CASE REPORT

Infected tracheal diverticulum mimicking an aggressive mediastinal lesion on FDG PET/CT: an interesting case with review of the literature

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ABSTRACT. The differential diagnosis for intense hypermetabolic mediastinal lesions on positron emission tomography (PET) could benefit from the combined morphological and metabolic information present in a fluorodeoxyglucose (FDG) PET/CT study. We report a case of an infected tracheal diverticulum mimicking an FDG-avid malignancy in a patient with a history of chronic lymphoproliferative disease. We review the literature for a systematic approach in the differential diagnosis of cystic mediastinal lesions. The embryological development of the normal tracheobronchial tree is reviewed, followed by a presentation of various congenital and acquired mediastinal lesions. The characteristic CT findings are described for each lesion and the avidity for FDG on PET is mentioned when references are available. This case emphasises that complicated benign processes should be considered in the differential diagnosis of an FDG-avid mediastinal lesion, even in subgroups of patients with significant risk factors for malignancy.

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The value of fluorodeoxyglucose (FDG) positron emission tomography (PET) in the initial staging, assessment of recurrence, response to therapy and follow-up of various oncological disorders is well established. FDG PET has widespread use in the diagnosis and staging of non-small cell lung cancer, especially in nodal staging and distant metastatic disease assessment [1–3]. PET/CT imaging has evolved to become the modality of choice for staging lymphoma, assessing therapeutic response and establishing patient prognosis [4, 5]. The literature is continuously increasing on the role of FDG PET/CT in the assessment of known lesions. A differential diagnosis approach for an FDG-avid lesion in a specific anatomical region without knowledge of histology is not described as clearly.

We report an interesting case of an infected tracheal diverticulum mimicking an FDG-avid malignancy. We review the literature for a systematic approach in the differential diagnosis of mediastinal lesions with cystic pattern that can present FDG uptake on PET studies.

Case report

A 74-year-old female presented to the emergency department following a recent fall. The patient had a history of chronic lymphoproliferative disease (Waldenström's macroglobulinaemia) and a previous hip fracture. Although

the initial radiographs did not show any new fractures, a chest radiograph revealed a large mediastinal mass making the trachea deviate, which prompted further investigation.

A chest CT (Figure 1) revealed a new right paratracheal mediastinal mass, which was not present on a chest CT performed 5 months earlier. This mass displaced the trachea and the oesophagus to the left, had a necrotic centre and was associated with small lymph nodes in the aortopulmonary window. The largest node was a 16 mm infracarinal node, there were unchanged 8 mm and 10 mm nodules in the spleen, a stable 2 cm node in the gastrohepatic ligament region and smaller stable retroperitoneal nodes.

An FDG PET/CT (Figure 2) was performed for staging purposes to identify possible lymphoma transformation of known chronic lymphoproliferative disease. It revealed intense FDG activity at the periphery of a large mediastinal mass lesion that had a relatively hypometabolic centre. The metabolic lesion measured more than 5 cm in diameter and had a maximal standardised uptake value (SUV) of 9.2. It extended from the sternal notch following the right paratracheal groove inferiorly to the antecarinal area and displaced the trachea and the oesophagus to the left. The known aortopulmonary window and the infracarinal nodes were not associated with abnormal FDG uptake. There was uniform uptake in the spleen parenchyma and the known retroperitoneal nodes showed no abnormal activity. The differential diagnosis was a solitary doughnut-shaped hypermetabolic mediastinal lesion, which included active neoplasm and infectious process.

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Figure 1. A chest CT performed at presentation showing a large right paratracheal mediastinal lesion displacing the trachea and the oesophagus to the left. Note the relatively hypodense necrotic centre.

The infectious process differential was raised owing to the significant discrepancy in activity between the main lesion and the local and retroperitoneal nodes. At this point, a histological correlation was recommended.

An exhaustive review of the patient's chart revealed that on a chest CT performed a year earlier, a right posterior tracheal diverticulum had been mentioned incidentally (Figure 3). The tracheal diverticulum was first mentioned in December 2006, measured 15 mm in diameter and demonstrated typical air content with septation.

As a result of mediastinoscopy and subsequent microbiological evaluation, the patient was diagnosed with an infected tracheal diverticulum. The abscess was drained and the patient hospitalised for intravenous antibiotic therapy. On a follow-up chest CT, a residual soft-tissue density remained visible in the upper mediastinum, but there was no evidence of aggressive behaviour in the interval. 6 months of clinical and radiographical follow-up confirm the benign, although important, nature of this mediastinal lesion.

Discussion

Embryology

Ghaye et al [6] summarised the embryological development of the normal tracheobronchial tree. It usually starts at 24–26 days as a median bulge of the ventral wall of the pharynx that develops at the caudal end of the laryngotracheal groove. At 26–28 days, this bulge gives rise to right and left lung buds. As the lung buds elongate, the trachea is separated from the oesophagus

Figure 2. Fluorodeoxyglucose positron emission tomography/CT selected images showing intense activity at the periphery of a large mediastinal mass with a relatively hypometabolic centre. The lesion had a maximal standardised uptake value of 9.2. It extended from the sternal notch following the right paratracheal groove inferiorly to the antecarinal area and displaced the trachea and the oesophagus to the left.

Figure 3. A chest CT performed a few months earlier than that in Figure 1, demonstrating the uninfected tracheal diverticulum. Note the air density within the rounded lesion as well as the communication with the trachea, which is typical for a tracheal diverticulum.

by lateral ingrowths of the mesoderm. At 28–30 days, the lung buds elongate into primary bronchi. At 30–32 days, the 5 lobar bronchi appear as outgrowths of the primary bronchi. The lobar bronchi elongate at 32–34 days and then rapidly branch to form all segmental bronchi by 36 days. Over the same period, the vascular supply to this tissue shifts from branches of the splenic plexus to definite pulmonary arteries as the lung bud plexus fuses with the sixth branchial arches [6, 7].

Berrocal et al [8] reviewed embryology, imaging key findings and pathology of various congenital anomalies of the tracheobronchial tree. They described the following tracheal anomalies: tracheomalacia, tracheal stenosis, tracheal bronchus, bronchial atresia and bronchogenic cyst. These are rare congenital malformations that are more frequently encountered in children.

Epidemiology

Tracheal diverticulum is relatively rare and accounts for about 2% of cases of air-filled cystic lesions in the neck. Goo et al [9] reported perhaps the largest series, with 64 proven cases. In previous literature, tracheal diverticulum has been described as paratracheal diverticulum, air cyst, bronchogenic cyst, tracheocele and lymphoepithelial cyst [10].

Histopathology

A diverticulum is lined by ciliated columnar epithelium and has a communication with the tracheal lumen. Two types of tracheal diverticula have been reported: acquired and congenital [11]. The acquired form is associated with chronic bronchopulmonary disease in the adult population and is thought to represent mucosal

herniation secondary to increased intraluminal pressure at a weak point of the tracheal wall. The congenital form occurs 4–5 cm below the true vocal folds or a few centimetres above the carina on the right side of the trachea. It usually contains smooth muscle and cartilage. During a CT scan, a communication with the tracheal lumen should be identified and the presence of cartilaginous rings within the wall of the diverticulum may help to determine if the lesion is congenital or acquired [10–12].

Bronchogenic cysts are congenital lesions which are thought to originate from the primitive ventral foregut and may be mediastinal, intrapulmonary or less frequently in the lower cervical area. About two-thirds of bronchogenic cysts are located in the mediastinum. They represent 40–50% of all congenital mediastinal cysts [13]. These cysts contain mucoid material and are lined by ciliated columnar or cuboidal epithelium. Their walls contain smooth muscle and often cartilage. If mediastinal, they may be located paratracheal (usually right-sided), carinal or hilar. They are most frequently located in the carina. They can also be found in the posterior and anterior mediastinum. If located in the anterior mediastinum, it may be necessary to distinguish them from cystic teratoma, thymic cysts or cysts derived from ectopic thyroid glands [8, 14, 15].

Bronchogenic cysts do not initially communicate with the tracheobronchial tree. Although most bronchogenic cysts in children are found incidentally, more than half are symptomatic. The symptoms vary with the size and the position of the cyst and may result from compression of the trachea or bronchi. This leads to coughing, wheezing, stridor, dyspnoea, cyanotic spells and pneumonia [8]. Infection occurs in 20% of cases with intraparenchymal cysts, usually secondary to communication with the tracheobronchial tree [16].

FDG-PET characteristics

Performing a Medline search for tracheal diverticulum and FDG did not yield any results. A search for bronchogenic cyst and FDG yielded a single reference. Sala and Coulden [17] report a case of bronchogenic cyst incidentally found on FDG PET/CT performed for pulmonary nodule evaluation. It demonstrated a photon-deficient area in the subcarinal region that corresponded to a bronchogenic cyst on CT images. Although the SUV measurement of this anomaly was not given, only low-grade uptake could be appreciated at the periphery of the doughnut-shaped lesion that demonstrated a cold centre.

CT characteristics

The CT features of benign mediastinal cysts are a smooth, oval or tubular mass with a well-defined thin wall that usually enhances after intravascular administration of contrast material. The cysts show homogeneous attenuation, usually in the range of water attenuation (0–20 HU), with no enhancement of contents and no infiltration of adjacent mediastinal structures [7]. Oesophageal duplication cysts may share many of the features of bronchogenic cysts. To distinguish an oesophageal

duplication cyst, its relation to the oesophagus should be assessed. Half of thoracic duplication cysts contain ectopic gastric mucosa. $^{99}\text{Tc}^{\text{m}}$ -pertechnetate may be of great value in the characterisation of such lesions [18], because the normal physiological distribution of pertechnetate is intense in gastric mucosa.

The walls of pericardial cysts are composed of connective tissue, a single layer of mesothelial cells and they usually contain clear fluid. Pericardial cysts are invariably connected to the pericardium and the majority of them occur in the right anterior cardiophrenic angle [19]. However, they can be seen as high as the pericardial recesses at the level of the proximal aorta [20]. The cysts are asymptomatic in most patients.

Most meningoceles are detected in adults [7]. During a CT scan they appear as well-defined, homogeneous low-attenuation paravertebral masses. Other possible findings include enlargement of intervertebral foramina and associated vertebral and rib anomalies or scoliosis [21].

Thymic cysts are uncommon and they can be congenital or acquired, unilocular or multilocular [22]. Simple congenital thymic cysts usually appear on a CT scan as well-defined water-attenuation masses with imperceptible walls [7]. Multilocular thymic cysts may appear as well-defined, heterogeneous cystic masses with clearly seen walls [23]. Some thymic cysts may demonstrate increased CT attenuation if a haemorrhage or an infection occurs and may be misdiagnosed as solid masses. Curvilinear calcification of the cyst wall occurs in a minority of cases [22].

Mature cystic teratomas (dermoid cysts) are cystic tumours derived from at least two of the three germ cell layers (ectoderm, mesoderm and endoderm). Cyst formation is typical and they are usually lined by tall mucus-secreting epithelial cells. The cysts are filled with sebaceous material and may contain hair. Mature cystic teratomas are the most common germ cell neoplasm. They occur more frequently in young adults. Most are asymptomatic and as a result are discovered incidentally. However, large tumours may cause chest pain, dyspnoea or other symptoms of compression [7]. The majority of dermoid cysts are in the anterior mediastinum, with only 3–8% occurring in the posterior mediastinum [24]. A fat fluid level within the mass is a finding that is highly specific to these cysts [7].

Lymphangiomas are rare (0.7–4.5% of all mediastinal tumours) benign congenital malformations consisting of focal proliferations of well-differentiated lymphatic tissue. They are most common in the neck and axilla, and about 10% extend into the mediastinum [25]. CT scans usually show a smooth lobulated mass which may mould to or envelope, rather than displace, the adjacent mediastinal structures [7]. Lymphangiomas usually have homogeneous low attenuation similar to water on a CT scan, but can also have a combination of fluid, solid tissue and fat. Calcification in lymphangiomas is rare [7].

The differential diagnosis must include tumours' cystic degeneration either as a spontaneous process or following therapy, thymomas, Hodgkin's disease, germ cell tumours, mediastinal carcinomas, lymph node metastases, nerve root tumours (schwannomas and neurofibromas) [7].

Finally, mediastinal abscesses are usually iatrogenic in nature following surgery, mediastinoscopy, an

oesophageal perforation or secondary to the spread of infection from an adjacent structure. The abscesses may appear as low-attenuation masses on a CT scan owing to fluid content. Air bubbles, contiguity or communication with an empyema or subphrenic abscess and clinical features usually permit identification of the infectious processes [21].

Conclusion

This article reports an infrequent complication of a rare tracheal malformation. The combination of a good response to antibiotic therapy, the radiographic findings and the favourable clinical evolution confirms the diagnosis of an infected tracheal diverticulum that was mimicking an intense FDG-avid mediastinal malignancy on the initial FDG PET/CT study. This case emphasises that simple or complicated benign processes should always be considered in the differential diagnosis of an FDG-avid mediastinal lesion on FDG PET/CT. This diagnosis should be considered even in subgroups of patients with significant risk factors and a medical history of malignancy.

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