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Imaging Findings of Pneumothorax Caused by Bronchial Cartilage Hypoplasia in a Dog

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Abstract : A 10-year-old, castrated poodle dog presented with a cough for 2 weeks, and the cough initially developed since very young age. On radiographs, pneumothorax was noticed by characteristics of radiolucent area without pulmonary markings along the thoracic wall and diaphragm, retracted lung lobes from the thoracic wall and severely decreased volume of the left cranial lung lobe with disconnected bronchus. Computed tomography (CT) findings identified several pulmonary air-filled cysts and collapsed lung with abnormal shape and non-tapered end of bronchus, bronchioles at the accessory lobe and left cranial lobe. Also, pneumothorax, pneumomediastinum and subcutaneous emphysema were found. Imaging diagnosis was the spontaneous pneumothorax caused by ruptured emphysematous bullae associated with congenital bronchial cartilage abnormality or bronchial tree malformation. On surgery, hypoplasia of the left cranial lobe, right middle lobe, and accessory lobe with a bulla where air was leaking was identified. The accessory lobe was partially resected and bronchial cartilage hypoplasia was confirmed by histopathologic examination.

Key words **:** bronchial cartilage hypoplasia, pneumothorax, computed tomography, lung lobectomy.

Introduction

Spontaneous pneumothorax is defined as the presence of air or gas in the pleural space, without iatrogenic or traumatic causes (8,14,16). Although reported causes include bacterial pneumonia, dirofilariasis, pulmonary abscesses and neoplasia, the primary cause of spontaneous pneumothorax in humans and dogs is rupture of subpleural bullae or blebs (7,8,16). The most common clinical signs include coughing, tachypnea, exercise intolerance, respiratory distress, depression, anorexia, and lethargy (16). Thoracic radiography is an excellent diagnostic means of spontaneous pneumothorax. However, based on previous reports, the use of thoracic computed tomography (CT) or thoracoscopy for identifying small pulmonary lesions such as pulmonary blebs and bullae is recommended due to its superior accuracy than radiographs (7, 8,16). Treatment is aimed at removing air in the pleural space by conservatively or definitively. Prognosis is good when surgical treatment involving resecting the pulmonary blebs and bullae with a partial or complete lung lobectomy is performed, while conservative treatment with thoracocentesis or thoracic drainage was not effective in resolving the pneumothorax caused by pulmonary blebs and bullae in dogs (7, 16). The purpose of this report is to describe a case of spontaneous pneumothorax caused by bullae with bronchial cartilage hypoplasia in a dog and provide radiographs and CT imaging features, intraoperative findings, and prognosis following surgical treatment.

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Case

A 10-year-old castrated male, Poodle dog weighing 7 kg with cardiac management by a referring veterinarian was referred for suspected pneumothorax. The cough has been developed when excited since very young age, and it worsened 2 weeks before. On hematology, serum biochemistry, and urinalysis, no abnormalities were detected except for mildly decreased RBC at 506 (reference interval: 570-880).

Through thoracic and abdominal radiography, pneumothorax was noticed by characteristics of radiolucent area without pulmonary markings along the thoracic wall and diaphragm, retracted lung lobes from the thoracic wall and severely

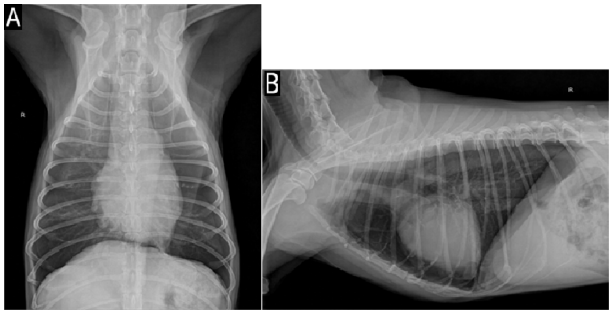

Fig 1. Survey thoracic radiographs show pneumothorax characterized by radiolucent area, retracted lung lobes, and decreased volume of the left cranial lung lobe on ventrodorsal (A) and lateral (B) views.

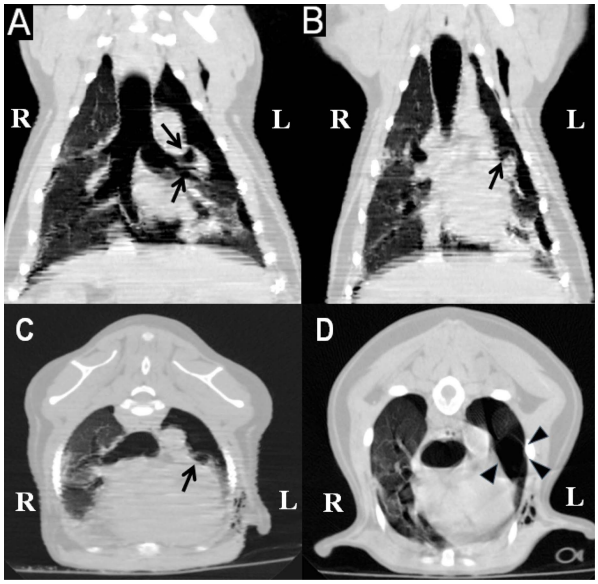

Fig 2. Dorsal (A, B) and transverse (C, D) CT images. Note the collapsed lobe with abnormally non-tapered end of bronchus (arrows) and probable bullae (arrow heads) at the left cranial lobe.

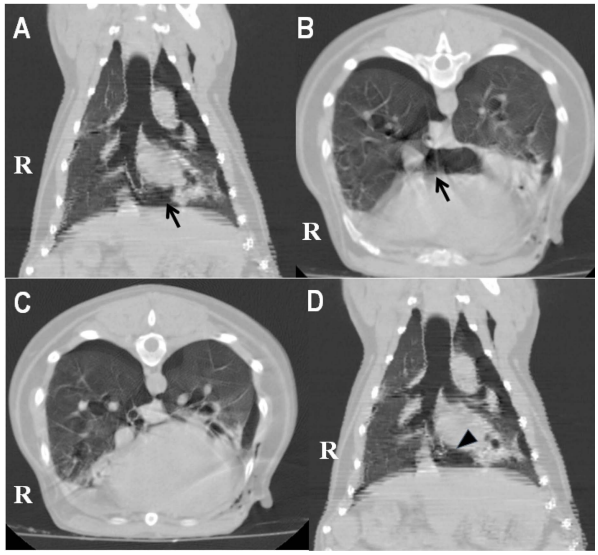

Fig 3. Dorsal (A, D) and transverse (B, C) CT images. Note the air-filled cyst (arrow head) suspected and collapsed lobe with abnormal shape of bronchus (arrows) at the accessory lobe.

decreased volume of the left cranial lung lobe with disconnected bronchus (Fig 1). Also, echocardiography was performed, compensatory hypertrophy with mitral and aortic regurgitation was identified.

CT images of the thorax were obtained from the thoracic inlet to the cranial part of L1 at 5 mm and 1 mm thickness under the general anesthesia. Several pulmonary air-filled cysts and collapsed lung with abnormal shape and non-tapered end of bronchus, bronchioles at the left cranial lobe (Fig 2) and accessory lobe (Fig 3) were identified. Also, pneumothorax, pneumomediastinum, and subcutaneous emphysema were found.

Imaging diagnosis was the spontaneous pneumothorax

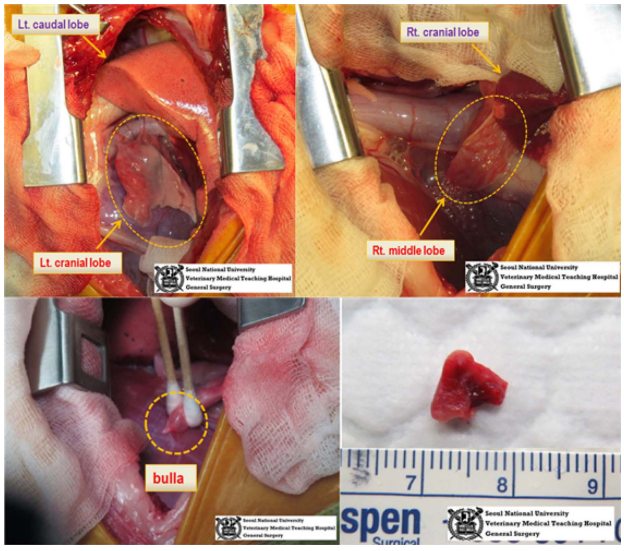

Fig 4. Hypoplasia of the left cranial lobe (A), right middle lobe (B), and a bulla (C, D) in the accessory lobe were identified on surgery.

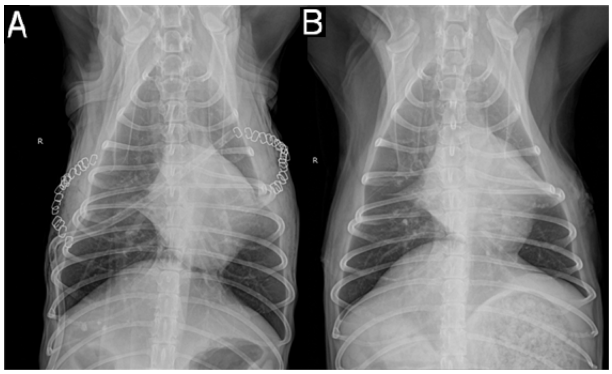

Fig 5. Survey thoracic radiographs show no signs of pneumothorax on the day of surgery (A) and on the 12 days after surgery (B).

caused by ruptured emphysematous bullae associated with congenital broncial cartilage abnormality or bronchial tree malformation.

Partial lung lobectomy of the accessory lobe was performed, and the hypoplastic left cranial lobe, right middle lobe, and accessory lobe were identified (Fig 4). Bronchial tubes and vessels running to the left cranial lobe were too short to separate with ease and there was no air leakage in this region, so partial lobectomy of the left cranial lobe was not performed. However, a bulla, approximately 0.7 cm in diameter, and air leakage in the accessory lobe were identified (Fig 4), hence the accessory lobe was partially resected including the bulla. As the bulla was removed, there was no more air leakage. A histopathologic diagnosis of resected accessory lobe was made.

On the 12th day after surgery, pneumothorax was not noticed on the thoracic radiographs (Fig 5). The dog was in a stable condition, and no characteristic symptoms were shown until 4 months after surgery.

Discussion

Pneumothorax occurs when air or gas enters the pleural space. Based on the reported studies, spontaneous pneumothoraces are mostly found in healthy, middle-aged, large-breed or deep-chested dogs that have no previous history of respiratory problems or lung disease (16). The most common clinical signs include lethargy, anorexia, depression, coughing, tachypnea, exercise intolerance, increased respiratory effort, and various degrees of respiratory distress. For some dogs, respiratory signs may develop rapidly and be very obvious, whereas for other dogs, initial clinical signs may be very nonspecific and respiratory signs may not develop until the pneumothorax progresses for over days (16).

According to the published reports in the human fields, pneumothoraces are divided into spontaneous pneumothorax, occurring without any preceding event, and traumatic pneumothorax due to direct or indirect trauma. Iatrogenic pneumothoraces, resulting from diagnostic or therapeutic medical procedures, are also categorized as traumatic pneumothoraces (12,14). Also, spontaneous pneumothoraces are divided into primary and secondary spontaneous pneumothoraces. While secondary spontaneous pneumothoraces are associated with underlying pulmonary pathology, no underlying pulmonary disease is present in patients with primary spontaneous pneumothorax.

For the primary spontaneous pneumothorax, blebs and bullae seem to be mostly associated with its pathogenesis (14). Pulmonary blebs are accumulations of air within the layers of the visceral pleura, most commonly located at the lung apices (16,18). They are formed when air escapes from within the lung parenchyma and travels to the surface of the lung and becomes trapped between the layers of visceral pleura. Grossly, blebs appear as small "bubbles" or "blister-like" lesions on the surface of the lung that range in size up to several centimeters in diameter.

On the other hand, pulmonary bullae are defined as air-filled spaces within the lung parenchyma that result from the destruction, dilatation, and confluence of adjacent alveoli (11, 16). Bullae could vary in size, with some being small (involving only a few alveoli), whereas others being very large (involving a majority of the lung) (3).

Several previous reports have described the pulmonary blebs, bullae, or bullous emphysema from dogs with spontaneous pneumothorax. However, differences in lesional terminology, description, and histopathological interpretation have resulted in conflicting information about pulmonary bleb and bulla lesions (5,15,19). In particular, the terms "bleb, bulla, and bullous emphysema" have been used interchangeably in some reports, making it difficult to determine the specific lesion being described. Also, the location and extent of the pulmonary lesions were not always reported, making it unclear as to whether lesions were focal, multifocal, or diffuse. Finally, different interpretations about the histopathological findings have resulted in uncertainty as to whether pulmonary blebs and bullae should be considered primary lesions or lesions that develop secondary to some other underlying cause.

Diagnosis and treatment of spontaneous pneumothorax in dogs appear particularly challenging, because the source of air leakage is not usually evident from the history, clinical examination, or thoracic radiographs (5,15,19).

Although thoracic radiography is effective for detection of pneumothorax, it has poor sensitivity for identifying bullae, blebs, and their location and number (7,8). In the human research, radiographic accuracy of detection for blebs and bullae ranges from 10% to 60.5% (5,6,17). In dogs, accuracy of detection for bullae and blebs on radiographs has ranged from 0% to 50% (8,15). Lesions identified radiographically could resulted in inadequate surgical approach because of underestimating both the number and location of lesions (8). Nevertheless, serial thoracic radiographs should be taken to identify other potential causes of pneumothorax such as pulmonary neoplasia, abscesses, or dirofilariasis (5,15,16).

In humans, CT is known as a more sensitive method for detection of blebs and bullae than radiography. In the human fields, accuracy of lesional detection via CT has been reported to range from 88% to 91.8% (10,18). Even in cases in which lesions are identified on radiographs, CT has several advantages in defining lesion in number, size, location, or surrounding structures, as well as enabling better differentiation of anatomic structures and their relationship to the lesions (8,17).

There are many previous studies about surgical and nonsurgical management for spontaneous pneumothorax in humans and dogs. Based on the reported results, conservative treatment with thoracocentesis or thoracostomy tube drainage should not be considered as a reliable means of treating pneumothorax caused by pulmonary blebs and bullae in dogs (16). Pneumothorax persisted or recurred in eight of eleven (73%) and seven of eight (88%) dogs with confirmed or presumed blebs and bullae after treatment with thoracocentesis or thoracostomy tube drainage (5,15,16). And pneumothorax persisted in all of twelve dogs in another study, despite of conservative treatment for 1 to 5 days (16). Therefore, surgical treatment should be pursued once other obvious causes of pneumothorax have been ruled out.

On the other hand, definitive treatment involves resecting the pulmonary blebs and bullae with a partial or complete lung lobectomy. Lesions may be present on multiple lung lobes, so each lobe should be thoroughly examined during surgery. Blebs and bullae typically appear as focal, translucent, "bubble-like" lesions mostly on the apices of the lungs, even so they could be located anywhere within the lung. The size, number, and location of the lesions on the each lung lobe will determine the amount of lung tissue that needs to be removed (16). Previous studies on spontaneous pneumothorax in dogs have reported fair to good results, with a 3% to 25% risk of recurrence after surgery (5,13,15,19).

Recently, thoracoscopy has been used diagnostically and therapeutically in small animals. Thoracoscopy compares favorably with the "open chest" procedure, because patients have less morbidity, less pain, quicker recovery, and shorter hospitalization (3,4,13). In a recent study, 3 dogs recovered quickly and fully after thoracoscopic treatment without any signs of recurrence at 18 to 29 months (4).

In this case study, several pulmonary air-filled cysts and collapsed lung with abnormal shape and non-tapered ends of bronchus and bronchioles were found on CT images, additionally, hypoplasia of the left cranial lobe, right middle lobe,

and accessory lobe was identified on surgery. Histologically, bronchial cartilage hypoplasia was diagnosed, and a bulla where air was leaking was found not in the other lung lobes but in the accessory lobe. Hence, the dog was expected to show favorable prognosis after partial lung lobectomy, and indeed has been showed progressive improvement of respiratory signs.

Conclusion

Radiographs and CT features in this case were helpful in determining resectability and eventually surgical planning about the lung lobe. The accessory lobe was partially resected, diagnosed histologically as bronchial cartilage hypoplasia and pulmonary atelectasis that was closely correlated to the imaging findings. The dog has recovered well so far.

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개에서 기관지 연골 저형성에 의해 발생한 기흉의 영상학적 진단 증례

김수연 · 김성수 · 이저순 · 윤수경 · 오현정 · 손정민 · 김보은 · 김완희 · 윤정희 · 최민철[1]

서울대학교 수의과대학

요 약 : 10년령의 중성화된 수컷 Poodle이 기흉이 의심되어 내원하였다. 어릴 때부터 기침증상을 보이다가 약 2주 전부터 악화되었으며, 혈액검사 및 요검사에서 유의적인 특이소견은 발견되지 않았다. 흉부 방사선 영상에서 기흉 소견 및 좌측 전엽 부위 기관지의 단절과 고도로 감소된 폐 실질이 관찰되었다. 흉부의 CT 영상 검사에서, 좌측 전엽과 덧엽에서 몇 개의 cyst들과 허탈된 폐가 관찰되었다. 또한 이 부위에서 기관지와 세기관지들은 비정상적인 형태로 끝부분이 가늘어지지 않았으며, 그 외에도 기흉, 기종격, 그리고 피하 기종이 확인되었다. 이러한 영상학적 특징들로부터 기종성 수포의 파열에 의해 발생한 자발성 기흉이 고려되었으며, 그 원인으로는 선천적인 기관지의 이상 또는 기관지 가지의 기형이 고려되었다. 부분적인 폐 덧엽 절제술이 실시되었으며, 좌측 전엽, 우측 중엽, 그리고 덧엽의 저형성이 확인되었고 특히 덧엽에서는 공기가 새고 있는 수포가 확인되었다. 조직병리학적 검사를 통해 기관지 연골 저형성으로 최종 진단되었다.

주요어 : 기관지 연골 저형성, 기흉, Computed Tomography, 폐엽 절제술