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Intrapulmonary and Gastric Teratoma
: Report of Two Cases

The lung and stomach are very unusual sites for teratoma. The histologic findings of intrapulmonary and gastric teratomas are not different from those arising in usual sites, such as the ovary or testis. However, preoperative diagnosis is sometimes difficult to make partly because of unusual location. We report here two cases of teratoma, one intrapulmonary teratoma and the other gastric. The intrapulmonary teratoma in our study had an endobronchial tumor growth, which rules out mediastinal teratoma. Meanwhile gastric teratomas usually present as a submucosal tumor and most cases are reported in infancy and childhood. Gastric teratoma in this study occurred in a 27-year-old man. To the best of our knowledge, this case of intrapulmonary teratoma is the eighth and the gastric teratoma is the first to be reported in Korea.

Key Words: *Teratoma; Lung; Stomach*

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INTRODUCTION

Although teratomas may occur anywhere in the body, they are found, in order of decreasing frequency, in the ovary, testis, sacrococcygeal region and mediastinum (1). Intrapulmonary or gastric teratoma is very exceptional due to its location. A search of the literature revealed only 42 intrapulmonary teratomas and 99 gastric teratomas worldwide (1-8). Intrapulmonary teratomas often occur in the upper lobe with adhesion to the anterior mediastinum, so it is difficult to distinguish whether they are an intrapulmonary or anterior mediastinal mass (1). Trivedi et al. (9) stressed the importance of determining its exact anatomic location by endobronchial approach before labeling a teratoma as being intrapulmonary in origin. In cases of gastric teratoma, most reported patients are diagnosed in infancy or childhood (4-6, 10). We report two cases of teratomas: an intrapulmonary teratoma showing an endobronchial tumor and a gastric teratoma occurred in a 27-year-old man.

CASE REPORT

Case 1

A 49-year-old man visited our internal medicine clinic with a 10-day history of cough and hemoptysis. He had been well until that time. On physical examination, inspiratory rale was audible at left upper field. A bronchoscopy revealed luminal narrowing and obstruction of anterior segmental bronchus but failed to find intrabronchial lesion. A chest computed tomography (CT) scan showed an intrapulmonary mass of heterogeneous density which had no distinct border to mediastinal structures (Fig. 1). No calcification within the lesion was noted. The tumor was located near the segmental bronchus, however, it is not clear whether it is an endobronchial mass or not. A percutaneous needle aspiration was performed. Cytologic smear showed a large amount of amorphous anucleated squames and lipid material intermixed with foamy histiocytes, lymphoplasma cells and foreign body giant cells. Preoperative diagnosis was lung cancer. He underwent a segmentectomy of anterior segment of the left upper lobe. At surgery, A $5 \times 5 \times 4$ cm, firm nodular mass was palpable in the anterior segment of right upper lung. It was focally adherent to normal appearing thymus and easily separated from the thymus.

The resected specimen, measuring $8 \times 7 \times 4$ cm, showed irregular fibrous thickening of the visceral pleura and a subpleural whitish nodular lesion. On opening along

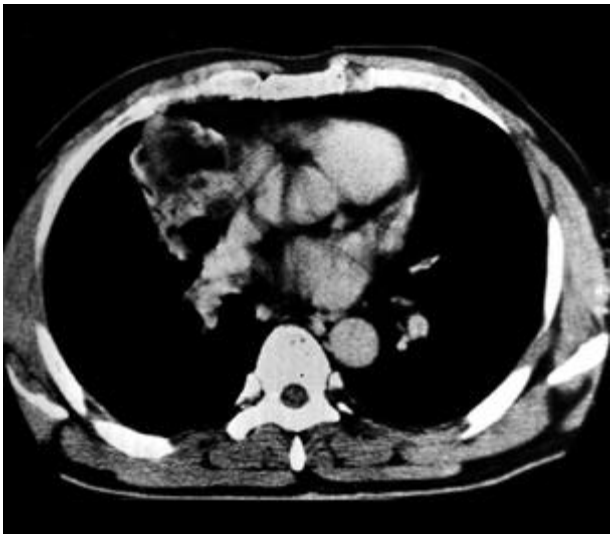

Fig. 1. A chest CT scan reveals an intrapulmonary mass of heterogeneous density having no distinct border from mediastinal structures.

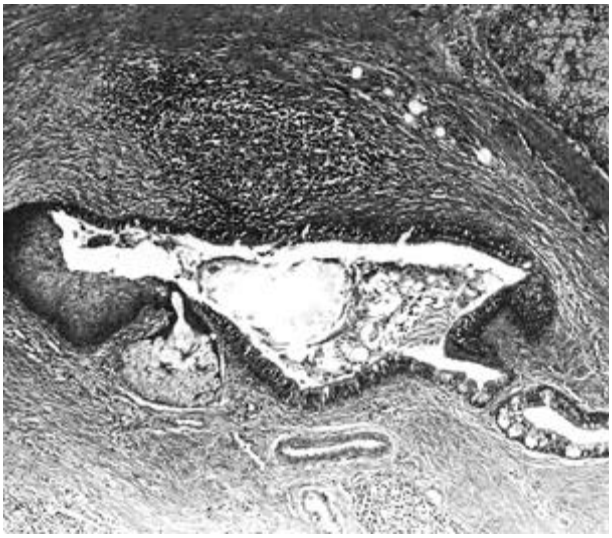

Fig. 3. A section of the teratoma showing keranizing stratified squamous epithelium with underlying sebaceous glands, ducts lined by columnar epithelium and fat (H&E, ×40).

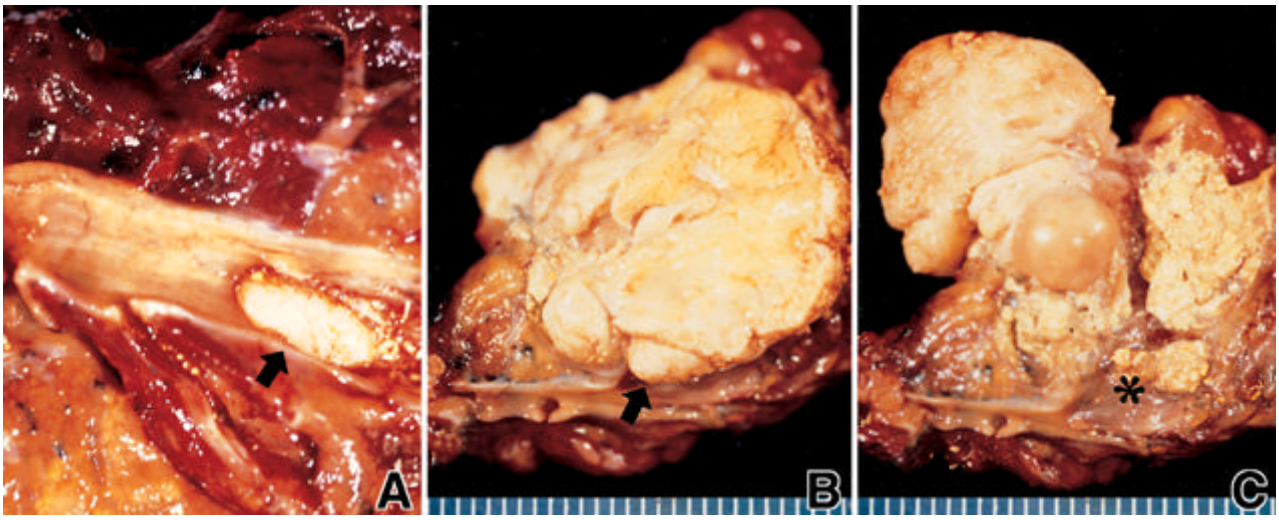

Fig. 2. On opening along the bronchial tree, a whitish 2×2×2.5 cm endobronchial nodule (arrow) is found 3 cm apart from the bronchial margin (A, B). The segmental bronchial mucosa (asterisk) is covered with whitish greasy material, and direct continuity of the tumor with the abutted bonchial mucosa is demonstrated (C).

the segmental bronchus, a 2×2.5×2 cm-sized endobronchial nodule was found (Fig. 2A). It was covered by yellowish-white greasy material and matted hair. After a careful dissection, a direct connection between the tumor and bronchial mucosa was demonstrated (Fig. 2B). The abutted bronchial mucosa showed erosion and hemorrhage, and was also covered with whitish greasy material (Fig. 2C). On serial cut sections, the mass occupying from the anterior segmental bronchus to the subpleural area was noted, and it was entirely surrounded by lung parenchyma. Cut surface revealed variegated appearance: the endobronchial protruding area was composed of usual skin and subcutaneous fat tissue, and the peripheral portions of the tumor showed mucus-filled cysts or yellowish solid appearance. The surrounding lung parenchyme was collapsed and firm in consistency. Histologically, the protruding intrabronchial nodular lesion was covered by keratinizing stratified squamous epithelium with hair follicles, hyperplastic sebaceous glands, apocrine and eccrine sweat glands (Fig. 3). Areas of mature cartilage, fat, foregut mucosa with smooth muscle coat, and fair amount of endo- and exocrine pancreatic tissue were also noted.

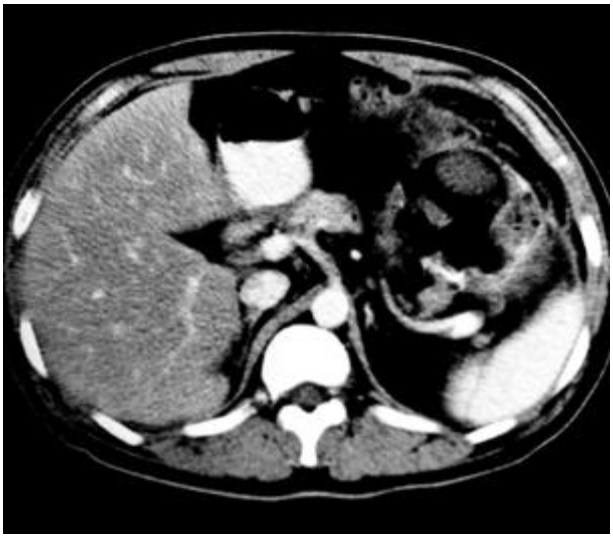

Fig. 4. An abdominal CT shows a large multilobulated gastric tumor of mixed density in the entire body along the greater curvature. Multifocal calcification is also noted within the tumor.

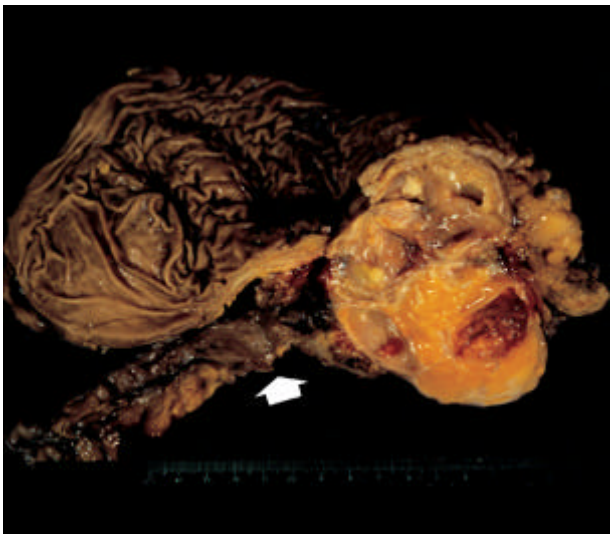

Fig. 5. Grossly, a large polypoid, multilobulated tumor, 9.5×7.5×5 cm is seen in upper body along the greater curvature of the resected stomach with focal adhesion to the transverse colonic wall (white arrow). Cut section of the tumor is largely yellowish solid, and some cystic areas filled with mucus material and multiple whitish calcific bony components are also noted.

The endocrine pancreatic tissue frequently showed hyperplastic proliferation exhibiting ribbon and festoon formation. Thymic tissue was noted here and there within the tumor. The adjacent bronchial mucosa showed diffusely hemorrhagic erosion with multifocal squamous metaplasia. The surrounding lung parenchyme showed alveolar septal fibrosis and diffuse infiltration of lymphoplasma cells, foamy macrophages, and multinucleated giant cells

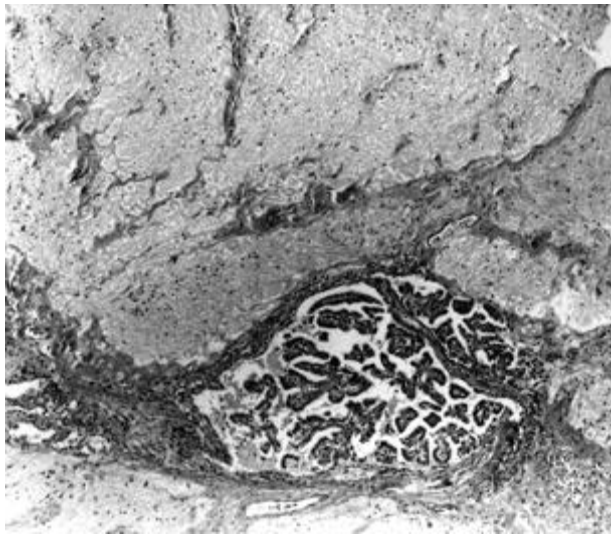

Fig. 6. Abundant neuroglial tissue and a focus of papillary structure mimicking choroid plexus is noted in the gastric teratoma (H&E, ×100).

as with patchy aggregates of keratin squames and endogenous lipid.

Case 2

A 27-year-old man visited our emergency room with a nine-day history of fever, chill and intermittent colicky pain in the left upper quadrant of abdomen and left flank area. He also complained nausea, vomiting, headache and epigastric discomfort. There was a 4-year history of episodic abdominal pain. On physical examination, there was tenderness in the epigastric area and left costovertebral angle, but no palpable mass. An upper gastrointestinal series was performed with no information because gastrografin did not pass through the esophagogastric junction. A computed tomography disclosed a large multilobulated gastric tumor of mixed density in the entire body along the greater curvature. The mass showed solid and cystic areas and calcification. A loop of transverse colon was adhered to the mass (Fig. 4). Gastroscopic examination revealed a huge polypoid mass with surface ulceration. Clinical impression was gastric cancer, so, he underwent extended total gastrectomy including entire stomach, a portion of distal pancreas, a spleen, and a segmentally resected transverse colon.

Grossly, a large polypoid, multilobulated tumor, 9.5×7.5×5 cm was noted in the body along the greater curvature of the resected stomach (Fig. 5). The serosal aspect of the mass showed adhesion to the transverse colonic wall. The mucosal surface of the tumor was ulcerative and a hard whitish bone component was exposed. Cut section of the tumor showed largely whitish to yel-

lowish solid area and some cystic portion filled with mucus material. Also there was considerable bone tissue. Histologically, the tumor was composed of irregularly arranged but well-differentiated tissue such as fat, smooth muscle, cartilage, lymphoreticular, bone with marrow component, foregut mucosa, lung parenchyma and neuroglia. A minute focus of papillary structures mimicking choroid plexus was also encountered (Fig. 6). No immature component was noted. The gastric wall was perforated with focal abscess formation between the gastric and colonic wall.

DISCUSSION

Teratomas may arise from the gonads or from extragonadal sites, like the sacrococcygeal and presacral area, the cervicopharyngeal area, the retroperitoneum, within the cranium, the thoracic or abdominal cavity. Intrapulmonary or gastric teratomas are extremely rare. Up to the present, 35 cases of intrapulmonary teratoma (1-3) and 99 cases of gastric teratoma have been reported outside our country (4-6). In Korean literature (7-8), 7 cases of intrapulmonary teratoma were noted but no gastric teratoma was found.

According to a theory proposed by Schlumberger in 1946 (11), intrathoracic teratomas, including mediastinal and intrapulmonary teratoma, have a common genesis and are thought to originate from the thymic tissue of the third pharyngeal pouch. While intrapulmonary teratoma may be the result of displacement or separation of the thymus during early embryogenesis. Pound and Willis further proposed that the primordial teratomatous focus in the potential mediastinum is caught up by the respiratory out-growth and located within the lung (12). Marchevsky (13) described that various intrapulmonary neoplasm such as malignant melanoma, thymoma, meningioma, glomus tumor, and germ cell neoplasms (choriocarcinoma and teratoma) also develop from ectopic tissue. Although their precise origin remains unclear, aberrant thymic tissue may be a candidate for its origin (2, 11, 12). On the other hand, there is a theory that primary pulmonary germ cell tumor represents unusual differentiation of somatic cell line (14, 15). Thymic tissue was frequently found, even in the central area of our tumor, so it was not a simply attached thymic tissue, moreover the present case was not mediastinal teratoma. It is not clear whether the thymic tissue present in our case is an element of teratoma or a aberrant thymic tissue. Presumably, however, the thymic tissue is significantly related with the pathogenesis of intrapulmonary teratoma.

As described above, intrapulmonary teratomas are exclusively rare, so it is necessary to exclude the possibility of mediastinal teratoma and metastasis from an extrapulmonary germ cell tumor. Also it is mandatory to prove intrapulmonary tumor location or a direct connection to the bronchus before making a diagnosis (9). Intrapulmonary teratomas predominantly occur in the upper lobes (65%), and many of them occur in the anterior segment, making one wonder about arising directly from included fragments of thymus (16). A direct connection with bronchial system has been found in less than 50% of the previously reported cases in the English literature (1), and in five out of seven cases reported in Korean literature (7, 8). Furthermore, obvious endobronchial protruding tumor component has so far been described in only four cases (1, 16). In our case, although the tumor was located in the anterior segment with focal adhesion to the thymus due to severe inflammation, it was mainly located in intrapulmonary area and completely surrounded by lung parenchyma. A connection to the bronchus and endobronchial tumor growth were evident. Moran et al. (17) reported seven cases of metastatic mature teratoma in lung from testicular teratocarcinoma and embyonal carcinoma that were indistinguishable with pathologic findings from primary intrapulmonary teratoma. So, when there is a single pulmonary nodule that microscopically is consistent with teratoma, they recommended that a detailed physical examination should be performed to rule out the presence of a small or occult extrapulmonary germ cell tumor. In the present case, systemic examination revealed no evidence of extrapulmonary germ cell tumor including testicular tumor, and normal serum level of α-fetoprotein.

There is an overall female predominance of teratomas, with approximately 68% of all teratomas occurring in female (18). Intrapulmonary teratomas showed no female predominance and except for a 10-month-old infant, most patients were diagnosed between the second and fifth decades of life (16).

Radiologically, intrapulmonary teratomas most often present as lobulated mass, and intratumoral calcification has been reported as the most important finding to make a diagnosis (1). A peripheral radiolucent area indicating air being trapped suggests a connection between the tumor and bronchial tree, helping to distinguish intrapulmonary teratoma from mediastinal one (1). However, it was not demonstrated in our case.

In 31 cases of intrapulmonary teratoma described in the literature (1), the clinical symptoms were rather nonspecific: chest pain (16 cases), hemoptysis (13 cases), and cough (12 cases). Expectoration of hair, so called trichoptysis, was seen in 4 of the 31 cases and was the most specific and interesting sign for this tumor (2). A Korean case with a 27-year history of trichoptysis was reported

(8). Hemoptysis was three times more common in patients with pathological or surgical evidence for bronchial connection, and tumors with large amounts of pancreatic tissue (1). It was thought that the erosion of the abutted bronchial mucosa might be due to an exocrine pancreatic secretion from the tumor (19). Support for this hypothesis was provided by the relatively common occurrence of exocrine pancreatic tissue in intrapulmonary teratomas (1, 16). In our case, hemoptysis was the main symptom and histologically, abundant pancreatic tissue and diffuse mucosal erosion were demonstrated.

The histogenesis of gastric teratoma has not been explained clearly yet. The germ cell theory that extragonadal teratoma originates from migrated totipotential germ cells has been generally accepted in gastric teratoma (4).

There is a striking male predominance in gastric teratoma with only 7 cases (7%) occurring in female (4). Additionally, gastric teratomas typically present in the first year of life as a palpable mass and approximately 94% of the reported patients were infants or neonates (4). To our knowledge, five adult cases of gastric teratoma has been reported with their ages being 83, 40, 37, 31 and 23, respectively (4, 10). Our case was a 27-year-old man who was considered to be the fifth oldest patient currently known.

Suggestive radiographic findings of gastric teratoma such as intratumoral calcification and mixed cystic and fatty components (4-6, 10) were typically noted in our case. Grossly, in our case, a large lobulated intraluminal polypoid mass with erosive mucosal surface was observed in the body along the greater curvature and was situated in the full thickness of the gastric wall. Therefore, it is clear that its origin was stomach. However, because of the rarity of this tumor and adhesion to the transverse colonic wall mimicking tumor infiltration, we had some difficulty in making preoperative diagnosis.

Gastric teratomas present as a palpable mass (75%) and/or abdominal distention (56%) in infancy or childhood (5), and epigastric discomfort and hematemesis or melena are main symptoms in adult (10). These symptoms were related to the large polypoid, exo/endogastric growth pattern and surface ulceration. Our case had recurrent upper abdominal and left flank pain, fever and chill for four years. These symptoms are thought to be related to degenerative and inflammatory change within the tumor.

Histological findings of our cases were consistent with the histologic criteria of teratoma and revealed mesodermal predominance, including bone and fat tissue. Also, abundant glial tissue and a focus of choroid plexus noted in our gastric teratoma were an interesting histologic feature.

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