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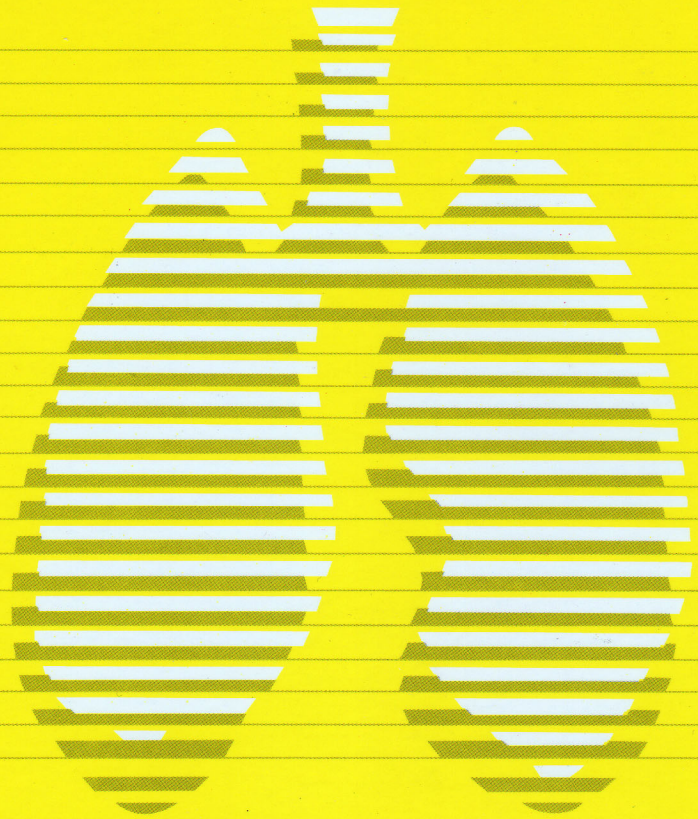

台灣胸腔暨重症加護醫學會

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原著

病例報告

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

Orginial Articles

Case Reports

Castleman's Disease in a Single Institution in Southern Taiwan

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Castleman's disease (CD) is a very rare lymphoproliferative disease, and the mediastinum is the most commonly involved site. Because of the rarity of CD, its etiology and pathogenesis are still uncertain. The initial impression of a CD-related mass is often that of a lymphoma, solitary malignancy, metastatic carcinoma or an infectious disease, rather than CD. The various types of CD are characterized by their distinctive lymphoid architectural changes in all nodal compartments, and can be divided into 4 types pathologically: unicentric, which has a hyaline vascular variant and a plasma cell variant, multicentric, and a new subtype, HHV-8-associated CD. The prognosis of CD depends on the subtype. The unicentric subtype usually has a good prognosis; however, recurrence or coexistence with malignancies or malignant sequelae is possible. In order to investigate this rare disease and its prognosis, we retrospectively reviewed all patients with CD with pathologically proven samples at Kaohsiung Medical University Hospital from 1990 to 2010. Five cases were recorded, all of which were of the unicentric type and hyaline vascular variant. The diagnoses were made based on surgical excisions, and no cases of recurrence were found after at least 1 year of follow-up, indicating the good prognosis. We present our case review and discuss the findings. ***(Thorac Med 2013; 28: 1-7)***

Key words: Castleman's disease, unicentric, hyaline-vascular type

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卡斯托曼病在南臺灣一醫院之經驗

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卡斯托曼病是一種少見的淋巴增生性疾病且縱膈腔是最容易被侵犯的地方。因為少見所以它的病因及病理機轉仍然不明，起初的表現可以類似淋巴癌、單一惡性腫瘤、轉移癌或是感染症。依據卡斯托曼病獨特的淋巴結構可以區分為四種病理型態，包括單一中心型（透明血管變異及血漿細胞變異）、多重中心型、及人類皰疹病毒第八型相關的卡斯托曼病。卡斯托曼病的預後與分型有關，單一中心型通常有較好的預後，但是其還是可能會復發或合併其他種類卡斯托曼病與惡性腫瘤同時發生。我們從 1990 年起至 2010 年 5 月回顧高雄醫學大學附設醫院所有以病理確定診斷卡斯托曼病的病例，共計有 5 例。所有病例皆為單一中心型及透明血管變異。這些病例皆有良好預後，於手術切除診斷後追蹤至少一年以上皆無復發。我們發表這個研究整理及討論我們的發現。*(胸腔醫學 **2013; 28: 1-7**)*

關鍵詞：卡斯托曼病，單一中心型，透明血管型

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Efficacy Comparison of Conventional Computed Tomography and Whole-body ^{18}F-Fluorodeoxyglucose Positron Emission Tomography for Staging in Non-small Cell Lung Cancer Patients

Ping-Chih Hsu, Chih-Hung Chen, Kung-Chu Ho*, Yi-Chen Wu**, Kuo-Chin Kao***, Chien-Ying Liu, Wen-Cheng Chang****, Ying-Huang Tsai***, Cheng-Ta Yang***

Objectives: The prognosis of non-small cell lung cancer (NSCLC) is strongly correlated with the disease stage. Both ^{18}F-fluorodeoxyglucose positron emission tomography (FDG-PET) and conventional contrast-enhanced computed tomography (CT) are commonly used for staging, and their accuracies may thus influence the clinical outcome of NSCLC treatment. In this study, we investigated the efficacy of both methods for staging, particularly mediastinal nodal staging, in NSCLC patients.

Methods: From November 2006 to July 2010, 596 newly diagnosed NSCLC patients who had received chest CT and subsequent FDG-PET for initial staging were enrolled for assessment. Of these, 173 patients who received surgical resection without neoadjuvant therapy were further analyzed based on the final pathological stage.

Results: Compared to CT, FDG-PET led to upstaging in 180 (30.2%) patients and downstaging in 29 (4.9%). The pathological results of the 173 (31.0%) patients who received surgical intervention without neoadjuvant therapy revealed that the accuracy rate in staging was 57.8% by CT and 60.7% by PET. The sensitivity, specificity, positive predictive value, negative predictive value and accuracy for mediastinal nodal staging (N2) were 42.9%, 95.0%, 52.9%, 92.3% and 88.4% by CT, and 62.0%, 96.0%, 68.4%, 94.8% and 91.9% by PET, respectively.

Conclusions: This study suggests that FDG-PET is superior to conventional CT on the clinical staging of NSCLC, but there was still a false positive rate up to 31.6% in mediastinal staging or metastasis by FDG-PET. The false positive may cause that some patients receive inadequate treatment. Therefore, if nodal involvement is suspected by either image study which may alter the decision for surgical treatment, another invasive procedure may be required for tissue proof. ***(Thorac Med 2013; 28: 8-17)***

Key words: non-small cell lung cancer, ^{18}F-fluorodeoxyglucose positron emission tomography (FDG-PET), staging, mediastinal lymph node

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傳統電腦斷層和正子掃瞄在非小細胞癌診斷分期上的準確度分析比較

徐稟智　陳志弘　何恭之 *　吳怡成 **　高國晉 ***　劉劍英　張文震 ****
蔡熒煌 ***　楊政達 ***

背景：非小細胞肺癌（non-small cell lung cancer, NSCLC）的分期和預後有重要的關連，正子掃瞄（^{18}F-fluorodeoxyglucose positron emission tomography, FDG PET）和傳統電腦斷層掃瞄已經普遍使用在非小細胞肺癌的診斷和分期上。然而這兩項檢查的準確度可能影響在臨床上的治療計畫，尤其是在縱膈腔淋巴結（N2）診斷方面。所以本研究目的在於分析這兩項檢查在非小細胞肺癌的分期準確度比較。

方法：本研究瀏覽了自 2006 年 11 月到 2010 年 7 月，596 名新診斷的非小細胞肺癌的病患，並且在初診斷時同接受正子掃瞄和傳統電腦斷層掃瞄者，分析其分期治療計畫的改變。在接受手術的病患中，以最後病理確定的診斷分期去比較這兩者檢查在分期上的準確度。

結果：相較於傳統電腦斷層，加上全身正子掃瞄之後，有百分之三十的病人分期上的改變是上升的，另外百分之四點九的病人分期下降，因此，百分之二十六點三的病人會改變原本的治療計畫。其中有一百七十三名病人在沒有接受任何前置輔助治療（Neoadjuvant therapy）下直接接受手術治療，以這些手術後的病理組織作為最後分期診斷的根據比較，電腦斷層的準確率為百分之五十七點八，全身正子掃瞄為百分之六十點七。對於縱膈腔淋巴結（N2）分期的診斷靈敏度（Sensitivity）、特異度（Specificity）、陽性預測值（Positive predictive value）、陰性預測值（Negative predictive value）和準確率（Accuracy），在電腦斷層分別為 42.9%, 95.0%, 52.9%, 92.3% 及 88.4%，而在正子掃瞄方面為 62.0%, 96.0%, 68.4%, 94.8% 及 91.9%。

結論：該研究顯示出正子掃瞄（FDG PET）對於非小細胞肺癌的臨床分期上是稍優於電腦斷層。但是在在縱膈腔淋巴結（N2）或遠處轉移分期診斷方面仍有三成左右的偽陽性率，這也可能使得有些病人失去痊癒治療的機會。因此，當這兩項檢查有懷疑癌細胞縱膈腔淋巴結侵犯時，或許進一步侵襲性的如經內視鏡超音波穿刺或縱膈腔鏡去直接取得組織證實是建議必要的。*(胸腔醫學* ***2013; 28: 8-17)***

關鍵詞：非小細胞肺癌，正子掃瞄，分期，縱膈腔淋巴結

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Pulmonary Alveolar Proteinosis Combined with Pulmonary Aspergillosis in an Elderly Patient: A Case Report and Literature Review

Ping-Hsien Tsou, Pin-Kuei Fu, Yi-Hsien Chen*, Gwan-Han Shen, Jeng-Yuan Hsu

Pulmonary alveolar proteinosis (PAP) is a rare disease characterized by the accumulation of surfactant lipids and proteins in the alveoli of the lungs, leading to respiratory insufficiency and alveolar macrophage dysfunction. The median age at diagnosis is 42 years. It is uncommon in the elderly and has a high morbidity, mainly from respiratory failure and infection. We report the case of a 74-year-old female diabetic patient with PAP and pulmonary aspergillosis complicated by severe hypoxemic respiratory failure and long-term ventilator dependence. The hypoxemia was resolved, lung infiltrates reduced, and the patient was weaned from the ventilator after successful anti-fungal therapy and sequential unilateral whole lung lavage. ***(Thorac Med 2013; 28: 18-24)***

Key words: pulmonary alveolar proteinosis, aspergillosis, long-term ventilator dependence

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老年病人肺泡蛋白沉積症合併肺麴菌感染：病例報告與文獻回顧

鄒秉誠　傅彬貴　陳奕先 *　沈光漢　許正園

肺泡蛋白沉積症是一種罕見的疾病。此疾病與肺表面活性劑在肺部累積，肺泡巨噬細胞失能有關，導致呼吸困難與肺部免疫機能缺失。此疾病診斷的平均年齡為 42 歲，老年人的肺泡蛋白沉積症並不常見。老年人常見的併發症包括嚴重呼吸衰竭與繼發機會性感染。我們報告一個 74 歲的肺泡蛋白沉積症病人，合併嚴重低血氧呼吸衰竭與肺麴菌感染。病人在抗麴菌治療後，接受了單側循序性的全肺灌洗，成功的治療了呼吸衰竭，並脫離了長時間依賴的呼吸器。*(胸腔醫學 **2013; 28: 18-24**)*

關鍵詞：肺泡蛋白沉積症，麴菌，長期呼吸器依賴

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Life-threatening Upper Airway Obstruction Caused by Massive Subcutaneous Emphysema during Mechanical Ventilation: A Case Report

Jiann-Hwa Kao, Wen-Kuang Yu, Te-Cheng Lien

Subcutaneous emphysema usually causes pain and swelling, and requires conservative treatment only. We present a case with severe subcutaneous emphysema during positive pressure mechanical ventilation. Life-threatening complications developed in our patient, including intubation difficulty due to upper airway obstruction and occult pneumothorax. We performed an infraclavicular skin incision for decompression, after which endotracheal intubation could be carried out. Computed tomography (CT) after intubation clearly revealed a collapsed extrathoracic upper airway, which was compressed by extremely inflated soft tissue. CT also showed occult pneumothorax not found in the chest X-ray. Occult pneumothorax was also managed by tube thoracostomy. We believe subcutaneous emphysema in mechanically ventilated patients should be considered an alarm sign sometimes deserving of more than conservative treatment. ***(Thorac Med 2013; 28: 25-30)***

Key words: subcutaneous emphysema, upper airway obstruction, occult pneumothorax

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於呼吸器使用時，嚴重的皮下氣腫造成之致命的上呼吸道阻塞：個案報告

高建華　余文光　連德正

皮下氣腫一向被認為只會影響到美觀，而且只需要保守性的治療。我們提出一個病患在正壓呼吸器使用的期間，產生了嚴重的皮下氣腫。很重要地，致命的併發症發生在我們的病患身上，包括了上呼吸道阻塞造成的插管困難，和隱藏性的氣胸。急救過後的胸部電腦斷層上顯示出，因為皮下軟組織嚴重的充氣，而被壓扁的胸廓外上呼吸道。我們緊急地在病人的鎖骨下方，進行皮膚切開以便減壓，之後，病人順利插入氣管內管，電腦斷層上亦顯示出 X 光上無法發現的隱藏性氣胸，氣胸也趕緊小心處理。我們相信，在使用呼吸器的病人中，皮下氣腫是一個警訊，而且需要的絕對不只是保守性治療。*(胸腔醫學* ***2013; 28: 25-30)***

關鍵詞：皮下氣腫，上呼吸道阻塞，隱藏性氣胸

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Tracheobronchomegaly and Ankylosing Spondylitis: A Case Report and Literature Review

Meng-Zhi Han, Han-Yu Chang

Tracheobronchomegaly, also called Mounier-Kuhn syndrome, is a distinct condition that involves marked dilation of the trachea and central bronchi, in association with chronic respiratory tract infections. It probably results from a congenital defect of the elastic and muscle fibers within the tracheal and bronchial walls. Secondary tracheobronchomegaly associated with Marfan syndrome, connective tissue diseases, ataxia-telangiectasia, ankylosing spondylitis, cutis laxa, light chain deposition disease, and others, has also been described. In patients with ankylosing spondylitis, the most commonly described pulmonary manifestations are upper lobe fibrosis, mycetoma formation, and pleural thickening. Tracheobronchomegaly is a rare finding in ankylosing spondylitis. We reported a male patient with both tracheobronchomegaly and ankylosing spondylitis. ***(Thorac Med 2013; 28: 31-37)***

Key words: tracheobronchomegaly, Mounier-Kuhn syndrome

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氣管支氣管巨大症與僵直性脊椎炎：病例報告及文獻回顧

韓孟志　張漢煜

氣管支氣管巨大症又被稱為 Mounier-Kuhn 症候群，是一種在氣管及支氣管會有顯著擴張且合併有慢性呼吸道感染的獨特疾病。其病因源於氣管及支氣管壁的彈性及肌肉組織之先天性缺陷。氣管支氣管巨大症亦可和馬凡氏症候群，結締組織疾病，毛細血管擴張性運動失調，僵直性脊椎炎，皮膚鬆弛症及輕鏈沉積病等疾病有所關聯。在此，我們報告一個有氣管支氣管巨大症合併僵直性脊椎炎的男性案例。*(胸腔醫學 **2013; 28: 31-37**)*

關鍵詞：氣管支氣管巨大症，Mounier-Kuhn 症候群

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Pitfall in Diagnosing Lymphoepithelioma-like Carcinoma of the Lung: A Case Report and Literature Review

Yi-Mou Wu, Gwan-Han Shen, Jeng-Yuan Hsu

Lymphoepithelioma-like carcinoma (LELC) of the lung is a rare form of lung cancer. The diagnosis requires tissue biopsy for histological and immunohistochemical stain. The prognosis is better than for other non-small cell lung cancers (NSCLC). We report a 60 year-old man who presented with a central pulmonary mass and pleural effusion. Scanty tissue from endobronchial ultrasonography with transbronchial needle aspiration (EBUS-TBNA) resembled squamous cell carcinoma morphologically. But biopsy via thoracoscopy showed high grade tumor cells arranged in a solid sheet pattern, infiltrated by varying numbers of lymphocytes. *In situ* hybridization for Epstein-Barr virus (EBV) encoded RNA showed localization of EBV genomes within the nuclei of tumor cells. LELC of the lung was diagnosed, and the patient received platinum-based chemotherapy. ***(Thorac Med 2013; 28: 38-43)***

Key words: lymphoepithelioma-like carcinoma (LELC), endobronchial ultrasonography transbronchial needle aspiration, squamous cell carcinoma, diagnosis

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診斷肺原發類淋巴上皮細胞癌上的陷阱－病例報告與文獻回顧

吳宜謀　沈光漢　許正園

肺原發類淋巴上皮細胞癌是一少見的肺癌。診斷需要組織切片來做免疫組織化學染色。其預後比其他的非小細胞肺癌好。我們報導一個 60 歲男性表現出肺中心部腫瘤併肋膜腔積液。氣管內視鏡超音波經氣管細針抽吸得到的少量細胞，形態學上像鱗狀上皮細胞癌。但經胸腔鏡切片發現高惡性度細胞排列成實質片狀，有不等的淋巴球浸潤且雜交檢測 EB 病毒編碼的 RNA 發現 EB 病毒基因在癌細胞核中。診斷為肺原發類淋巴上皮細胞癌且病人接受鉑金類為主的化學藥物治療。*(胸腔醫學 **2013; 28: 38-43**)*

關鍵詞：肺原發類淋巴上皮細胞癌（LELC），氣管內視鏡超音波經氣管細針抽吸，鱗狀上皮細胞癌，診斷

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A Primary Endobronchial Schwannoma Coexisting with an Ipsilateral Lung Adenocarcinoma: A Case Report and Literature Review

Po-Chiang Chan, Chun-Chi Chang, Ching-Hsiung Lin

Primary endobronchial schwannomas are relatively rare, benign tumors of neurogenic origin. Their presence within the bronchus may lead to cough, hemoptysis, dyspnea and obstructive pneumonia. We present the case of an 83-year-old man with a primary endobronchial schwannoma simultaneously occurring with an ipsilateral bronchogenic adenocarcinoma, and review the literature on primary endobronchial schwannomas. To date, primary endobronchial schwannomas coexisting with lung adenocarcinomas have not been reported, and their relationship is still unknown. We should always keep in mind that endobronchial benign tumors may mimic endobronchial involvement of malignancies. Bronchofiberscopic examination with biopsy is warranted for any endobronchial tumors, and will have an important influence on the staging, treatment, and outcome of coexisting cancers. ***(Thorac Med 2013; 28: 44-50)***

Key words: endobronchial schwannoma, lung adenocarcinoma

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原發性支氣管內許旺細胞瘤併存同側肺腺癌：病例報告與文獻回顧

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原發性支氣管內許旺細胞瘤是相對罕見的神經原性良性腫瘤。生長於氣管內可能導致咳嗽、咳血、呼吸困難與阻塞性肺炎。我們報告一位 83 歲男性同時存在原發性支氣管內許旺式細胞瘤與同側肺腺癌，並整理回顧支氣管內許旺式細胞瘤的相關文獻。就我們所知，文獻中未曾有與同側肺腺癌併存之支氣管內許旺細胞瘤的個案報告，而兩者之間的相關性依然未知。我們必須留意支氣管內良性腫瘤可能相似於氣管內轉移之惡性腫瘤的情形，因此針對支氣管內腫瘤進行軟式支氣管鏡切片是必要的，對於併存之癌症的分期、治療及預後有重要的影響。*(胸腔醫學 **2013; 28: 44-50**)*

關鍵詞：支氣管內許旺細胞瘤，肺腺癌

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Obstructive Sleep Apnea and Hypopnea Syndrome Originating from an Unusual Anatomical Obstructive Site – A Case Report

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Bilateral vocal cord palsy is not an uncommon complication in patients who have experienced neck or thoracic surgery. We describe a male patient who had a past history of thyroid cancer with mediastinum invasion and recurrent laryngeal nerve impairment. Post-thyroidectomy flexible laryngoscope examination revealed bilateral vocal cord palsy. He was referred to the sleep outpatient services due to the new onset of loud snoring when sleeping following the tracheostomy decannulation. Full-night polysomnography (PSG) showed severe obstructive sleep apnea and hypopnea syndrome (OSAHS), and flow-volume loop revealed variable extra-thoracic upper airway obstruction. The level of larynx abnormality contributing to OSAHS has not been clearly determined until now. We present this case to emphasize that laryngeal examinations and flow-volume loop should be implemented in patients with a history of neck or thoracic surgery and a new onset of loud snoring. ***(Thorac Med 2013; 28: 51-56)***

Key words: vocal cord palsy, obstructive sleep apnea and hypopnea syndrome

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罕見解剖構造異常引起阻塞性睡眠呼吸中止症－病例報告

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對於經歷頸部或胸腔手術的患者而言，術後因喉返神經受損引起雙側聲帶麻痺並非少見之併發症。在此病例報告中，一位男性甲狀腺癌病人術後纖維喉鏡檢查發現雙側聲帶麻痺並接受氣切手術。歷經階段性氣切更換與吞嚥訓練後移除氣切。但自氣切移除後，病患開始出現睡眠時打鼾之情況而轉介睡眠門診。經評估後，此病患並無常見引發睡眠呼吸中止症之上呼吸道解剖構造異常。經多頻道睡眠生理檢查顯示有重度阻塞性睡眠呼吸中止症。對於喉部構造異常引起之阻塞性睡眠呼吸中止症目前仍只有少數病例報告發表且治療仍以手術方式為主。我們藉此病例報告提醒對於曾接受頸部或胸腔手術患者新產生之睡眠呼吸中止症候群時，除常見引發之危險因子評估外，纖維喉鏡檢查與肺功能檢查釐清確切阻塞位置是非常重要的。
*(胸腔醫學 **2013; 28: 51-56**)*

關鍵詞：聲帶麻痺，阻塞性睡眠呼吸中止症

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Pulmonary Physiology before and after Bronchopulmonary Lavage for Pulmonary Alveolar Proteinosis – A Case Report

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Pulmonary alveolar proteinosis (PAP) is a diffuse lung disease characterized by the accumulation of lipoproteinaceous material in the alveoli. Although bronchopulmonary lavage (BPL) is the most widely accepted and effective treatment for patients with moderate to severe hypoxemia, there are few reports about the changes in pulmonary physiology before and after BPL. We report a rare case which was diagnosed as PAP with high plateau pressure. The patient received prolonged mechanical ventilation for 2 months, and was weaned from the ventilator after 2 sequential BPL sessions. We also propose an integrated algorithm which physicians can use when they perform BPL in such critically ill patients. In addition, we present the serial respiratory parameters and serial chest X-rays obtained before and after the BPL to highlight the impact of BPL on PAP in pulmonary physiology. Finally, we provide a figure to illustrate the morphological changes in the pulmonary alveoli and interstitial space, to explain the effect of BPL on PAP. This report is the first on the physiological changes in the lungs and the respiratory parameters before and after the implementation of BPL. ***(Thorac Med 2013; 28: 57-64)***

Key words: bronchopulmonary lavage, pulmonary alveolar proteinosis, plateau pressure, pulmonary physiology

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肺泡蛋白沉積症的支氣管肺泡灌洗前後生理參數變化－病例報告

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肺泡蛋白沉積症（PAP）是一種因肺泡累積脂蛋白物質導致瀰漫性肺疾病。對於中度至嚴重症狀和低血氧症患者，雖然支氣管肺泡灌洗是最被廣泛接受和有效的治療，但鮮少文獻討論支氣管肺泡灌洗前後的肺生理變化。我們報告一個病例：肺泡蛋白沉積症併發 2 個月呼吸衰竭，在接受 2 次支氣管肺泡灌洗治療後，成功脫離呼吸器，而我們也提出了一個處理嚴重肺泡蛋白沉積症的流程圖。在病患接受支氣管肺泡灌洗治療前後，我們記錄了呼吸參數及胸部 X 光片來探討支氣管肺泡灌洗對肺泡蛋白沉積症病患的肺部生理影響。最後，我們以繪圖的方式試著去形容肺泡及肺間質在支氣管肺泡灌洗的前後變化。這份報告是第一篇詳細討論支氣管肺泡灌洗前後肺部和呼吸參數的生理變化之相關文獻。*(胸腔醫學* ***2013; 28: 57-64)***

關鍵詞：支氣管肺泡灌洗，肺泡蛋白沉積症

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