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CASE REPORT

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An intrapulmonary teratoma associated with bronchiectasia containing various kinds of primordium: a case report and review of the literature

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Abstract An intrapulmonary teratoma (IPT), multiloculated and bronchiectatic, with two polyps inside a 23-year-old man is reported. The IPT, a very rare benign cystic lesion, was communicating with segmental bronchus and was removed by a segmental resection from the upper lobe of the left lung. The teratoma contained various kinds of primordial derivatives, such as mesoderm, ectoderm, and endoderm. Though 65 cases of IPT have been reported in the literature (1839–1996), in the present case there were over 15 germ derivatives, the largest number reported to date. The tumor contained thymic tissue, apart from mediastinum, which may be significant in relation to the pathogenesis of IPT. Clinical manifestations, age, and gender distributions and the kind of germ cell derivatives are discussed.

Key words Pulmonary teratoma · Bronchiectasia · Germ cell derivatives · Third pharyngeal pouch

Introduction

Intrapulmonary teratomas (IPT) are exceedingly rare compared with similar teratomas occurring in ovarium, testis, sacrum, retroperitoneum, oral floor, and mediastinum. This paper reports a benign multiloculated IPT in a 23-year-old man and a literature review from 1839 to 1996 [2, 6, 7, 8, 14].

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Case report

A 23-year-old man was admitted to Iwaki Kyouritsu General Hospital with left chest pain, fever, general malaise, cough and yellow sputum on 10 August 1996. A chest X-ray showed a large opacity in the left upper lobe (Fig. 1A), and computed tomography (CT) of the chest revealed an abnormal shadow with an air crescent lesion in the left upper lobe (S4+5+3) (Fig.1B). He had been admitted to another hospital with a diagnosis of pulmonary abscess

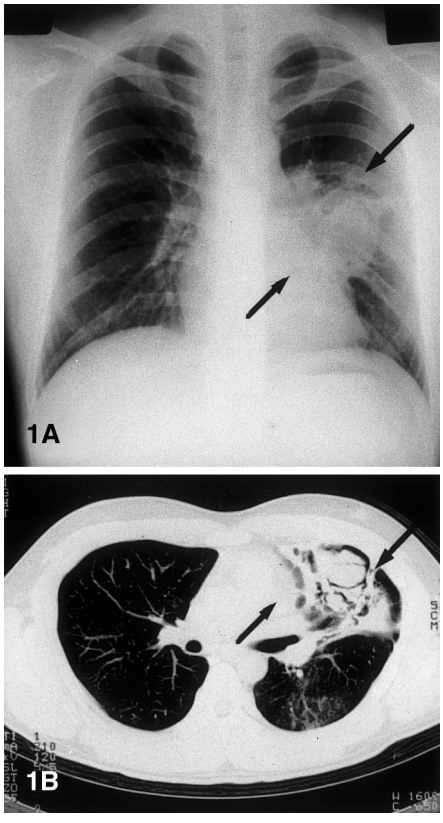

Fig. 1 Chest X-ray roentgenogram revealed an ill-defined round lesion in the upper lobe of the left lung (A). Computed tomography showed an air crescent lesion at S4+5 and S3 in the left lung (B)

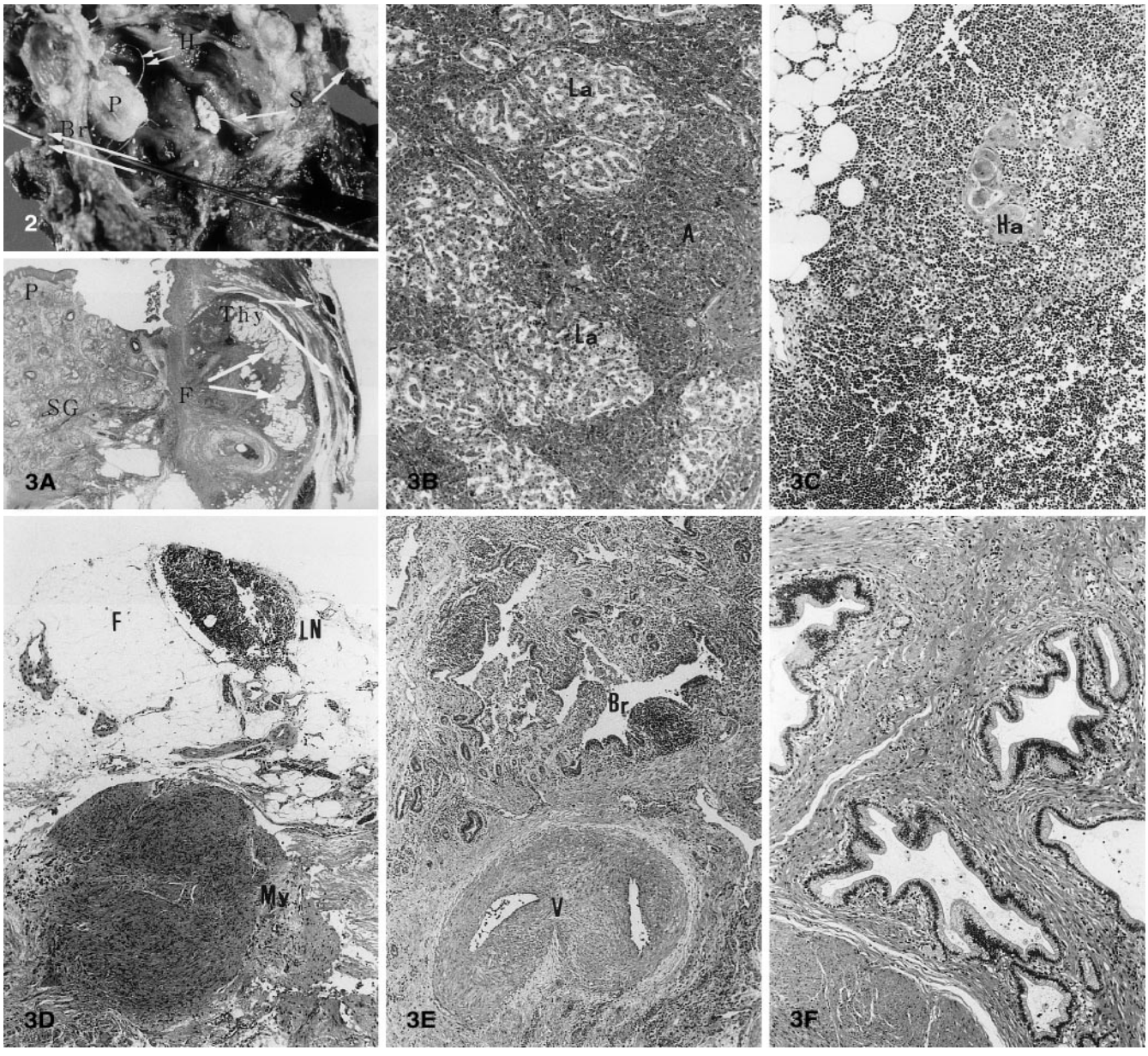

Fig. 2 Resected tumor of the upper lobe of the left lung. A multiloculated bronchiectatic lesion contained yellowish-white sebum (*S*) and hard polyps (*P*) with non-pigmented hairs (*H*) at the upper region of teratoma. The lesion connected to the main bronchus (*Br*)

Fig. 3 **A** The polyps (*P*) covered by squamous epithelium contained hair follicles, hyperplastic sweet glands (*SG*), adipose tissue (*F*), and thymus (*Thy*) bordered by thin fibrous tissue from the mediastinum. **B** The pancreatic tissue was composed of Langerhans' islands (*La*) and acinus (*A*). **C** Thymic tissue contained many Hassall's corpuscles (*Ha*) and lymphocytes. At the base of the ectatic lesion, leiomyoma nodules (*My*), lymph node (*LN*), fat cells (*F*) (**D**), thickened vessels (*V*), muscle, bronchus (*Br*) (**E**), high columnar epithelium (**F**), peripheral nerve, bone, and bone marrow were detected

from April to May 1994 and from August to September 1995. Since bronchoscopy revealed no abnormal findings at the end of 1995, he had stopped visiting the hospital.

After admission to our hospital, he was treated with antibiotics until 9 September 1996 for prolonged fever. He complained of discharge of hair-containing sebum during the hospitalization. The white blood cell count was 1800/mm^3, c-reactive protein was 18.65 mg/dl, and erythrocyte sedimentation rate was 58/1 h and 96/2 h. Cytological analysis of sputum revealed neither abnormal cells nor hair-containing sebum except with candida albicans. On 11 November, a left upper lobectomy was performed under the diagnosis of pulmonary abscess. A standard segmental resection was performed. The patient was discharged on 30 November with an uneventful postoperative recovery.

Pathology

A multiloculated bronchiectatic lesion, 6.0×5.0×3.0 cm, connected to the main bronchus was found in the anterior segment of the left upper lobe. It contained yellowish-

white fragile sebum and keratinous debris (5.5×3.0 cm) mingled with fine non-pigmented hairs. The lesion was lined by dark grayish bronchial mucosa, and two yellowish-white firm polyps, measuring 1.8×1.5×1.3 cm and 1.5×1.3×1.3 cm, respectively, were found in the upper region (Fig. 2). The bronchiectatic lesion was lined by ciliated columnar epithelium with squamous metaplasia. The polyps with non-pigmented long hairs were covered by squamous epithelium which contained inflammation, hair follicles and hyperplasia of apocrine and eccrine sweet glands (Fig. 3A).

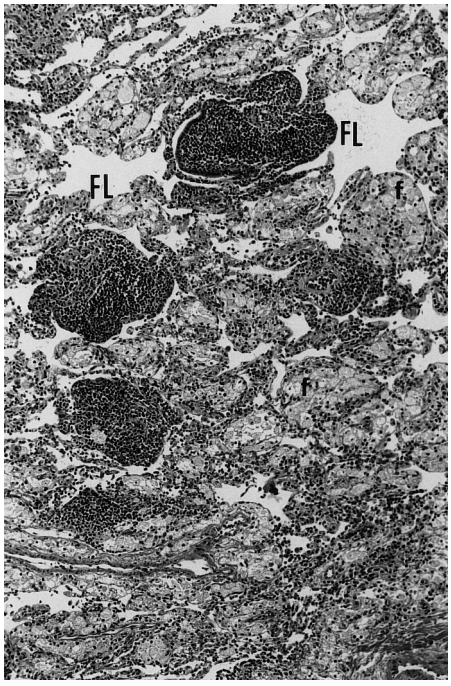

Fig. 4 Lipoid pneumonia with voluminous foamy cells (*f*) in the alveoli of atelectatic lung parenchyma which was surrounding the teratomatous lesion. Many lymph follicles were observed (*Fl*)

There was ectopic pancreatic tissue (1 cm in diameter), which was composed of many irregularly shaped Langerhans' islands and acinus (Fig. 3B), at the base of the polyps beneath the adipose tissue.

Several thymic tissues with Hassall's corpuscle, demarcated by thin fibrous tissue from mediastinum, were located in the distal region of the teratomatous lesion (Fig. 3C). In addition, at the periphery of the ectatic lesion, there were various kinds of teratomatous components, such as immature bronchi, simple columnar epithelium, thickened vessels, small leiomyoma nodules, peripheral nerve, lymph node, adipose tissue, muscle, bone, and bone marrow (Fig. 3D–F). The neighboring lung tissue with tiny yellow granules microscopically coincided with lipoid pneumonia including numerous foamy cells in the alveoli (Fig. 4). There were numerous small lymph follicles in the interstitium of the lung. Neither immature nerve tissues nor brain tissues were identified. A pathological diagnosis of benign IPT was made.

Discussion

This paper describes a 23-year-old man with histologically detectable IPT with over 15 derivatives, the largest number reported to date. IPT is an extremely rare tumor compared with mediastinal teratoma. Only 65 cases of IPT, including 35 in Japan, have been reported in the literature (1839–1996), and the diagnosis was usually made based only on the excised specimen [2, 3, 6, 7, 8, 14]. The age and gender distributions for IPT are shown in Fig. 5. The teratoma occurs in adults as well as in children [3], with the age of the subjects ranging from 10 months to 68 years. Most patients are diagnosed in the first or second decade of their lives (45%). It had been described for the occurrence of IPT that females are more commonly affected than males, but IPT is equally prevalent in women (34 cases) and men (31 cases) [2, 3, 6, 7, 8, 14]. Hemoptysis and trichoptysis, indicating a

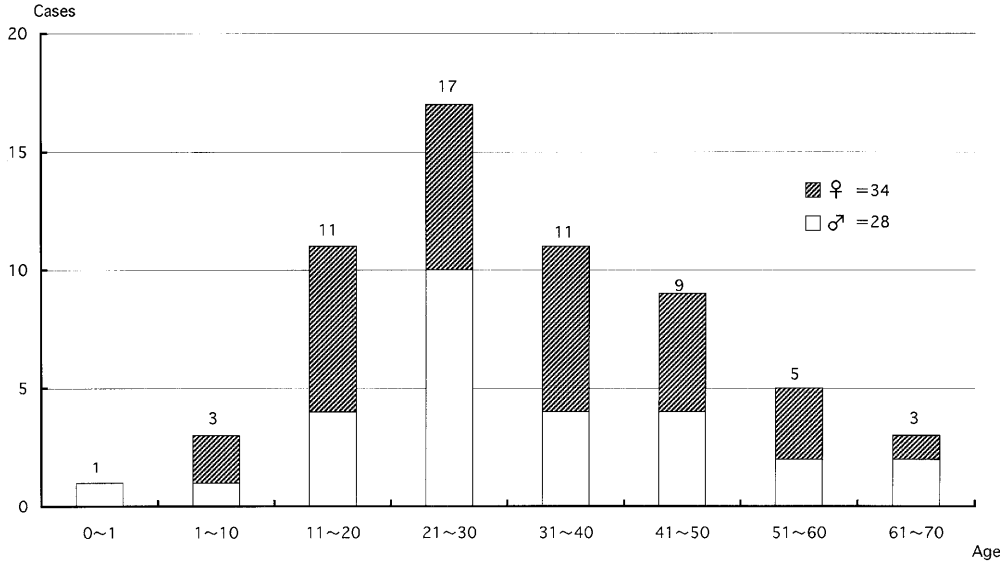

Fig. 5 Age and gender distribution in 62 cases of intrapulmonary teratoma (1839–1996)

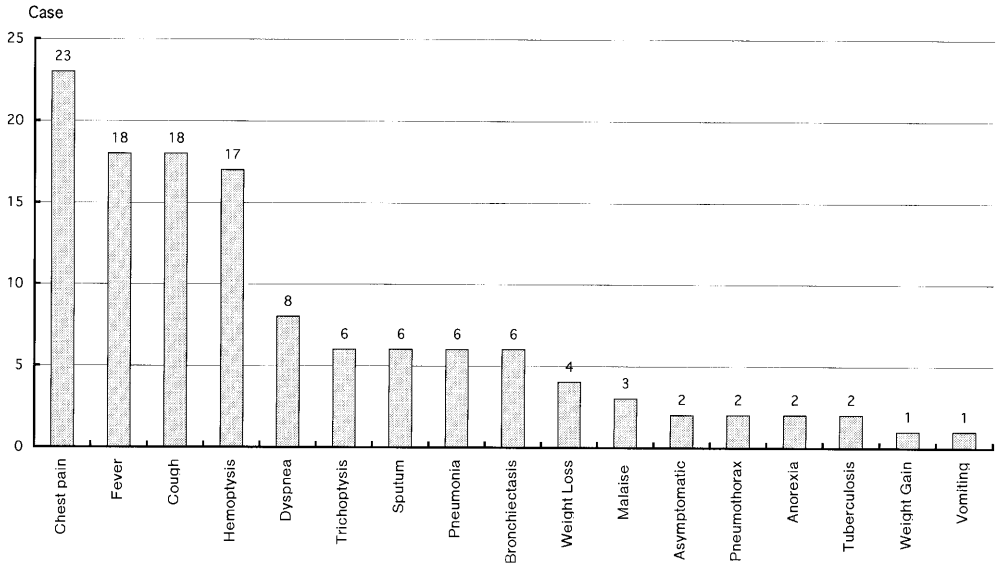

Fig. 6 Symptoms in 46 cases of intrapulmonary teratoma (1839–1996)

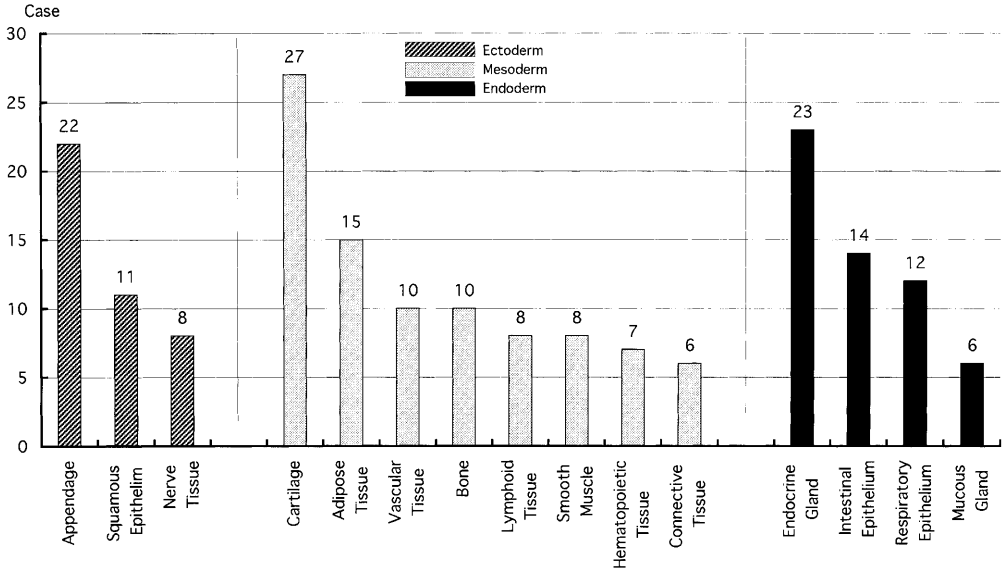

Fig. 7 Germ cell derivatives in 27 cases of intrapulmonary teratoma (1839–1996)

connection to the bronchial system, are not uncommon in patients with endobronchial tumor or bronchiectasis [2, 4, 5]. The cystic tumors in IPT, the walls of which are partly lined by squamous epithelium, contain characteristic inspissated brownish hair containing sebaceous materials, like those observed in the present case. Other recorded symptoms, including chest pain, fever, cough, dyspnea, and sputum, are probably due to bronchiectasia and lipoid pneumonia surrounding the teratomatous lesion. The most commonly reported symptoms of IPT are shown in Fig. 6 [2, 3, 6, 7, 8, 14, 15]. Clinical manifestations such pleuritic types of chest pain appeared to be nonspecific and the unusual nature of the lesion made it difficult to arrive at a preoperative diagnosis [7]. The occurrence of congenital cardiovascular anomalies in IPT patients has also been reported [1].

The chest X-ray of a patient with IPT usually reveals a lobulated opacity within the upper lobe mass in the lung, and calcification within the lesion [15], cavitation [2], or peripheral translucent areas might be a helpful diagnostic feature [1, 2, 4]. The patients showed either cystic or solid tumors which grew slowly [13]. A peripheral radiolucent area indicating air within the cavity reflected either communicating of the teratoma to the bronchial system or masses within a bronchus. This helped to distinguish IPT radiologically from mediastinal teratoma [7]. The presence of the crescent-shaped translucent area within the mass raised the possibility that the lesion might be a fungal mass, hydatid cyst, pericyst, endocyst, blood clot, or lung abscess [2]. However, theoretically, the neoplasm might be differentiated [2]. It had been reported that the great majority of cases of IPT are found in the upper lobes and usually on the left side [7, 8, 13], but a search of the literature revealed no predilection of the right lobes (32 cases) or the left (32 cases), but predilection of the upper lobes of both lungs (left upper 28

cases; left lower 4 cases; right upper 21 cases; right middle 7 cases; right lower 5 cases) [2, 3, 6, 7, 8, 14].

Since the late 1950s, an increasing number of studies have examined the histological and clinicopathological features of IPT. IPT is characterized by a variety of mesodermal, ectodermal, and endodermal derivatives that are indicative of the potential of the parent cells to differentiate along the line of the three germ layers; skin and appendages, and cartilage, fat, muscle, vascular or hematopoietic tissues, together with intestinal and respiratory epitheliums or pancreatic tissues (Fig. 7) [6, 7, 8, 9, 10, 14]. Each IPT in general contained less than 10 derivatives, whereas the present case contained over 15 derivatives.

Approximately one-third of the tumors in published cases demonstrated malignancies (18 of 65 cases). Apparently, this designation has been used loosely based on the presence of immature tissues within the teratoma rather than on evidence of infiltration and metastasis. This practice may account for the unexpectedly good results obtained by surgical treatment of some purportedly malignant lesions [1, 2, 6, 7, 8, 14]. Malignant teratoma has occurred at all ages from a 10-month-old infant to a 68-year-old man. Malignant teratoma presented in more women (12 cases) than men (5 cases) – with one case in which the gender was unknown – and was more liable to present at the right lung (12 cases) than the left (6 cases). In addition, the size of the teratoma was not predictive of malignancy as it ranged from 1×1.5×30 cm to 16×14×30 cm [8]. Though the origin of IPT remained as obscure as that of mediastinal teratoma [12], it was considered that the mediastinal teratoma and IPT developed in relation to the thymus as derivatives of the third pharyngeal pouch. In the third to sixth gestational weeks, thymic tissue and the lungs are derived from the third pharyngeal pouch and the ventral side of the foregut, respectively. It was further proposed that IPT occurs by displacement or early separation of thymic tissue with the migration along the course of the developing lung bud [1, 2, 11]. In the present case, it was thought that various kinds of derivatives expanded in to the bronchus instead of the lung parenchyma without direct connection to the anterior mediastinum.

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