Note (Surgery)

Congenital lobar emphysema concurrent with pneumothorax and pneumomediastinum in a dog

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Running title: CONGENITAL LOBAR EMPHYSEMA IN A DOG
ABSTRACT

A two-year-old castrated male Pomeranian dog was referred with the chief complaints of coughing and subcutaneous emphysema. On physical examination, the crepitant areas were palpable. When auscultated, the right chest was absent of respiratory sound, while the sound of the opposite side was enhanced. Radiographs presented pneumothorax and pneumomediastinum. On computed tomography, hypoattenuated bulla-like lesion at right middle lung lobe and trapped air in mediastinum were shown. After patient stabilization, surgery for excision of affected lobe was performed. During follow-up period, there were no recurrence and complication on radiographic examination. Based on clinical and pathological findings, the dog was diagnosed as congenital lobar emphysema.

KEY WORDS: congenital lobar emphysema, dog, pneumomediastinum, pneumothorax
Congenital lobar emphysema (CLE) is rare pulmonary disease in dogs, and the majority of affected dogs were between 6 weeks to 6 months of age [1, 2, 10]. The most common clinical sign is progressive dyspnea with coughing and exercise intolerance [2, 5]. It predominantly occurs in unilateral cranial lobe or right middle lobe, and has distinct histopathological characteristics [1, 2, 10]. We present a case of CLE with concurrent pneumothorax, pneumomediastinum and subcutaneous emphysema, that was successfully corrected with surgical treatment and definitively diagnosed through histopathological staining, in an adult dog.

A two-year-old castrated male Pomeranian dog, weighing 2.7 kg, was referred to the Kyungpook National University Veterinary Medical Teaching Hospital with the chief complaints of subcutaneous emphysema and acute onset of coughing. No previous clinical sign related with respiratory tract had been noted. On physical examination, the crepitant areas were palpable on head, neck and lumbar regions. When auscultated, the right chest was absent of respiratory sound, while the sound of the opposite side was enhanced. Despite such changes, only mild tachypnea with coughing was observed. Visible mucous membrane color was normal. The dog was normothermic and had regular pulse. There were no history of trauma and noticeable superficial soft tissue injury. Complete blood count and blood chemistry values were normal.

Lateral radiographs presented the radiolucent space between heart and sternum. The cranial mediastinum had heterogeneous opacity, and major vessels of mediastinum were visualized, which can be the evidence of pneumomediastinum. In ventrodorsal radiographs, radiolucent bulla-like lesion with left shift of heart and mediastinum was visualized in the right thorax. Caudal displacement of right hemidiaphragm and
hyperlucency of right thorax, implying pneumothorax, were seen. Subcutaneous
emphysema was extended from head to the lumbar region (Fig. 1).

On computed tomography (CT), pneumothorax was obviously observed in right
thorax. The hypoattenuated bullae-like lesion on the right middle lung lobe was shown.
Pneumomediastinum was seen as trapped air around trachea, esophagus and major
vessels (Fig. 2).

After patient stabilization and sufficient oxygenation, lobectomy was performed
with right intercostal thoracotomy. The incision was made between 5th and 6th rib. Lack
of negative pressure within thoracic cavity was identified during thoracotomy. Grossly,
the middle lung lobe had extremely scanty parenchyma (Fig. 3). After the ligation of
blood vessels and bronchus, affected lung lobe was excised. When positive pressure was
given with ventilation machine, collapsed cranial, caudal and accessory lobes were
recovered to normal shape. Prior to closing the thorax, warm sterile saline was filled
within the thoracic space to confirm the absence of air leakage, and Jackson-Pratt drain
tube was placed.

Tissue samples were fixed in 10% buffered formalin, embedded in paraffin wax
and sectioned. Sections were stained with hematoxylin and eosin (HE), and serial
sections were prepared for staining with Masson-trichrome and immunohistochemistry.
The following primary antibodies were used: cytokeratin (AE1/AE3, Nichirei
Biosciences, Tokyo, Japan) and α-smooth muscle actin (SMA, Dako-Japan, Kyoto,
Japan). Each antibody was visualized using 3-3’-Diaminobenzidine (DAB, Dako-Japan)
and were counterstained with hematoxylin.
On histopathological examination, marked diffuse enlargement and over-
distention of alveoli and alveolar ducts with disruption of alveolar septa were shown
(Fig. 4). The affected bronchus was lined with low cuboidal and immature epithelium
instead of normal ciliated pseudostratified columnar epithelium. Cartilaginous
components and bronchial glands around bronchi were not observed. The visceral pleura,
alveolar septa, peribronchial spaces and interlobular septa were expanded by extensive
fibrosis. The fibrosis area was stained intensely with Masson’s trichrome (Fig. 5). A
small number of lymphocytes, plasma cells and macrophages were scattered throughout
the alveolar and peribronchial spaces, and occasionally, foamy macrophages were also
observed in alveolar lumens.

Immunostaining for \( \alpha \)-SMA demonstrates abnormal bronchial smooth muscle
architecture with disorganized muscle bundle structure (Fig. 6). In addition, \( \alpha \)-SMA
labeled the markedly increased number of pulmonary vessels surrounding bronchi. The
cuboidal epithelial cells lining the affected bronchus stained positively for AE1/AE3. In
some parts, the epithelium was pseudostratified and invaginated into the underlying
stroma (Fig. 7). Taken together, the dog was definitively diagnosed as CLE.

The histopathological findings in our case, including markedly distended alveoli, the
absence of bronchial cartilage, dispersed smooth muscle and minimal aspect of
inflammation, are the distinctive characteristics of CLE, and they correspond to previous
reports [1, 2, 5, 10]. The aplasia or dysplasia of cartilage around bronchi is main cause
of CLE. It would lead to bronchial collapse on expiration and obstruction on inspiration,
and eventually cause over-inflation of affected lobe [1, 10]. In the present case, an
increased number of pulmonary vessels in areas of peribronchial fibrosis were
demonstrated by \( \alpha \)-SMA immunostaining, indicating the formation of new blood vessels
or angiogenesis. A previous study has described that the angiogenesis is commonly found in pulmonary fibrosis. The possible mechanism of angiogenesis in pulmonary fibrosis is that it is stimulated by various angiogenic chemokines, such as vascular endothelial growth factor and fibroblast growth factor released by alveolar epithelial cells, macrophages and myofibroblasts in fibrotic areas [3].

It is well known that the radiographic features of CLE include hyperlucent lobes and mediastinal shift [1, 5, 6, 8, 10]. However, this case showed not only the general image findings of CLE, but also new findings, such as concurrent pneumothorax and pneumomediastinum. The pneumothorax by CLE could occur by two mechanisms. First, the rupture in visceral pleura of affected lobe could directly discharge air to pleural space [2]. Second, ruptured alveoli could indirectly raise pneumothorax. The air from the ruptured alveoli could cause pneumomediastinum by expansion through the perivascular or peribronchial connective tissues, and it is called as Macklin effect [11]. Mediastinum communicates with the subcutaneous tissue of neck region and with retroperitoneum through thoracic inlet and aortic hiatus, respectively. Therefore, air could migrate to the other regions, according to the severity of disease [4]. Excessively increased pressure by the free air in mediastinum could subsequent rupture the mediastinal pleura, pericardium or visceral pleura, and it could cause pneumothorax [9]. Although we could not identify the precise pathway of pneumothorax in this case, it was probably caused by one or both of above mentioned mechanisms, and at least, causative lesion was resolved by the surgical excision of affected lobe.

According to the previous reports, conservative treatment including intrathoracic tube drainage could not completely resolve CLE, and the most effective treatment was surgical excision of affected lung lobe [1, 5, 7]. In our case, the symptoms associated
with CLE were gradually alleviated and disappeared over 2 weeks after surgery. Then, Jackson-Pratt drain tube was removed. During 6 months of follow-up period, there were no recurrence and complication on radiographic examination.

In our best knowledge, this is the first case report of CLE showing pneumothorax, pneumomediastinum and subcutaneous emphysema in dog. The dog was diagnosed with a modern imaging device and advanced histopathological staining, and successfully corrected with surgical treatment.
REFERENCES


FIGURE LEGENDS

Fig. 1. The right lateral and ventrodorsal thoracic (a) and abdominal (b) radiographs of the dog showing pneumothorax (black arrowheads), pneumomediastinum (white arrowhead) and subcutaneous emphysema (arrows). Note the bulla-like lesion (asterisk).

Fig. 2. The computed tomography images of the dog at the level of cranial (a) and middle (b) of thorax. Note the trapped air around the trachea, esophagus and major vessels (arrows). The air-filled and bulla-like shaped lesions are seen (asterisk).

Fig. 3. The macroscopic findings of middle lung lobe show the extremely scanty parenchymal tissues.

Fig. 4. Severely dilated alveolar cavity and congestive blood vessels are observed. HE staining. Bar = 500 µm. A: alveolar space, B: bronchi, P: pleura, V: blood vessels.

Fig. 5. Proliferation of connective tissues especially in subpleura and perivascular areas are observed. Masson-trichrome staining. Bar = 500 µm. A: alveolar space, B: bronchi, P: pleura, V: blood vessels.

Fig. 6. Smooth muscles surrounding the bronchi occasionally dispersed (asterisks), and an increased number of pulmonary vessels are seen. Immunohistochemistry (α-SMA). Bar = 250 µm. A: alveolar space, B: bronchi.
Fig. 7. The epithelial cell layer of bronchi is irregularly arranged, and some cells invaginated into the underlying stroma (arrows). Immunohistochemistry (AE1/AE3). Bar = 250 µm. A: alveolar space, B: bronchi.
Fig. 1.
Fig. 2.

Fig. 3.
Figs. 4–7