, as seen on follow-up chest radiographs and the patient began oral intake on the 11th day after operation. Although infection, the main complication of catheter drainage, occurred during the course of hospitalization, the patient recovered after adequate antibiotics treatment for sepsis. The patient was discharged in a stable condition with a pig-tail tube for pseudocyst drainage.

In summary, pancreatic pseudocyst complicated with ascending mediastinitis is a surgical emergency. Early detection and an aggressive approach are crucial in reducing the high mortality rate of acute mediastinitis. The essential elements of treatment should be a combination of surgical debridement and broad-spectrum antimicrobial agents. A minimally invasive thoracoscopic operation may have the same effect as invasive thoracotomy, and percutaneous catheter drainage is as effective as surgery for treatment of pancreatic pseudocyst.

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胰臟假囊腫引發急性化膿性縱隔腔炎合併頸部侵犯

游群翔　賴吾為*　張漢煜

胰臟假囊腫是一種急性或慢性胰臟炎所引發的併發症。從西元 1951 年，約有五十多篇文獻報導胰臟假囊腫合併侵犯縱隔腔的個案，然而，鮮少有報告胰臟假囊腫併發上升性縱隔腔炎。因此到目前，我們報告第一位因胰臟假性囊腫引發急性上升性縱膈腔炎合併侵犯頸部的個案。這位病人有慢性酒精性胰臟炎的病史，因發燒、吞嚥困難和頸部腫脹來到急診室。由胸腔 X 光片發現縱隔腔變寬且合併左側肋膜腔積液。從縱隔腔電腦斷層影像，發現有胰臟假性囊腫，並且沿著縱隔腔往上引發縱膈腔炎和侵犯頸部。此病患接受胸腔鏡手術，頸部切開引流和胰臟假囊腫電腦斷層指引經皮引流術，目前此病人已恢復健康並於門診追蹤。*(胸腔醫學* ***2011; 26: 73-79)***

關鍵詞：胰臟炎，縱隔腔腫瘤

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Impending Asphyxia Caused by Migration of Tracheal Expandible Stent: An Unexpected Complication

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The use of covered self-expanding metallic stents (SEMS) for malignant tracheo-esophageal fistula can lead to complications. We report such a case, in which local migration of a tracheal stent caused it to erode through the trachea and invade the esophagus, leading to compromise of the airway. This situation was finally resolved by emergency tracheostomy, thereby saving the patient. The stent was retrieved and a long tracheostomy tube was put in place so as to bypass the fistula. Similar complications could not be found in the literature. ***(Thorac Med 2011; 26: 80-84)***

Key words: tracheal stent, malignant tracheo-esophageal fistula, migration

Introduction

Malignant tracheo-esophageal (T-E) fistula, a serious complication of esophageal cancer, can aggravate the patient's underlying condition and even cause death due to repeated aspiration and pneumonia attacks [1]. The treatment options include chemotherapy, irradiation, bypass surgery, esophageal exclusion and placement of a covered self-expanding metallic stent (SEMS); the latter is the most commonly used [2]. Choosing from among the above options must depend on the patient's physical status. Most patients receive palliative placement of a SEMS in the trachea or esophagus because of technical feasibility, unresectability of the tumor or unfitness for surgery. Possible complications after the SEMS placement are granulation, epitheliation, migration and perforation [3]. These may induce severe airway symptoms, including dyspnea, stridor, airway obstruction, and finally death. Perforation is caused by the force of the stent's expansion, which is sufficient to erode through the wall [3]. We herein report a case in which the tracheal stent continued to erode into the esophageal wall, with the proximal end eroding into the lumen. This caused severe airway compromise the membranous portion of the tracheal SEMS.

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Case Report

A 74-year-old male who was a heavy smoker and an alcoholic suffered from progressive dysphagia for 2 months. During esophagoscopy, a fungative-meaning not clear mass was detected in the upper third of the thoracic esophagus. Biopsy revealed squamous cell carcinoma. Because of his poor physical condition, we suggested concurrent chemoradiotherapy (CCRT) as the treatment option. We performed feeding jejunostomy for nutritional maintenance. The treatment course was uneventful until 5 months later, when he complained of progressive productive cough with saliva-like sputum. T-E fistula, located 5 cm above the carina, was confirmed by bronchofiberscopy (Figure 1).

Under fluoroscopic and bronchofiberscopic guidance, placement of a tracheal covered self-expanding metallic stent (Ultraflex, Boston Scientific, USA), 1.6 cm in diameter, 4 cm in length, and with a 2 cm membranous cover, was performed. After the procedure, the patient's symptoms greatly improved. We further consulted a radiotherapist who suggested CCRT be continued for as long as the patient could tolerate it.

Two weeks later, the patient was brought to our emergency department with severe dyspnea. Cold sweats, tachycardia, anxiety, stridor and gasping were noted. O_2 saturation was 90% in room air and the respiratory rate was 27 to 30 breaths per minute. Bronchofiberscopy and chest computed tomography (CT) were performed immediately, and showed the tracheal SEMS crossing the trachea and the esophagus and compromising the airway (Figures 2, 3). Because of impending asphyxia, tracheostomy was performed immediately in the emergency room, lest the patient suffer an arrest during transfer to the operating room. Under endotracheal tube general anesthesia, the 2^{nd} to 5^{th} tracheal rings were incised through a cervical transverse incision. The stent was then retrieved with a long hemostatic clamp. A long endotracheal tube was then inserted and the fistula bypassed. Its tip was fixed just above the carina. The symptoms were then relieved.

After the procedure, bronchofiberscopy re-

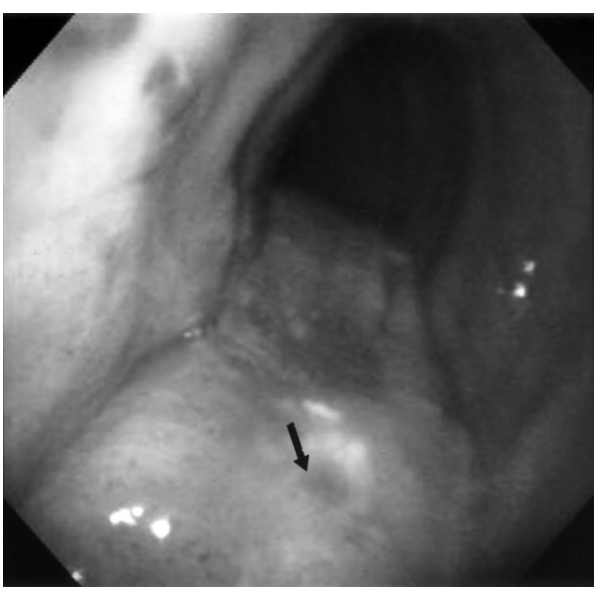

Fig. 1. Bronchofiberscopy (arrow) revealed T-E fistula.

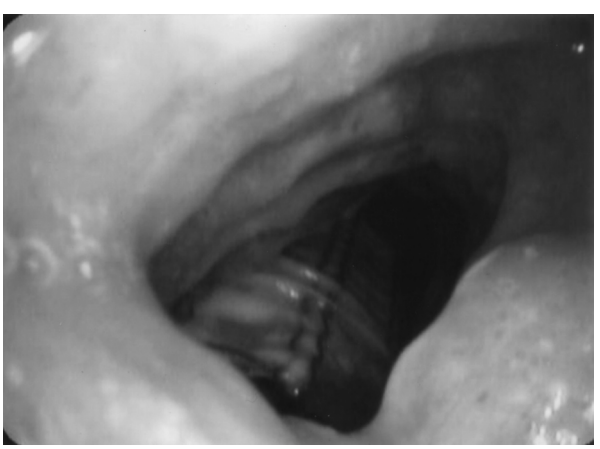

Fig. 2. Bronchofiberscopy revealed a SEMS crossing between the trachea and esophagus. Note the compromised airway.

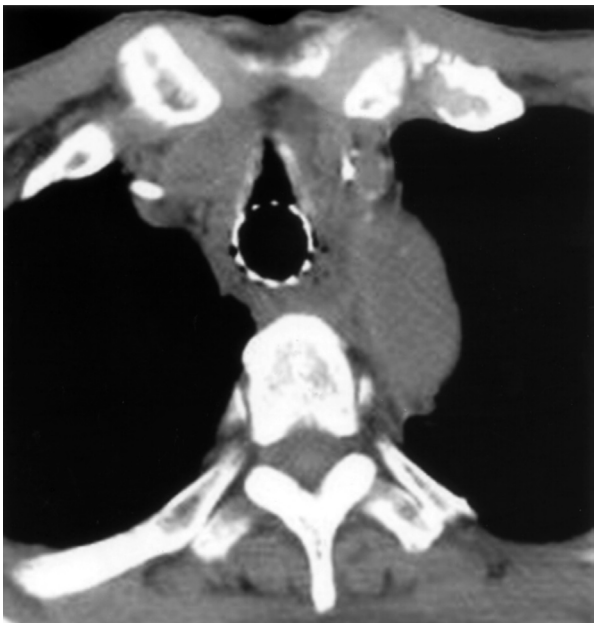

Fig. 3. Chest CT scan revealed erosion of the tracheal stent into the esophagus.

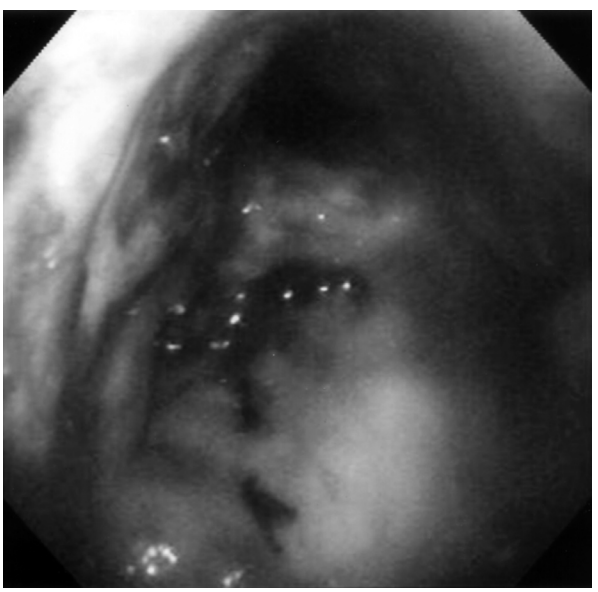

Fig. 4. Follow-up of bronchofiberscopy after the retrieval of the stent revealed serious destruction of a tracheal membranous portion.

vealed serious destruction of a tracheal membranous portion (Figure 4). Thereafter, the tracheostomy tube was changed every 4 weeks under bronchofiberscopic guidance.

Unfortunately, the patient expired 3 months later because of cachexia and an aggravated general condition. Prior to his death, his breathing was smooth and he was free from aspiration or respiratory infection.

Discussion

Tracheal stents are used for unresectable malignant obstructions or to seal off malignant T-E fistula, especially in those patients who are unfit for surgical procedures [3-4]. The advantages are that tracheal stent placement is technically feasible in most centers, less invasive compared with surgery, and results in dramatic symptomatic relief. However, migration, fracture, granulation and perforation are among the disadvantages [4-5].

A possible explanation for this patient's condition is that the perforation in the membranous portion, originally caused by CCRT, was exacerbated by the SEMS. The SEMS then gradually invaded the esophageal wall and finally bridged the 2 organs. To our knowledge, no similar report can be found in the literature.

In such a scenario, and because of airway obstruction and impending asphyxia, the patient should be (1) correctly diagnosed and (2) managed immediately. Images studies cannot offer much help for diagnosis. CT scan might provide some clues, but the transferring time would be long and the diagnosis delayed. Bronchofiberscopy has become the only reliable means of diagnosis.

Removal of the SEMS through rigid bronchoscopy or tracheostomy is the only option after the diagnosis. The former requires a skillful technique. However, we decided to perform a tracheostomy, since the proximal end of the stent was above the sternal notch. Also, it could be performed immediately in the emergency

department.

This complication should always be kept in mind. We recommend the application of 2 stents, simultaneously at the trachea and esophagus in the event of T-E fistula [6-8]. First, the airway obstruction can be alleviated. Shortly, thereafter, oral intake can be resumed. With this, the migration of the tracheal stent and erosion of the esophageal wall can be prevented. Also the symptoms of T-E fistula would be lessened, even if they worsened after further CCRT [9-11].

It is difficult to determine whether continuation of CCRT after emergency episodes is necessary. The authors suggest further CCRT if (1) the patient's performance status allows, and (2) after the placement of tracheal and esophageal stents instead of the emergency tracheostomy tube. Otherwise, the patient should receive the best supportive care owing to his short life expectancy. Although our patient survived for only 3 months afterwards, he enjoyed unhindered breathing before he died, so our management should be considered successful.

In conclusion, a SEMS crossing between the trachea and esophagus is a life-threatening emergency. Correct diagnosis and immediate treatment is the only option to prevent death.

Acknowledgement

The authors thank Jadzia Chou for her revision of the article.

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可擴展氣管支架移位所造成的窒息：未曾預料的併發症

姜宏興* 莊逸君** 李憲斌* 李瑞英* 李彥龍* 張鈺堂* 周世華*,***

自我擴展金屬支架（SEMS）常用於惡性氣管食道瘻管。作者現報告因氣管支架持續侵蝕造成支架移位至食道同時造成氣道阻塞，危在旦夕之病例。患者於急診室隨即接受緊急氣管造口，並且移除支架，重新放置加長型的氣管造口管繞過瘻管處，挽回一命。文獻上尚未報導類似的併發症。*(胸腔醫學* ***2011; 26: 80-84)***

關鍵詞：氣管支架，惡性氣管食道瘻管，移位

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A Rare Case of Tuberculosis-Associated Immune Reconstitution Inflammatory Syndrome after Resection of Thymoma

Guo-En Huang*, Wann-Cherng Perng, Chien-Wen Chen

Tuberculosis-associated immune reconstitution inflammatory syndrome (IRIS) after human immunodeficiency virus (HIV) treatment is currently a well-accepted concept. IRIS also occurs in non-HIV-infected patients. We presented a 30-year-old HIV-negative man with thymoma, CD4 lymphocytopenia and disseminated tuberculosis. He received anti-tuberculous therapy and responded well initially. Resection of the thymoma was performed 2 months after the initiation of anti-tuberculous therapy. Neck lymph nodes enlarged and became confluent 1 week after surgical resection of the thymoma. A cutaneous fistula developed and pus smear disclosed abundant acid-fast bacilli. The CD4 count at that time was higher than at admission, and the clinical course was compatible with tuberculosis-associated IRIS. The neck lymphadenopathy improved gradually with continuous anti-tuberculous therapy and repeated surgical debridement. Tuberculosis-associated IRIS may occur after rapid restoration of the immunodeficiency status. ***(Thorac Med 2011; 26: 85-92)***

Key words: thymoma, CD4 lymphocytopenia, disseminated tuberculosis, immune reconstitution inflammatory syndrome

Introduction

During the treatment of human immunodeficiency virus (HIV) infection, a paradoxical worsening of the pre-existing opportunistic infection may occur in those who are receiving concurrent highly active antiretroviral therapy (HAART). After the severely compromised immunity of the HIV-infected person is restored, vigorous and dramatic reconstitution of pathogen-specific host responses may develop [1-2]; this phenomenon is termed immune reconstitution inflammatory syndrome (IRIS). The likelihood of IRIS correlates with the extent of $CD4^+$ T cell immune suppression prior to the initiation of HAART, and the degree of viral suppression accompanying an increase in the circulating CD4 cell count following the initiation of

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HAART [3]. This phenomenon has also been reported in non-HIV immunocompromised hosts with concurrent opportunistic infection, such as solid organ transplantation recipients who underwent a withdrawal of immunosuppressive agents, neutropenic patients who are recovering from neutropenia, and patients whose tumor necrosis factor (TNF)-alpha therapy has been discontinued. When immunosuppressive factors are removed from these non-HIV hosts and appropriate antimicrobial therapy is started, the T helper responses suddenly become strong and are not balanced by anti-inflammatory responses [4]. We report a non-HIV infected young man with thymoma, CD4 lymphocytopenia and disseminated tuberculosis. IRIS, manifested as a worsening of cervical lymphadenitis, developed after anti-tuberculous therapy and resection of the thymoma.

Case Report

A 30-year-old Taiwanese man presented with dysphagia and fever for 1 month. He also had night sweats, productive cough, progressive dyspnea and body weight loss (15 kg within 1 month), and a tender nodule at the right side of his neck. On physical examination, his temperature was 39.2^{o}C, respiratory rate 18/min, pulse rate 137/min and blood pressure 118/78 mmHg. There were several soft, non-tender, movable lymph nodes along both sides of his neck, the largest being 5 cm in diameter. Oral thrush was found at the posterior pharyngeal wall and soft palate.

Chest radiography showed a large anterior mediastinal mass, along with faint tiny nodules in both lung fields, and interstitial and nodular infiltrates in the left upper lobe (Figure 1). High resolution computed tomography of the chest showed a large soft tissue mass (7.2 x 5.8 cm) with heterogeneous enhancement at the prevascular space of the left anterior mediastinum, diffuse nodules scattered in both lung fields, conglomerated nodules in the left upper lung, and enlargement of the supraclavicular lymph nodes

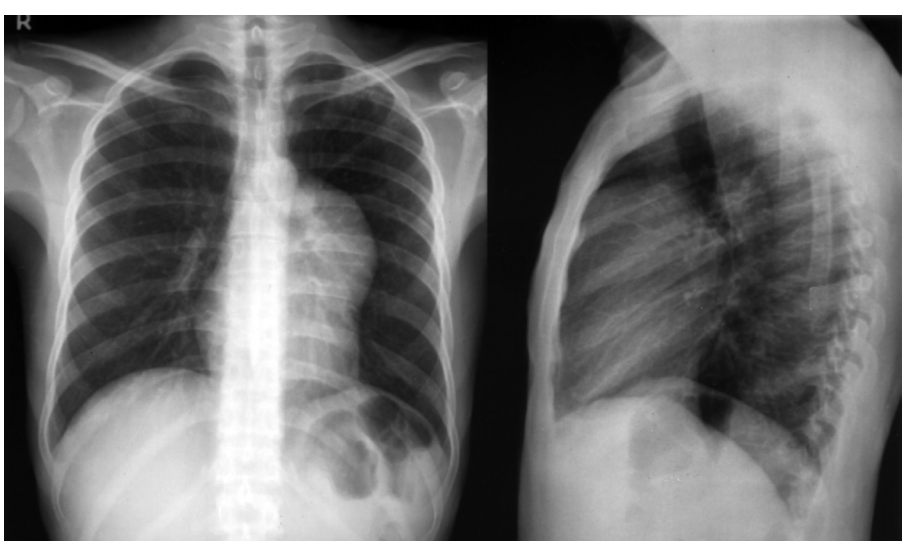
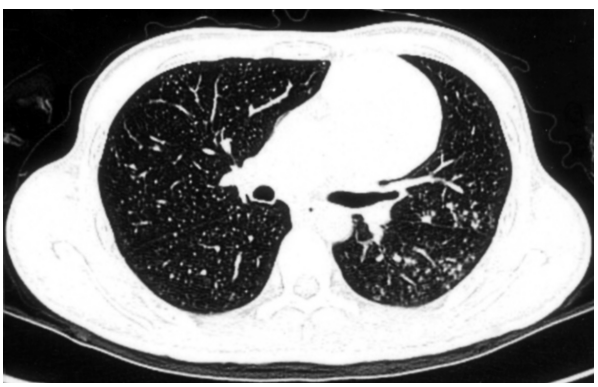
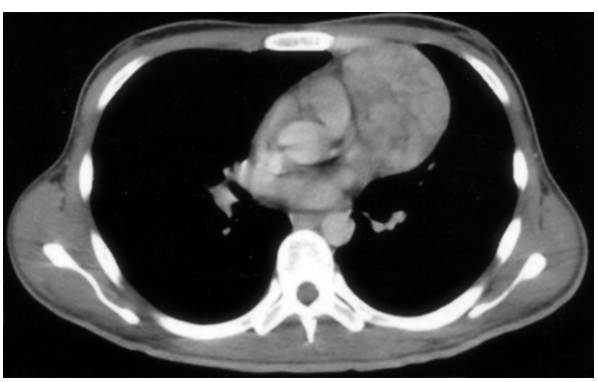


Fig. 1. Chest radiology on admission shows a large anterior mediastinal mass, along with faint tiny nodules in both lung fields and interstitial and nodular infiltrates at the left upper lobe.

(Figure 2). Brain magnetic resonance imaging (MRI) was performed to exclude infection of the central nervous system, and the results were normal. Laboratory analyses revealed normocytic anemia (Hb 11.4 g/dl, MCV 92.6 fl), and an elevated white blood cell count (14400 10^6/μL), with neutrophils 84.8% and lymphocytes 12.5%. The antibody test for HIV was negative. The lymphocyte surface marker analysis yielded $CD4^+$ lymphocytopenia (23%, 232 cells/μL; normal 35-50%, >500 cells/μL) and an inverted CD4:CD8 ratio (0.88, normal 1.0-2.0). Serum immunoglobulin levels were normal. Sputum acid-fast stain revealed many acid-fast bacilli (AFB) and sputum culture later grew *Mycobacterium tuberculosis*. Sputum polymerase chain reaction (PCR) for *Mycobacterium tuberculosis* complex was also positive. Sonography-guided transthoracic biopsy of the mediastinal mass yielded a benign thymoma. Excisional biopsy of the neck lymph node showed chronic inflammatory cell infiltration with fibrosis, a negative AFB stain, and a negative Mycobacterium species culture. The patient was admitted to an isolation ward and full-dose anti-tuberculous chemotherapy with isoniazid, ethambutol, rifampin, and pyrazinamide was started.



(a)



(b)

Fig. 2. High resolution computed tomography of the chest shows diffuse tiny nodules scattered throughout both lung fields and a patchy opacity at the left apical lung (a, lung window). A large soft tissue mass (7.2 x 5.8 cm) with heterogeneous enhancement at the prevascular space of the left anterior mediastinum. (b, soft tissue window).

The acid-fast stain of the sputum turned negative after 3 weeks of anti-tuberculous therapy. There was no more fever and the neck lymphadenitis subsided. The patchy infiltrates in the left upper lobe gradually resolved on the radiograph, and the miliary nodules diminished. Nine weeks after the initiation of anti-tuberculous therapy, the patient received surgical resection of the thymoma. On gross examination, the tumor was encapsulated. The microscopic examination showed the tumor was composed of predominantly medium-sized spindle epithelial cells with slight atypia, mixed with lymphocytes. The histology was compatible with thymoma, WHO type B3, Masaoka stage II (Figure 3). Anti-tuberculous therapy was continued and directly observed. The mycobacterial isolate was sensitive to all the drugs tested, including isoniazid, rifampin, ethambutol and streptomycin.

However, a paradoxical reaction with newly developed neck lymphadenitis developed a week after the surgery (10 weeks after the initiation of anti-tuberculous therapy; the radiotherapy had not been started at that time) (Figure 4). He had not received any immunosuppres-

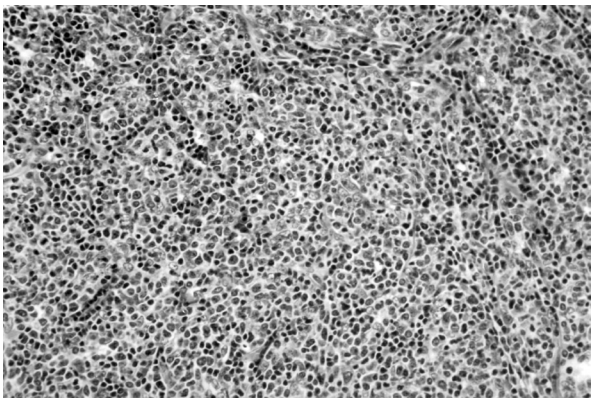


Fig. 3. Pathology of the thymoma
Sheet-like tumor cells with few lymphocytes. Round or elongated nuclei with inconspicuous nucleoli. (compatible with WHO classification type B3)

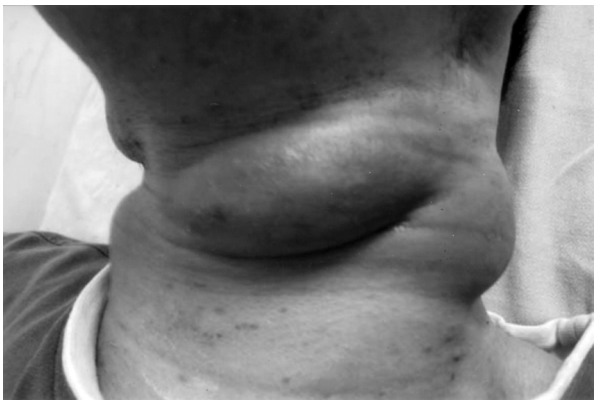


Fig. 4. One week after surgical resection of the thymoma, tuberculosis-associated immune reconstitution inflammatory syndrome developed in the form of confluent, swelling lymph nodes and cutaneous fistula that drained pus with strongly positive AFB.

sive agents, including steroids, before the development of lymphadenitis. The neck lymph nodes became confluent, and a cutaneous fistula developed that drained pus with strongly positive AFB. The PCR for *Mycobacterium tuberculosis* complex of the drained pus was positive but mycobacterial culture showed no growth 2 months later. There was no change on the chest radiographs as compared with 2 weeks previously. We rechecked the lymphocyte surface markers, which revealed a partial recovery of the lymphocyopenia. His $CD4^+$ lymphocyte count was 385/μL and CD4:CD8 ratio was 1.3. The patient underwent surgical debridement of the neck lymph nodes several times and the anti-tuberculous therapy was continued. There was no more fever or body weight loss. The confluent lymphadenitis gradually resolved and the fistula finally healed. Five months after admission, he was discharged with anti-tuberculous therapy. At the time of discharge, the miliary lesions on chest X-ray were almost completely resolved. The absolute CD4 count was still below the normal range (154.8/μL) and the CD4/CD8 ratio was 1.09. The patient did not come back for follow-up after discharge.

Discussion

The criteria of IRIS have yet to be established. In HIV-infected individuals, most definitions required the presence of acquired immunodeficiency syndrome (AIDS) with a severe CD4 lymphocytopenia, the presence of an antigen (bacteria, viruses or fungi), a positive virologic and immunological response to HAART therapy, and the exclusion of an alternative diagnosis (such as drug resistance or poor compliance with therapy). In non-HIV patients, a dysregulated inflammatory response was also reported when there was a rapid reconstitution of immunodeficiency. No laboratory marker is considered as a reliable indicator of IRIS [5]. Tuberculosis-associated IRIS is particularly well recognized. The most common clinical manifestations are fever, lymphadenopathy and deterioration of respiratory symptoms. In HIV-infected patients, the onset of tuberculosis-associated IRIS was strongly associated with anti-viral therapy, mostly within 2 months after the initiation of HAART. Extrapulmonary TB

was a risk factor [6]. However, long before the HIV epidemic, clinical illness consistent with IRIS had been reported in non-HIV patients following anti-tuberculosis treatment [7]. A paradoxical worsening of pre-existing infection may occur after initiation of anti-tuberculosis therapy, although it is usually self-limited; most of the reported cases were extrapulmonary tuberculosis [8]. In non-HIV patients with lymph node tuberculosis, Hawkey *et al*. [9] reported that the incidence of a paradoxical reaction after anti-tuberculous therapy was 23%. In this retrospective study, the median onset time was 46 days after initiation of anti-tuberculous therapy. Hosts immunocompromised due to solid-organ transplantation, women during the post-partum period and patients recovering from chemotherapy–induced neutropenia have also been reported to develop IRIS when there is a sudden improvement in immune function. The mechanism is thought to be a sudden change in the dominant T helper responses to inflammation that is not well balanced by anti-inflammatory responses [4].

In non-HIV infected patients with tuberculosis, a CD4 lymphocytopenia also indicates advanced disease and immunodeficiency. In a West African study [10], 14% of patients who were hospitalized due to pulmonary tuberculosis presented with CD4 cell subsets below 300 cells/ml in the absence of HIV infection. On the other hand, disseminated tuberculosis is thought to be a cause of profound lymphocytopenia. In most cases, T cell subsets were not evaluated until the patient became overtly ill [11-12]. Lymphocytopenia may predispose to miliary tuberculosis, whereas miliary tuberculosis may result in lymphocytopenia [10-11]. It is difficult to differentiate which condition presents first. The interaction between CD4 cell subsets and mycobacteria is complex. Increases in CD4 cell subsets and improvement in immune function may trigger clinical manifestations of tuberculosis [12].

Thymomas are the most common thymic neoplasm, and immunological dysregulation is 1 of the paraneoplastic syndromes of thymoma. The immunological alternations include inverted CD4:CD8 cell ratios, B-cell lymphopenia and hypogammaglobulinemia [13-14]. Although in previous reports the removal of the thymoma in this subset of patients did not reliably induce the recovery of immunodeficiency [15], thymectomy has been reported to improve chronic diarrhea in those who had thymoma with immunodeficiency [16]. In this case, the deterioration of lymphadenitis occurred after the resection of the thymoma. The clinical course resembled the tuberculosis-associated IRIS that occurs in HIV-infected patients (Figure 5). This implies a certain degree of improvement in the immune status after surgical resection of the thymoma. The IRIS was probably a response to surgical intervention and anti-tuberculous therapy.

In HIV-infected patients, TB-associated IRIS was thought to be a self-limiting syndrome, and that anti-TB therapy, anti-viral therapy should be continued. Manosuthi *et al*. [17] reported there was a tendency toward a higher subsequent mortality and morbidity, and that affected patients may need hospitalization for supportive care and even anti-inflammatory agents such as steroids to minimize the deterioration of symptoms. In our patient, the presentation of TB-associated IRIS was mainly the worsening of lymphadenitis. The confluent lymphadenitis persisted and required surgical intervention. The cutaneous fistula closed under repeated debridement, as well as the adminis-

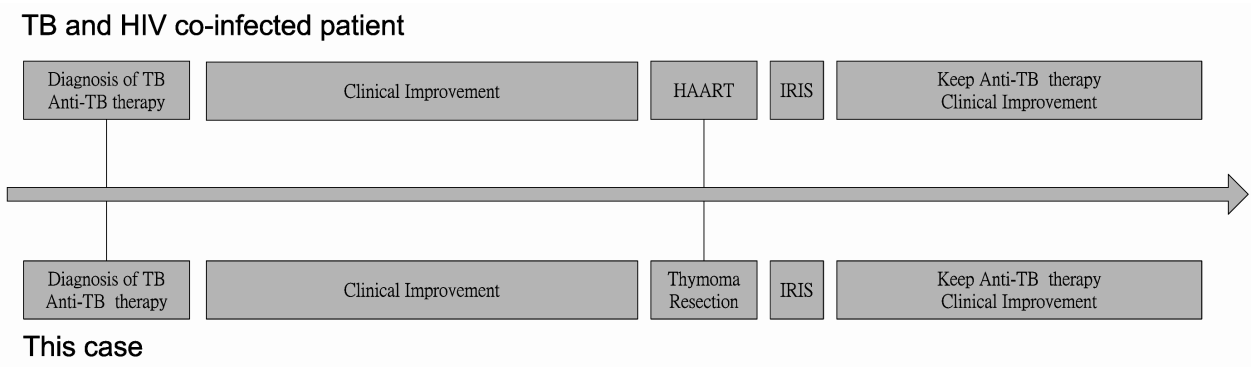


Fig. 5. Similarity between the clinical course of this case and that of patients with HIV infection, tuberculosis and IRIS.

tration of effective anti-tuberculous therapy. Supportive care includes drainage or excision of lymph nodes; the continuation of anti-tuberculous therapy was also a principle of treating TB-associated IRIS in non-HIV patients with TB adenitis [9].

Conclusion

We report a patient with disseminated tuberculosis, CD4 lymphocytopenia and a thymoma who developed tuberculosis-associated IRIS 8 days after thymectomy, and 71 days after the initiation of anti-tuberculous therapy. The patient was not HIV-infected but thymoma-associated CD4 lymphocytopenia predisposed him to disseminated tuberculosis. After the initiation of anti-tuberculous therapy and thymectomy, the neck lymphadenitis worsened; IRIS may be occurred as the the immune suppressive factors was removed. When the CD4 lymphocytopenia improved, IRIS improved after the continuation of anti-tuberculous therapy and surgical debridement.

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一個造成結核相關免疫重建炎性症後群的罕見原因：胸腺瘤切除

黃國恩* 彭萬誠 陳健文

結核病相關之免疫重建炎性症候群目前已經是一個被廣泛接受的觀念。然而，在 HIV 陰性病患身上，只有少數的案例被報導過。我們報告一位 30 歲 HIV 測試陰性之男性病患，具有胸腺瘤，CD4 淋巴球低下併發瀰漫性肺結核。病患接受了抗結核的治療，並且臨床症狀有改善。經過 2 個月的有效抗結核治療之後，病患接受手術切除胸腺瘤。然而在術後一周之後，病患被發現產生新的淋巴結腫大以及融合之淋巴炎，並形成皮膚瘻管且流出具大量耐酸桿菌之引流液。此時再檢測 CD4 淋巴球數目，發現已有改善，故確定免疫重建炎性症候群之診斷。此為罕見之非 HIV 之感染，在切除胸腺瘤後發生結核病相關之免疫重建炎性症候群之病例。臨床醫師需注意任何的治療或處置若造成免疫力快速重建，則可能產生免疫重建炎性症候群。*(胸腔醫學* ***2011; 26: 85-92)***

關鍵詞：甲狀腺，CD4 淋巴球缺乏，瀰漫性結核，免疫重建發炎症候群

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Suspected Aspiration Pneumonia in a Patient with Esophageal Compression Related to Cervical Spine Osteophytes

Hui-Chia Tien, Chung-Yi Lin, Chien-Liang Wu

This report describes a case of aspiration pneumonia with a very rare etiology – cervical spine osteophytes. The initial symptoms of this 65-year-old man were progressive dysphagia and weight loss for 1 year. He had been admitted recently for aspiration pneumonia. Radiological examination indicated anterior cervical spine osteophytes at the C4-C5 level, which obstructed the esophagus, and consequently induced frequent choking. This can be easily diagnosed by cervical X-ray, but overlooking the findings could easily lead to severe complications. After undergoing an anterior cervical disectomy at C4-C5, the patient's nutritional status and the above-mentioned symptoms improved. ***(Thorac Med 2011; 26: 93-98)***

Key words: pneumonia, dysphagia

Introduction

Aspiration pneumonia is commonly seen in the pulmonary service. The associated risk factors include COPD, heart disease, malignancy, diabetes mellitus, living in a nursing home, cerebral infarction and aging [1-2]. Among these factors, aging, which results in a decrease in swallowing and the cough reflex, plays the most important role. Aspiration pneumonia caused by anterior cervical spine osteophytes is rare. Since cervical symptoms are usually not obvious, physicians may overlook the differential diagnosis of cervical spine osteophytes. We report a 65-year-old man with ankylosing spondylitis who was admitted for aspiration pneumonia caused by anterior cervical spine osteophytes.

Case Report

A 65-year-old male, an ex-smoker (30 packs per year), had been diagnosed with ankylosing spondylitis 40 years previously with regular follow-up in a local hospital.

He complained of progressive dysphagia, choking easily, poor appetite and weight loss of 15 kg (from 60 to 45 kg) during the past year. He also had had yellow sputum and numbness of both hands for a long time. Half a year before this admission, he went to a local hos-

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pital where thorough examinations, including abdominal sonography, abdominal computed tomography (CT), panendoscopy and tumor markers, were performed to exclude malignancy. The results of the abdominal CT, abdominal sonography and tumor markers were unremarkable. He was discharged with the diagnosis of gastro-esophageal reflux. He used a proton-pump inhibitor regularly, but the symptoms still progressed. One month before this admission, he experienced poor sleep quality and emotional disturbance, for which he visited a psychiatrist, but he still felt very unwell after anxiolytics and hypnotics use.

On this occasion, he was admitted to our ward with exacerbation of cough with increasing yellow sticky sputum for 4 days. Fever was also noted during hospitalization. Physical examination was normal, except right lower lung crackles. Laboratory tests showed leukocytosis (15.70 x 10^3/uL) and elevated C-reactive protein (19.41 mg/dL). Chest X-ray (Figure 1) showed right lower lung infiltrates, compatible with pneumonia. Because of the patient's choking easily and numbness in both hands, cervical X-ray (Figure 2) was performed, which showed symmetrical syndesmophytes bridging intervertebral disc spaces at the level of C4-C5. In the esophagogram (Figure 3a) report, choking easily was found to result from posterior compression of the esophagus by a marked spur formation at the C4-C5 level. Contrast medium aspiration in the right lower lung (Figure 3b) was also found during the exam. After moxifloxacin and piperacillin treatment for 1 week, the fever subsided and the right lower lung consolidation resolved itself. Three sets of sputum culture confirmed that there was no pathologic bacterial growth. Spinal magnetic resonance imaging (MRI) (Figure 4) revealed disc bulging and

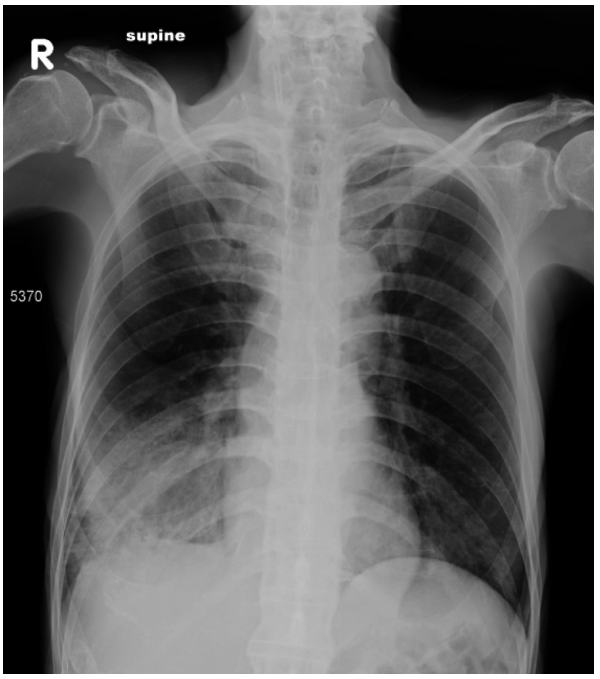

Fig. 1. Chest X-ray showed right lower lung infiltrates, compatible with pneumonia.

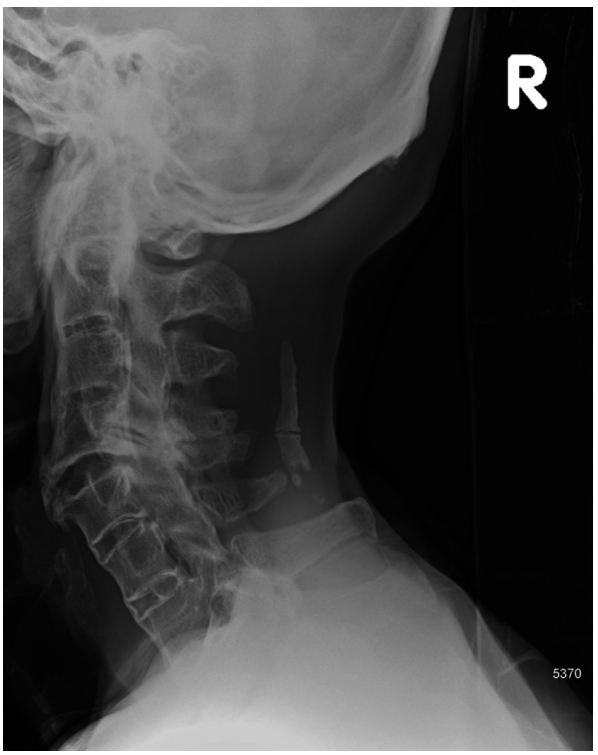

Fig. 2. Cervical X-ray showed symmetrical syndesmophytes bridging intervertebral disc spaces at the level of C4-C5.

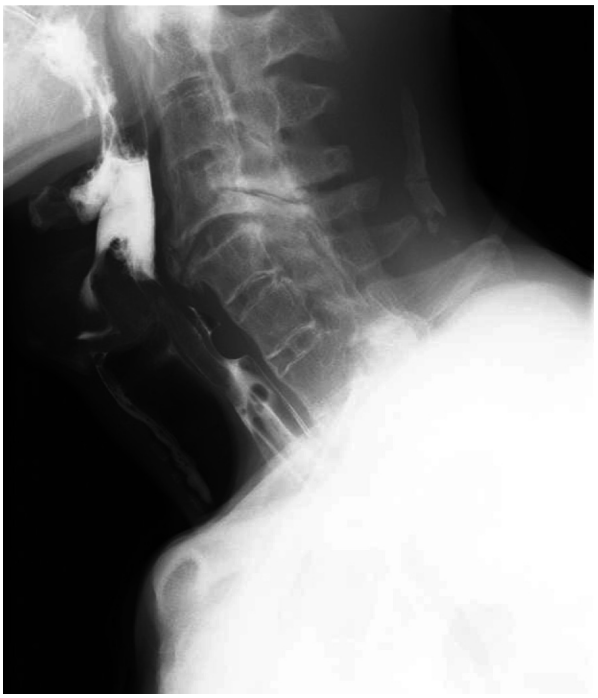

Fig. 3a. In the esophagogram, posterior compression of the esophagus by spur formation at the C4-C5 level.

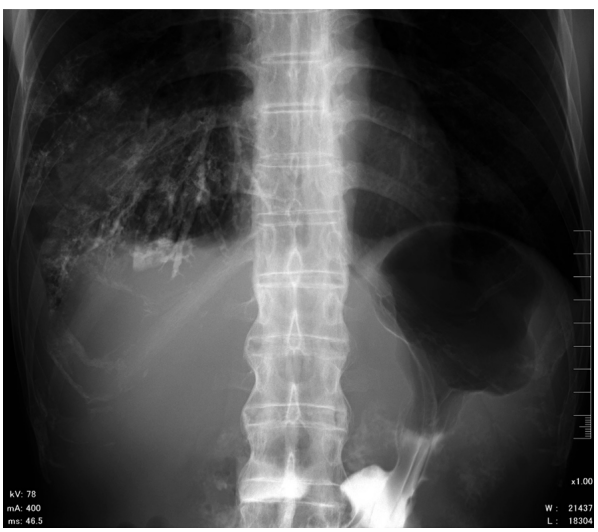

Fig. 3b. Contrast medium aspiration in the right lower lung was noted during the exam.

hypertrophy of the ligamentum flavum at the C4-C5 level causing spinal stenosis and mild cord compression. The neurosurgeon recommended operation, and the patient successfully

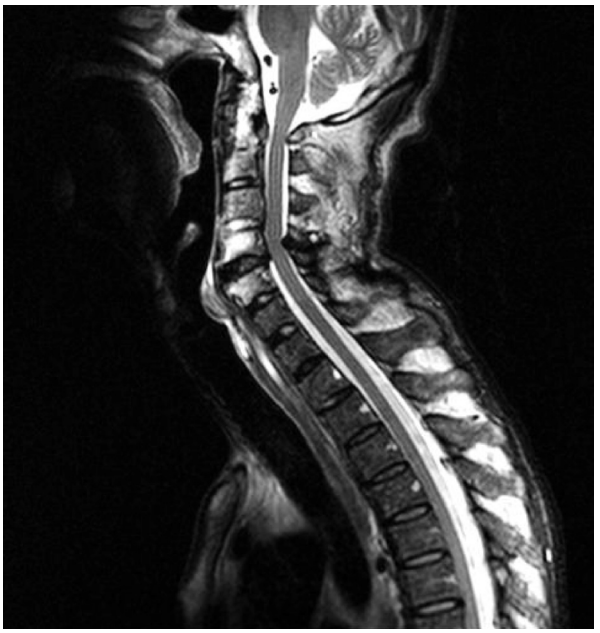

Fig. 4. Spinal magnetic resonance imaging revealed disc bulging and hypertrophy of the ligmentum flavum at the C4-C5 level with cord compression.

underwent C4 and C5 discectomy. One month after surgery, he was followed up at our outpatient clinic. The dysphagia was significantly improved.

Discussion

Aspiration pneumonia, also called "deglutition pneumonia" and "hypostatic pneumonia" [2], is defined as the misdirection of oropharyngeal secretions or gastric contents into the respiratory tract [1], followed by findings of inflammation in the chest X-ray [2]. This type of aspiration can be divided into 2 categories: silent and apparent aspiration [2]. Silent aspiration, which means the swallowing of nasalpharyngeal secretion due to decreased swallowing and cough reflexes [2], usually occurs at night. This condition is frequently found not only in mechanically ventilated patients, but also in healthy elderly persons with gradually

decreased swallowing and cough reflexes. Apparent aspiration occurs when persons with swallowing disorders direct food into the airways during meals [2]. Sedative and sleeping medications also lead to silent and apparent aspiration due to the muscle relaxation effect [2].

The common disorders related to aspiration are anatomic abnormalities, such as neoplastic diseases, neuromuscular disorders, and gastroesophageal reflux [3]. Cervical osteophytes are rarely mentioned except in patients with ankylosing spondylitis (AS) and diffuse idiopathic skeletal hyperostosis (DISH), also known as physiologic vertebral ligamentous calcification) [4]. In 1926, Mosher reported 2 cases of cervical spondylitic dysphagia [3, 6], and thereafter, sporadic case reports were published. Many hypotheses have been put forth, but the mechanism of cervical osteophyte-induced dysphagia remains unclear. Anterior cervical osteophytes are often found in middle-aged men without symptoms. Only a few patients have experienced dysphagia because of esophageal compression by cervical osteophytes, and associated pneumonia is even rarer. A review of anterior cervical ostephytes by Saffouri and Ward reported only 7 cases of dysphagia in 116 patients (6.03%) with cervical osteophytes [7]. The C4-C7 vertebral body is usually affected because at this level the esophagus is relatively less flexible [5] and closely fixed to the cricoid and dense soft tissue around the thyroid [3]. Below C7, the esophagus is flexible, so it can change shape to allow the bolus to pass. Osteophytes may also result in perifocal inflammation around the esophagus, with follow-up adhesion or fibrosis [3]. Our patient also had osteophytes at the C4-C5 level that compressed the esophagus, which was partly fixed to the peripheral tissue, leading to progressive dysphagia.

A lateral spine film and esophagogram can confirm the diagnosis [4]. MRI provides a definite anatomic picture of osteophytes for preoperative evaluation.

Conservative treatment should be administered first. Surgery is indicated in patients who have severe esophageal obstruction and life-threatening symptoms, such as recurrent aspiration and cachexia. Nutritional counseling is also of great importance [4].

In conclusion, we have reported a middle-aged male patient with ankylosing spondylitis who had experienced numbness of both hands for a long time. During the most recent year, he suffered from progressive dysphagia, choking easily, and weight loss of 15 kg. These symptoms led to his mood disorder, for which he visited a psychiatrist. Thereafter, he took sleeping medication for 1 month, which might ultimately have resulted in aspiration pneumonia. After a meticulous history-taking and extensive differential diagnosis, cervical osteophytes-induced dysphagia and aspiration were diagnosed. Eventually, he was treated appropriately, and life-threatening complications were reduced to a minimum.

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一個疑似吸入性肺炎病人併有頸椎骨刺造成的食道壓迫

田蕙嘉　林長怡　吳健樑

本文報告一個罕見的吸入性肺炎病例，它的病因為頸椎骨刺。這位 65 歲的男性起初的症狀為在過去一年有漸進性的吞嚥困難和體重減輕。最近則因為吸入性肺炎住院。放射線診斷顯示在頸椎前部第四節及第五節有骨刺導致食道開口阻塞以致於容易嗆到。這個病因可以經由頸部 X 光簡單地被診斷，但是疏忽卻會導致嚴重的併發症。患者接受頸椎前部第四節及第五節椎間盤切除後，他的營養狀況和症狀都有改善。*(胸腔醫學* ***2011; 26: 93-98)***

關鍵詞：肺炎，吞嚥困難

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Gastrointestinal Metastasis of Primary Lung Mucoepidermoid Carcinoma: A Case Report

Chun-Sheng Chen, Diahn-Warng Perng, Yu-Chin Lee

Primary lung cancer is a common neoplasm, and frequently metastasizes to internal organs such as the lung, liver and adrenal gland; however, it is relatively rare for lung cancer to metastasize to the gastrointestinal tract. The common symptoms are gastrointestinal bleeding, abdominal pain, obstruction, and perforation. The most common histology of small intestinal metastasis from lung cancer is adenocarcinoma. Gastrointestinal tract metastasis is an extremely poor prognostic indicator of lung carcinoma. We described an 80-year-old male with mucoepidermoid carcinoma of the lung, left lower lobe, with mediastinal lymphadenopathy and lung-to-lung metastases, cT2N2M1, stage IV status post-chemotherapy who suffered from an acute onset of abdominal pain. The abdominal CT revealed a suspected ruptured hollow organ. Emergency laparotomy revealed ileum perforation, and the pathology of the surgical specimen proved intestinal metastasis from lung cancer. The patient expired about 2 weeks after the laparotomy. ***(Thorac Med 2011; 26: 99-103)***

Key words: lung carcinoma, gastrointestinal metastatic tumor

Introduction

Primary lung cancer is a common neoplasm, and frequently metastasizes to internal organs such as the lung, liver and adrenal gland; however, it is relatively rare for lung cancer to metastasize to the gastrointestinal (GI) tract. The common symptoms are GI bleeding, abdominal pain, obstruction, and perforation. Herein, we describe a case of symptomatic GI metastasis from lung cancer.

Case Report

This 80-year-old male visited our emergency room on 1 July 2008 because of a worsening cough and dyspnea for a week. He was a smoker, consuming 1.5 packs of cigarettes per day for 50 years, and had a history of mucoepidermoid carcinoma of the lung, left lower lobe, with mediastinal lymphadenopathy and lung-to-lung metastases, cT2N2M1, stage IV status post-oral Ufur plus vinorelbine for 3 cycles, which was started on 21 April 2008; palliative radiotherapy with a total dose of 4000cGy over the tumor was started on 24 June 2008. He was

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admitted to our hospital for treatment of suspected pneumonia on the same day. However, the patient complained of acute abdominal pain on 9 July. Physical examination revealed diffuse tenderness with rebounding pain throughout the abdomen. Emergency abdominal CT was arranged on the same day, and showed free air in the abdomen, but more in the upper abdomen. In addition, ascites was also noted. Hence, a ruptured hollow organ was considered. Emergency laparotomy was performed immediately. Exploration of the abdominal cavity revealed a small perforation at the ileum, at 170 cm above the ileocecal valve, with a soft tissue mass inside; several small nodules at the mesentery were also noted. Segmental resection of about 20 cm and anastomosis were performed. Histological analysis of the specimen (Figure 1) showed the presence of a transmural infiltration of adenocarcinoma cells which were immunoreactive for CK5/6 and CK7, but negative for CK20, TTF-1 and CDX2 stains. Intracytoplasmic mucin vacuoles were observed, and mucosal necrosis and acute and chronic inflammation were also seen. Lymphatic invasion was noted. A comparison with the previous biopsy specimen of the primary lung tumor (Figure 2) showed a similar morphological and immunohistochemical finding. Ileum metastasis from lung cancer with the complication of intestinal perforation was confirmed. After the operation, the patient recovered slowly and could not be weaned from mechanical ventilator support. Unfortunately, an episode of ventilator-related pneumonia occurred on 26 July 2008, followed by septic shock with multiple organ systemic dysfunction. The patient expired on 28 July 2008.

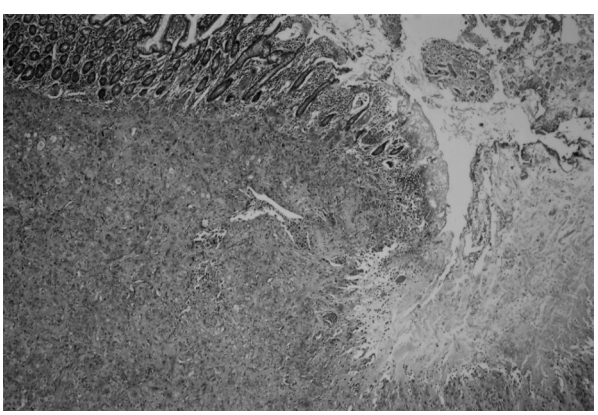

Fig. 1. (H&E stain X 40) The specimen was obtained from the ulcerative site of the ileum, which showed transmural infiltration of adenocarcinoma cells. Mucosal necrosis and acute and chronic inflammation are also seen. The finding was similar to the morphological and immunohistochemical findings of specimens from primary lung cancer.

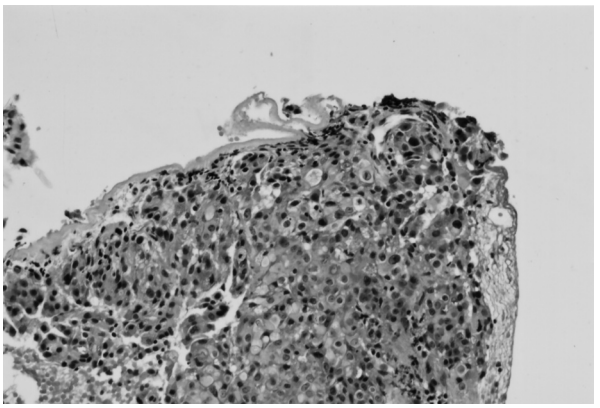

Fig. 2. (H&E stain X 200) The specimen was obtained from the primary lung tumor, which revealed bronchial tissue with non-small cell carcinoma, and is composed of neoplastic cells arranged in solid and focal glandular patterns. Focal squamoid differentiation is noted. The morphological findings are compatible with those of mucoepidermoid carcinoma.

Discussion

Lung cancer is the most frequent cause of cancer death in the world. About 50% of patients with lung cancer have distal metastasis at the time of diagnosis. The preferential sites of extra-pulmonary metastasis are the lymph nodes, liver, adrenal gland, bone, and brain. GI metastasis of primary lung carcinoma is

considered to be very rare, although there is approximately 4.7-14% prevalence at autopsy [1-2]. Acute abdominal emergencies due to GI metastasis from carcinoma to the lung are extremely rare. Yang [3] reported that the clinical prevalence of symptomatic GI metastasis of lung cancer was 1.77% (6/339). The small bowel was the most common GI metastatic site of lung cancer. The jejunum had a slightly higher prevalence [4-5], and the duodenum as a site of metastasis was relatively rare [6-7]. Esophageal metastasis was rare, but direct extension was common, based on autopsy reports [2]. Most cases are asymptomatic with rare symptoms of dysphagia. The middle 1/3 of the esophagus is the most common site of metastasis, mainly due to its proximity to the lymphatic-rich tracheal hilar region. Gastric metastasis from lung cancer is very rare and often asymptomatic. Autopsy reports range from 0.2% to 1.7% in several series [1-2], and only a few symptomatic cases have been published [8-11]. These cases presented with symptoms of epigastric pain, chronic bleeding, anemia, and hematemesis. Large bowel metastasis from lung cancer is also very rare and in an autopsy study from Japan, the colonic metastasis rate was only 0.5% [1].

Patients with GI metastasis of lung cancer are often asymptomatic. The most common symptoms are abdominal pain, nausea, vomiting, anemia, and weight loss [2, 12]. GI metastasis of lung cancer can cause serious complications, such as perforation, obstruction, intussusception, and GI hemorrhage. These findings generally present after the diagnosis of the primary disease, but can occur synchronously or before diagnosis of the primary lesion [12-14].

The most common histological types of lung cancer that cause small bowel perforation are adenocarcinoma and squamous cell carcinoma [4]. GI bleeding developed more commonly in large cell carcinoma [15]. The pathogenesis of small bowel metastasis has been thought to be tumor cell spread via the hematogenous and lymphatic routes. The tumor invades the full thickness of the bowel with permeation of the submucosal vessels. As a result, the blood supply may be compromised, leading to the observed necrosis and ulceration of the mucosa. Thus, ischemia is likely the underlying mechanism for hemorrhage, and may also predispose the bowel to perforation [16]. Chemotherapy of the primary bronchogenic carcinoma with small intestine and stomach tumor infiltration has been reported to lead to tumor necrosis and small bowel or stomach perforation [17].

The prognosis of patients with GI metastasis of lung cancer is extremely poor. In reported series [1, 18], the mean survival was 66 days, with 50% of patients not surviving beyond 30 days. Survival time was not affected by the therapy to the primary site of the cancer or its metastases [19]. This indicates that small bowel metastasis is a poor prognostic indicator of lung carcinoma.

Because of improvements in chemotherapy, supportive care for lung cancer patients, and the extension of life expectancy, we may encounter an increasing number of cases of GI metastases in the future. Thus, we should pay attention to GI metastatic signs such as GI bleeding, abdominal pain, nausea, vomiting, or less commonly, ileus. Development of GI symptoms after chemotherapy should be carefully managed in patients with GI metastasis because of the possibility of chemotherapy-induced perforation.

In conclusion, acute abdomen as a presentation of lung cancer metastasis is rare. The small bowel is the main location in the GI tract where

hemorrhage or perforation occurs. When life-threatening complications such as perforation or intussusception develop, they often need emergency surgery and subsequent adjuvant chemotherapy. However, a poor prognosis is still predicted.

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原發性肺部黏液表皮樣癌合併腸胃道轉移：病例報告

陳俊升　彭殿王　李毓芹

肺癌是一常見之腫瘤且常常會有其他器官之轉移，如肺臟、肝臟及腎上腺。然而，肺癌產生腸胃道之轉移仍然相當少見。而當肺癌產生腸胃道之轉移時，常常是沒有症狀的，如果有產生症狀，最常見的則是腸胃道出血、腹痛，腸阻塞及腸穿孔。而最常產生小腸轉移的原發性肺癌是肺腺癌。當肺癌合併腸胃道轉移時，通常代表著相當不好的預後。我們在這一邊提出一個80歲的男性病患，他本身是粘液表皮樣肺癌合併肺臟及縱膈腔淋巴結轉移之第四期肺癌患者。此患者突然產生急性腹痛，經電腦斷層檢查診斷為腹部中空器官破裂。經緊急剖腹手術發覺為回腸穿孔，手術取下之標本證實此為原發性肺部腫瘤合併腸胃道轉移所致。然而此病患在手術後約兩周後還是不治死亡。*(胸腔醫學* ***2011; 26: 99-103)***

關鍵詞：肺癌，腸胃道轉移腫瘤

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Congenital Bronchial Atresia Presenting as a Cavitary Lesion – A Case Report

Chia-Hsiang Li, Yi-Heng Liu, Chih-Yen Tu, Chuen-Ming Shih, Wu-Huei Hsu, Chia-Hung Chen

Bronchial atresia is a rare congenital anomaly usually identified as an incidental finding during routine examinations in adulthood. We report a case in which routine chest radiograph revealed bronchial atresia with a cavitary lesion surrounded by hyperinflated lung tissue and few vascular markings. Chest computed tomography (CT) revealed a cavitary lesion with air-fluid level and segmental emphysematous changes. Bronchoscopy revealed a very small orifice at the beginning of the bronchus.

The initial chest radiograph findings of bronchial atresia can mimic those of many pulmonary diseases. The diagnosis of congenital bronchial atresia can be confirmed on the basis of chest CT findings. Bronchoscopy can be helpful in excluding the possibility of other pulmonary diseases in doubtful cases. ***(Thorac Med 2011; 26: 104-107)***

Key words: bronchial atresia, cavitary lesion, computed tomography

Introduction

Bronchial atresia was first reported by Ramsay and Byron in 1953 [1]. It is an uncommon anomaly caused by focal obliteration of the bronchial lumen and the absence of communication between a lobar, segmental or subsegmental bronchus and the central airway [2-4]. Stenosis results in the collection of mucus in the bronchi and formation of a bronchocele. In most patients with bronchial atresia, the anomaly is detected as an incidental finding during routine examinations. Bronchial atresia is associated with various abnormalities, including pulmonary sequestration, congenital adenomatoid malformation, congenital lobar emphysema, bronchogenic cyst, pericardial defects, and aplastic lungs [2-4]. Herein, we report a case of bronchial atresia with a tubular mass, which was diagnosed by chest radiography. The chest computed tomography (CT) and bronchoscopy findings confirmed this diagnosis.

Case Report

A 17-year-old non-smoking woman underwent routine chest radiography during a health check-up. Her chest radiograph revealed a

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cavitary-like lesion surrounded by hyperinflated lung tissue with a few vascular markings on the left upper lobe (Figure 1). She was symptom-free. The results of all laboratory tests and physical examinations were normal.

A chest CT scan revealed a thin-walled cavitary lesion with air-fluid level and emphysematous changes in the anterior segment of the left upper lobe. These findings were consistent with those for mucocele. No other pulmonary masses were visible on the CT image (Figure 2). Bronchoscopy revealed a very small orifice at the beginning of the anterior segmental bronchus of the left upper lobe, which possibly corresponded to the "atretic" anterior segment (Figure 3). The results of microbiologic and cytologic examinations of the washed specimens from the cavitary lesion were negative.

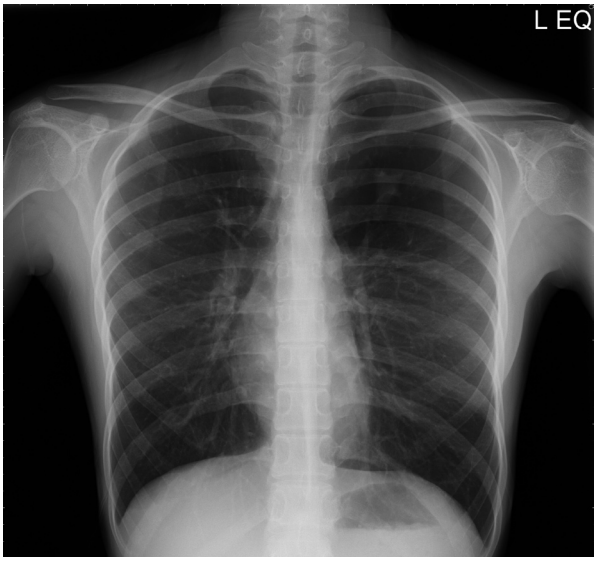

Fig. 1. Chest radiograph revealed a tubular mass surrounded by hyperinflated lung tissue with a few vascular markings on the left upper lung field.

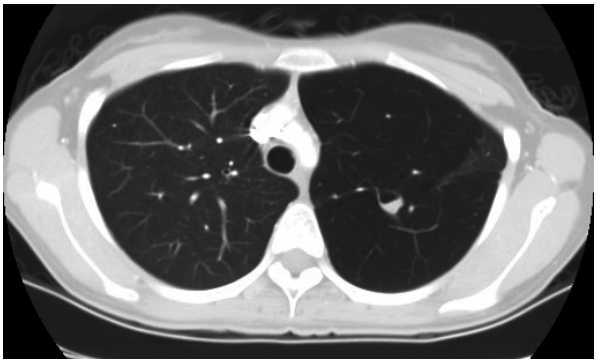

Fig. 2. Chest computed tomography scan showed a cavitary lesion with an air-fluid level and emphysematous changes in the anterior segment of the left upper lobe.

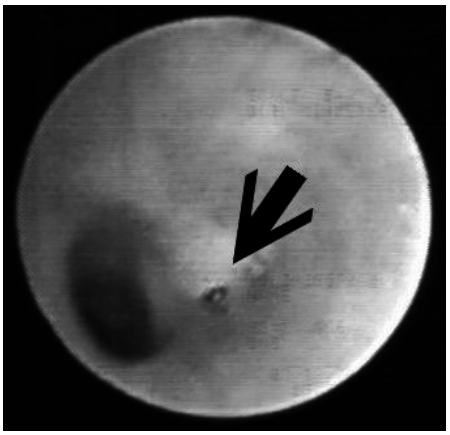

Fig. 3. Bronchoscopy revealed a very small orifice at the beginning of the anterior segmental bronchus of the left upper lobe.

The patient was diagnosed with congenital bronchial atresia and received no further intervention. Follow-up examination 1 year later revealed that she was asymptomatic, and no changes were observed in the chest CT scan.

Discussion

Bronchial atresia is a pulmonary anomaly of unknown cause in which a segmental bronchus does not communicate with the central airways. Although a pathological report is not available for our patient, the combination of mucocele, bronchial occlusion and local emphysematous changes supports our diagnosis of bronchial atresia, on the basis of Matsushima's criteria [5].

There are 2 possible theories about the

pathogenesis of bronchial atresia. First, the separated primitive bronchial bud may have continued to develop, but lost connection with the central airway. The second proposal has attributed its cause to a local vascular insult which resulted in failure of canalization of the bronchial buds [9].

Bronchial atresia is characterized by a branching mass or mucocele formed by the mucus-filled dilated bronchi distal to the atretic segment. The lung hyperinflation is caused by collateral ventilation from the adjacent normal lung via a check valve mechanism; this ventilation occurs through the pores of Kohn and the channels of Lambert [3, 9]. Bronchial atresia usually involves a segmental bronchus and is most commonly found in the posterior apical segment of the left upper lobe [6-9]. This condition is usually diagnosed in the second or third decade of life and exhibits male predominance, with an estimated prevalence of 1.2 cases per 100,000 males [10]. About half to two-thirds of the reported patients were asymptomatic before diagnosis. Recurrent pneumonia, dyspnea, cough, or hemoptysis has been reported in some cases [10].

Congenital bronchial atresia is a rare and benign condition. Initial radiographic examination of patients with this condition may occasionally reveal findings that resemble those of serious underlying pulmonary diseases. The criteria proposed by Matsushima *et al*. (mucocele, bronchial occlusion and local emphysema) may serve as an objective basis for its diagnosis. [5]

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以開洞性病灶來表現的支氣管閉鎖症—病例報告

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支氣管閉鎖症通常是意外發現的肺部結構異常，它在主要氣道和細支氣管間失去了交通性。影像學上的表現常可和其他疾病混淆。我們提出一位年輕女性，在常規的胸部放射線檢查中發現管狀的腫塊，電腦斷層表現為開洞性的病灶伴隨周邊的肺氣腫變化。支氣管鏡檢查顯示在左上葉的前分支開口處狹窄。細胞學和細菌培養皆為陰性。電腦斷層是最佳的檢查利器，而支氣管鏡檢查可協助排除其他疾病。
(胸腔醫學 ***2011; 26: 104-107)***

關鍵詞：支氣管閉鎖症

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Interventional Bronchoscopy Using Flexible Bronchoscope and Metallic Stent without Fluoroscopy to Liberate a Patient from Acute Respiratory Failure Due to Esophageal Cancer-Related Tracheal Invasion – A Case Report

Cheng-Nan Yeh, Hung-Jen Chen, Chia-Hung Chen, Chih-Yen Tu, Te-Chun Hsia, Chuen-Ming Shih

Esophageal cancer with airway invasion can cause central airway obstruction. Acute respiratory failure is 1 of the most severe complications of patients with central airway obstruction, and in these patients, interventional bronchoscopic procedures with implantation of self-expandable metallic stents (SEMSs) can facilitate weaning from mechanical ventilation. We describe a modified method that was used to successfully liberate an esophageal cancer patient who was intubated with a 6.5-mm endotracheal tube (ETT) and who was ventilator-dependent due to advanced esophageal cancer-related central airway obstruction. We implanted an Ultraflex covered stent using a flexible bronchoscope without insertion through the 6.5-mm ETT and the patient was weaned from the ventilator shortly after the procedure. ***(Thorac Med 2011; 26: 108-113)***

Key words: esophageal cancer, respiratory failure, mechanical ventilation, Ultraflex

Introduction

The most common malignancies that invade the central airway are lung cancer (30%) and esophageal cancer (30%) [1-2]. Acute respiratory failure is 1 of the most severe complications of esophageal cancer patients with central airway obstruction. Interventional bronchoscopic procedures with or without implantation of self-expandable metallic stents (SEMSs) can facilitate weaning from mechanical ventilation [3-5]. Traditionally, rigid bronchoscopy under general anaesthesia and flexible bronchoscopy under fluoroscopic guidance have been the most common methods utilized for therapeutic bronchoscopic procedures and stent implantation in mechanical ventilator-dependent patients. However, some patients are not ideal candidates for transfer to the operation room for rigid bronchoscopy with a general anaesthesia because of

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the severity of their illness and comorbidities. Moreover, the use of fluoroscopy equipment and additional radiation exposure limit the availability of fluoroscopy in the intensive care unit (ICU). We report the successful liberation from a ventilator of a ventilator-dependent esophageal cancer patient who was intubated with a 6.5-mm endotracheal tube (ETT) due to advanced esophageal cancer-related central airway obstruction using a flexible bronchoscope with a SEMS implantation without fluoroscopic guidance in an ICU.

Case Report

A 47-year-old man was diagnosed with esophageal squamous cell carcinoma (cT-4N1M0 stage IIIb) in February 2010. His chest computed tomography (CT) scan revealed that the esophageal cancer had invaded the trachea (Figure 1). He was admitted for jejunostomy to prepare for concurrent chemoradiotherapy (CCRT) for his esophageal cancer on 1 May 2010; however, prior to beginning CCRT, he experienced progressive dyspnea with stridor. Ten days after admission, acute respiratory failure developed, along with ventilator dependence due to the central airway obstruction; he then was intubated with a 6.5-mm ETT due to the narrowing trachea (Figure 2). The patient was not a good candidate for surgery or transfer to an operation room for rigid bronchoscopy with a general anesthesia because of the severity of his illness and his advanced esophageal cancer. Therefore, we chose to use flexible bronchoscopy with stent implantation without fluoroscopic guidance in the ICU. The following day, we inserted the bronchoscope (BF-260; Olympus; Tokyo, Japan) through the patient's oral cavity under local anesthesia and sedation (*i.e.*, midazolam) into the space between the tracheal wall and the 6.5-mm ETT (Figure 3A). Then, the bronchoscope was navigated to the proximal end of the lesion, and we evaluated the length of the airway stenosis using a flexible bronchoscope after we had slowly pulled back the 6.5-mm ETT. A guide wire was inserted through the bronchoscopic channel, passing through the

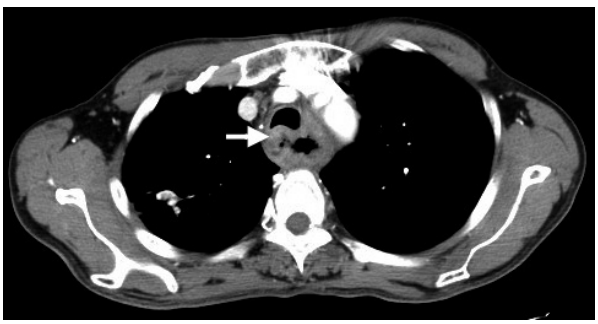

Fig. 1. The chest computed tomography (CT) in February 2010 revealed esophageal cancer (white arrow) with tracheal invasion-related tracheal stenosis.

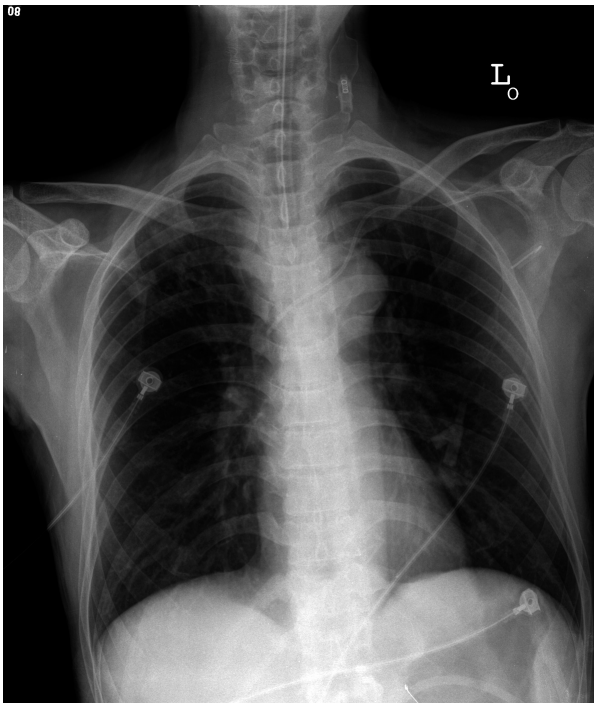

Fig. 2. Chest radiography revealed a 6.5-mm endotracheal tube in the stenotic trachea.

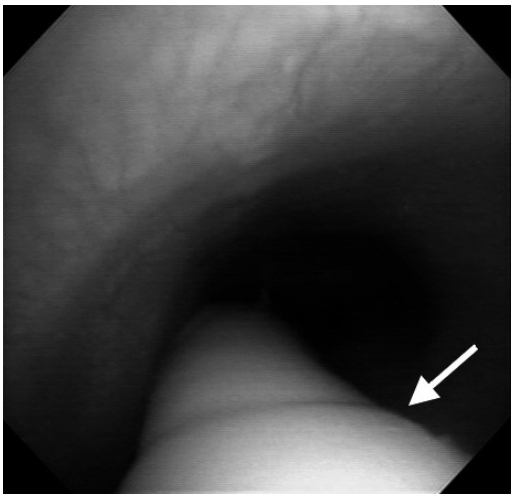

Fig. 3A. The bronchoscope was inserted into the space between the tracheal wall and the 6.5-mm ETT (white arrow).

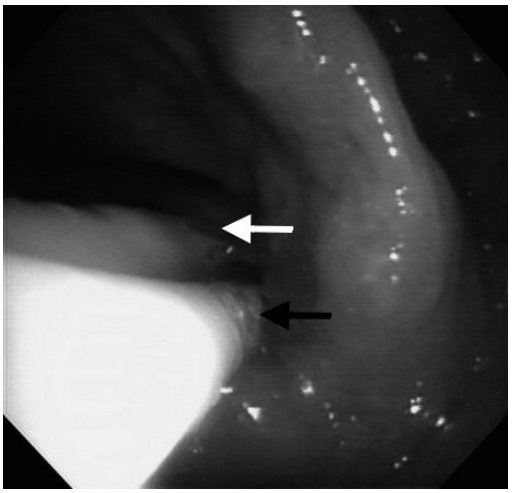

Fig. 3C. A SEMS (black arrow) was inserted between the tracheal wall and the 6.5-mm ETT (white arrow) through the vocal cord.

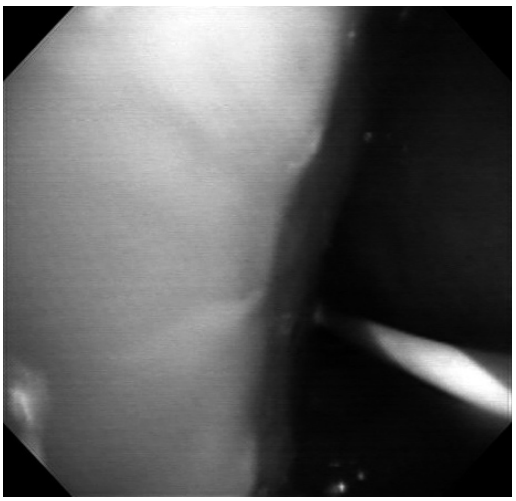

Fig. 3B. The guide wire was inserted through a flexible bronchosope to the lesion.

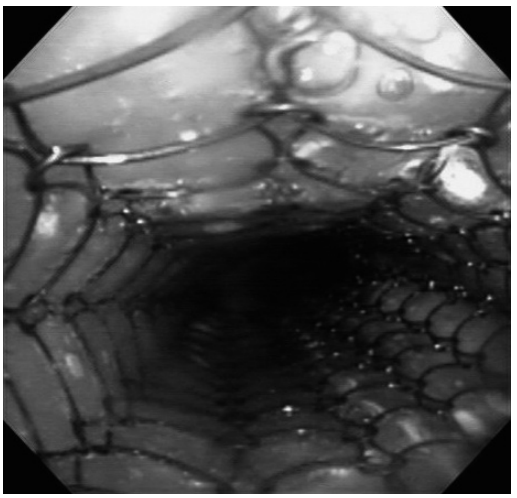

Fig. 3D. An Ultraflex stent (Boston Scientific) was successfully implanted and restored airway patency.

lesion site (Figure 3B), and the scope was then removed. The bronchoscope was reintroduced into the space between the tracheal wall and the 6.5-mm ETT to inspect the location of the guide wire, and was positioned next to the guide wire (Figure 3C). Under direct bronchoscopic visualization, the delivery catheter (Boston Scientific) was advanced over the guide wire to release the Utrafllex covered stent (18 mm x 6 cm) (Figure 3D). The delivery catheter and guide wire were then withdrawn, leaving the bronchoscope to check the position of the stent. Fifteen minutes were needed to finish our procedure, and there was no desaturation <90%, bleeding or hypotension (systolic blood pressure <90 mmHg) during the procedure. Soon after the tracheal stent implantation, the patient was liberated from the ETT without ventilator dependence

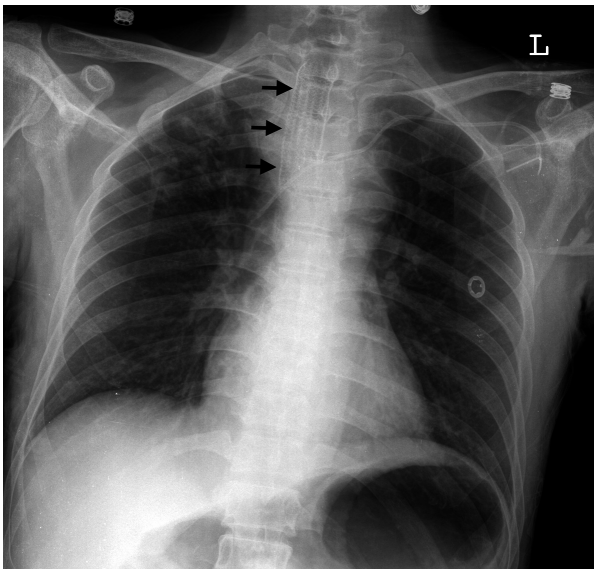

Fig. 4. Chest radiography after the procedure revealed a metallic stent (black arrows) in the tracheal lumen.

(Figure 4). After weaning, he began CCRT for his esophageal cancer.

Discussion

Advanced esophageal cancer can lead to airway involvement and has a poor prognosis. Approximately 34~50% of patients with esophageal cancer have been reported to develop airway involvement, with a slight predilection for the left bronchus [6], and 5~10% develop esophago-respiratory fistula [7]. In these patients, laser resection, electrocautery, argon plasma coagulation and stenting are techniques that can provide immediate relief, in contrast to the delayed effects of cryotherapy, brachytherapy and photodynamic therapy, in the palliative setting of alleviating central airway obstruction [8]. Metallic airway stent insertion can achieve symptomatic relief in the majority of patients with airway obstruction from both extrinsic compression and direct tumor invasion, and has also been shown to be useful in the treatment of tracheo-esophageal fistulas [9]. SEMS implantation and bronchoscopic tools can be used individually or in combination to match the needs of patients with airway disorders.

A consensus has been established that individuals presenting with imminent suffocation or respiratory failure with stenosis ≥50%, often have subtotal obstruction of the central airway [10]. Airway protection using an endotracheal tube is usually required to save the patient's life prior to referral. With the advances in airway stents and insertion techniques, the use of interventional bronchoscopic procedures has been reported to facilitate weaning from the ventilator and extubation in these patients [3-4]. The interventional bronchoscope can be used through the ETT in patients with acute respiratory failure and ventilator dependence. Clinically, there are 2 common types of airway stents (silicone stent and self-expanding metallic stent) available for interventional bronchoscopy [11]. The use of silicone stents is preferred in patients with benign conditions and can be inserted using a rigid bronchoscope. The rigid bronchoscope with a larger working channel can provide better access and allow for safer procedures with ventilator preservation, but patients need to receive general anesthesia. The self-expanding metallic stent can be implanted by using a flexible bronchoscope with fluoroscopic guidance in mechanically ventilated patients. Since some patients are not candidates for general anesthesia, and fluoroscopy equipment is not available in our ICU, a modified procedure using a flexible bronchoscope without fluoroscopic guidance to implant SEMS is feasible in patients with central airway obstruction-related respiratory failure [12]. Our patient had advanced esophageal cancer-related central airway obstruction which increased the

risk of transfer to the operation room and the use of general anesthesia. Therefore, we used our modified procedure to implant SEMSs in an ICU setting. The rate of successful weaning from the mechanical ventilator and survival after airway Ultraflex stenting was 68.7% (11 out of 16 cases) in Taiwan using this method [13].

Our patient suffered acute respiratory failure due to esophageal cancer invading the trachea and he was intubated with a 6.5-mm ETT. Traditionally, the bronchoscope or SEMSs are inserted through ETTs, but the smaller 6.5-mm ETT limited the ability to insert the bronchoscope or SEMSs. Therefore, we modified the procedure for the smaller lumen of the 6.5-mm ETT by passing the bronchoscope and SEMS through the space between the tracheal wall and the 6.5-mm ETT. This new modified method of flexible bronchoscopy with SEMS implantation without fluoroscopic guidance in the ICU is feasible and is quite possibly suitable for use in patients who have been intubated with smaller ETTs (<7.0 mm) due to central airway obstruction.

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在無放射線透視下使用軟式支氣管鏡及金屬支架讓食道癌併氣管侵犯呼吸衰竭的病人脫離呼吸器—病例報告

葉政男　陳鴻仁　陳家弘　涂智彥　夏德椿　施純明

食道癌併發呼吸道的侵犯可以造成上呼道的阻塞。而急性呼吸衰竭是上呼道阻塞最嚴重的併發症之一。在上呼吸道阻塞導致呼吸衰竭的病人，介入性支氣管鏡合併可自行擴張的金屬支架置入可加速病人脫離呼吸器。我們報告一種修改過的方式，讓一位因食道癌併上呼吸道阻塞導致急性呼吸衰竭且插了 6.5 mm 氣管內管的病人，成功的脫離了呼吸器。我們使用軟式支氣管鏡而不經由 6.5 mm 氣管內管的方式置入 Ultrflex 薄膜覆蓋式支架，經治療後病人很快的就脫離呼吸器。*(胸腔醫學* ***2011; 26: 108-113)***

關鍵詞：食道癌，呼吸衰竭，呼吸器，Ultraflex

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Cervical Bronchogenic Cyst: Report of an Adult Case

Kuan-Chun Lin, Teddy U Kuo*, Shi-Ping Luh**

Bronchogenic cysts are foregut-derived congenital lesions that are usually located in the mediastinum and detected in the pediatric patients. Rarely have they been reported in the adult neck. Herein, we report a 42-year-old male presenting with progressive respiratory distress due to airway compression by a growing left cervical mass lesion. Upon resection, a thin-walled cystic mass was noted and bronchogenic cyst was proved by its internal lining with ciliated columnar epithelium and cartilage. A review of the literature on cervical bronchogenic cyst in adults was also conducted. ***(Thorac Med 2011; 26: 114-119)***

Key words: bronchogenic cyst, cervical, adult

Introduction

Bronchogenic cysts are rare congenital malformations of ventral foregut development [1]. They are predominately located in the mediastinum or intrapulmonary regions, and found in the pediatric population [2-3]. Bronchogenic cysts located in the cervical regions of adults are extremely rare, and only 17 cases had been reported in the literature by the end of 2009 [1, 4-12]. Herein, we report an adult male who underwent surgical resection for cervical bronchogenic cyst.

Case Report

A 42-year-old male patient was referred to our clinic with the complaint of progressive dyspnea due to a growing left cervical mass found in the most recent 2 months. Upon examination, a soft, smooth, non-pulsatile mass was palpated deeply in the left lower cervical region. No dysphagia, body weight loss, hand tremor or palpitation was noted. Chest X-ray revealed the trachea was deviated to the right side, with a mass shadow on its left side (Figure 1). Chest computed tomography (CT) revealed a 6 x 5 x 4 cm cystic mass on the left lower pole of the thyroid and the paratracheal region (Figure 2). The serum-free thyroxin (free T4) and thyroid stimulating hormone (TSH) levels were normal. Cystic goiter or thyroglossal cyst was suspected pre-operatively.

Elective surgery through a left cervical lower collar curvilinear incision, with rightward extension crossing slightly over the midline, was planned for the patient. Surgical exploration revealed a cystic mass located on the left lower

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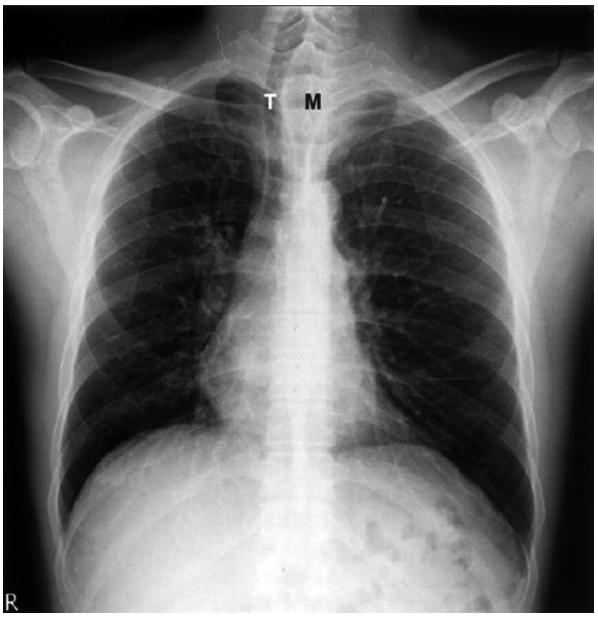

Fig. 1. Chest X-ray revealed the trachea (T) was deviated to the right side, with and a mass (M) shadow on its left side.

pole of the thyroid region, extending from the cricoid cartilage area down to the intrathoracic paratracheal region, with severe compression of adjacent structures (Figure 3). The cystic mass was completely excised uneventfully.It was grossly well-defined, and contained yellow-greenish-colored, thick and sticky mucoid fluid (Figure 4). In the microscopic exam, the cystic mass was surrounded by a thick fibrous tissue wall in which focal cartilage islands were noted. It was internally lined by ciliated columnar epithelium (Figure 5). Focal mural degeneration, hyalinizing fibrosis, foam cell collection and a cholesterol cleft were also found. Thyroid tissue was not identified.

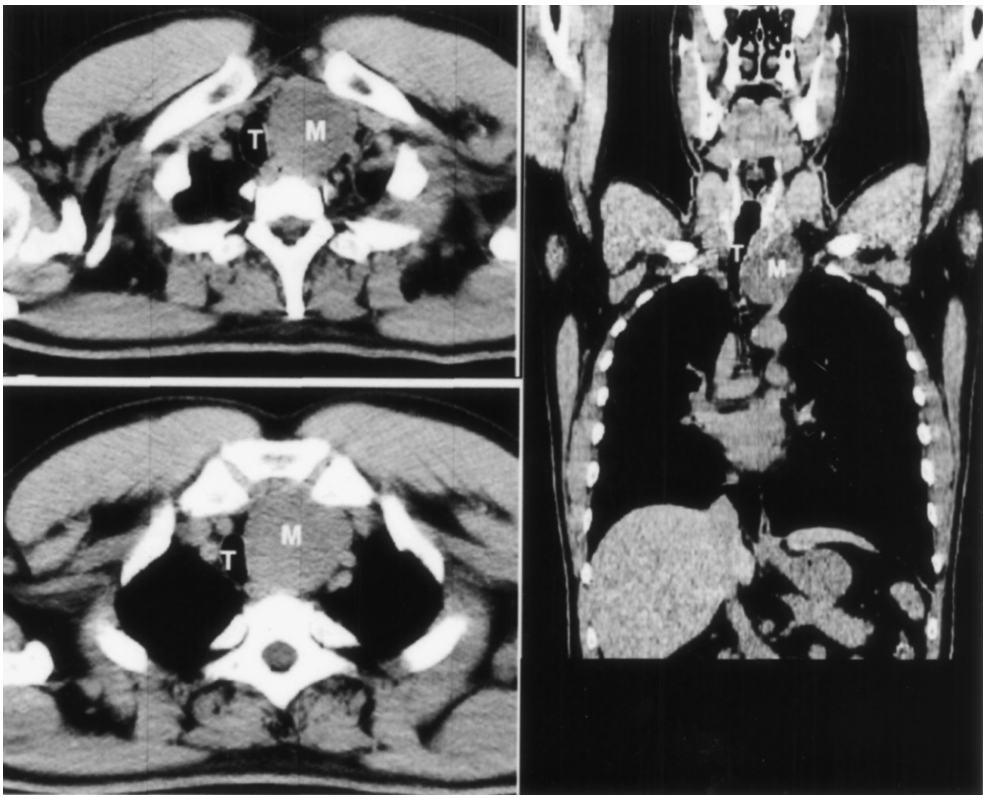

Fig. 2. Chest computed tomography (CT) revealed a 6 x 5 x 4 cm cystic mass (M) on the left lower pole of the thyroid and the paratracheal (T) region.

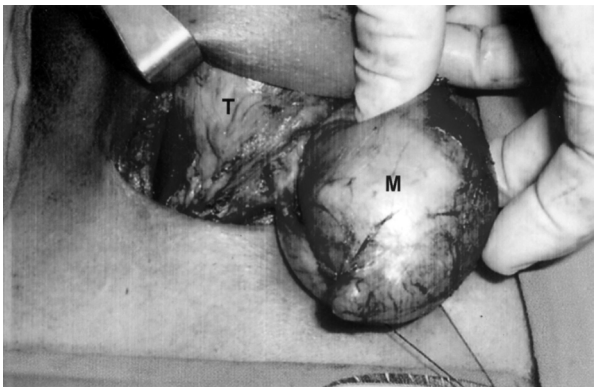

Fig. 3. Surgical exploration revealed a cystic mass (M) extending from the cricoid cartilage area down to the intrathoracic paratracheal region (T), with severe compression of adjacent structures.

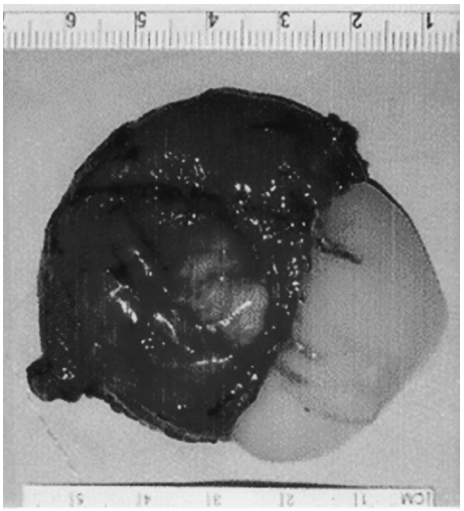

Fig. 4. Gross picture of the cyst. It was well-defined and contained yellow-greenish-colored, thick and sticky mucoid fluid.

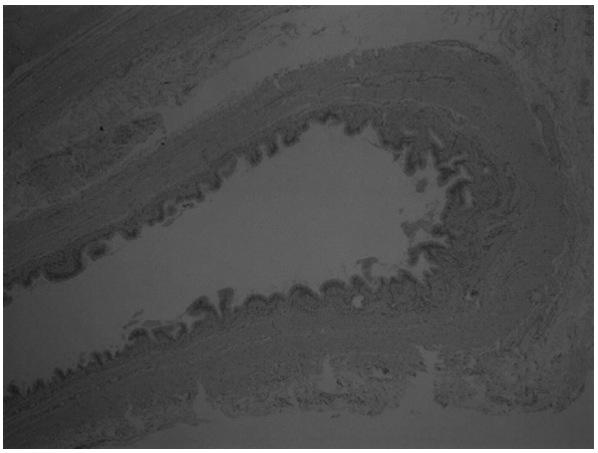

Fig. 5. In the microscopic exam, the cystic mass was surrounded by a thick fibrous tissue wall in which focal cartilage islands were noted, and internally lined with ciliated columnar epithelium.

Discussion

Bronchogenic cysts are congenital sacs that result from maldevelopment of the primitive foregut. Although they occur predominantly in the chest, there are reports of lesions in extrathoracic locations [5]. Intra-thoracic cysts are usually located in the mediastinum and intrapulmonary regions around the hilum. A fibrous cord or a patent bronchus-like tract might connect the cyst and the tracheobronchial trees [9]. Bronchogenic cysts can migrate into the superficial pre-sternal or supra-sternal or other extra-thoracic regions, such as deep and lateral cervical areas, or the subdiaphragmatic area [4, 13].

Bronchogenic cysts located in the cervical regions and in adults are extremely rare, and only 17 cases had been reported in the literature by the end of 2009 [1, 4-12]. The thyroid and para-thyroid were the most frequently affected regions, as in the present case, followed by the supra-clavicular or supra-sternal notch. The male to female ratio was four to one.

Bronchogenic cyst, either intra- or extra-thoracic, is usually asymptomatic. However, related symptoms, such as cough, dysphagia, and dyspnea with respiratory distress could appear if a cyst of the cervical area became larger and compressed adjacent vital structures. Fever, abscess, sinus drainage or fistula formation could also appear when infected [14]. The risk of developing infectious complications would be increased if communications between the cysts and airways existed [15]. Malignant changes arising from bronchogenic cysts have also been

reported, and occurred almost exclusively in adults [16-17]. Other rare but fatal complications, such as air embolism or superior vena cava syndrome, have also been reported [1, 8].

The diagnosis of cervical bronchogenic cyst should be differentiated from that of other cervical lesions, such as thyroglossal cysts, cystic goiter, branchial cleft cysts, thymic or thyroid cysts, cystic hygromas, teratomas and cystic neuromas [12]. CT or magnetic resonance imaging (MRI) can be helpful for pre-operative evaluation [18]. However, confirmation of this diagnosis requires histopathological examination of the surgical specimen, with cytology being rarely performed [4].

The choice of treatment for bronchogenic cyst is surgical resection. Surgery will be indicated when the cyst becomes infected or enlarged compressing the adjacent organs [3]. Neck exploration and dissection through a transcervical approach can remove the cyst successfully, even if it is partially extended into the thoracic cavity, as in the present case. Complete excision is mandatory to reduce the risk of recurrence or the possibility of malignant change in the cysts [16-17].

In conclusion, the possibility of a bronchogenic cyst should be considered when neck masses are noted, not only in children, but also in adults. Surgical excision is the choice for definitive treatment to prevent possible complications from the cyst.

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報告一位支氣管性囊腫發生於頸部的成人案例

林冠群　郭騰云*　陸希平**

支氣管性囊腫是一種由前腸衍生的先天性病變，通常發生部位是在縱膈腔內或是小孩族群。支氣管性囊腫發生部位是在頸部或是成人族群是很罕見的報告。本文描述一位 42 歲男性主訴有漸進性的呼吸窘迫症的症狀，主因是左側頸部有一顆正在成長中的腫塊壓迫到呼吸道。經手術切除之後，看到一個薄膜的囊狀腫塊及內層有柱狀型的纖毛上皮以及氣管軟骨，所以證實是支氣管性囊腫。*(胸腔醫學 **2011; 26: 114-119**)*

關鍵詞：支氣管性囊腫，頸部的，成人

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