A tracheal lesion simulating an aneurysmal bone cyst

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A 37-year-old man presented to the emergency department with the chief complaints of persistent cough and shortness of breath for approximately 2 months. At least two episodes of mild hemoptysis prior to presentation were reported. His past medical history was significant only for chronic low back pain secondary to multiple herniated discs. The chest radiograph at the time of presentation showed a subtle nodular opacity projecting over the region of the distal trachea (*Figure 1*). The lungs were clear. Subsequent chest computed tomography (CT) demonstrated a soft tissue mass measuring up to 1.7 cm in the distal trachea at the level of the aortic arch (*Figure 2*). A small central focus of calcification was present in the lesion. The mass appeared to arise from the right anterolateral tracheal wall and invade the adjacent mediastinum. The differential diagnosis included primary malignancies of the trachea such as squamous cell carcinoma, adenoid cystic carcinoma, mucoepidermoid carcinoma, and carcinoid. Benign tracheal tumors such as papilloma, hamartoma, leiomyoma, and amyloidoma were also considered.

Rigid bronchoscopy demonstrated that the mass was located 1.5 cm cephalad to the carina and occupied approximately 80% of the tracheal lumen. Following biopsy, pathology initially reported a mesenchymal spindle cell tumor with bone and numerous osteoclast-like giant cells. The tumor was subsequently resected and tracheal reconstruction was performed. Final pathology demonstrated the previous findings, as well as blood-filled cystic spaces surrounding cartilaginous elements (*Figure 3*). These findings were consistent with an aneurysmal bone cyst arising from and extending through the cartilaginous rings of the trachea. No additional lesions were identified within the tumor specimen or the resected tracheal segment.

FINAL DIAGNOSIS: Aneurysmal bone cyst of the trachea.

Figure 1. (a) Posteroanterior and (b) lateral chest radiographs demonstrate a nodular opacity projecting over the distal trachea (white arrows).

Figure 2. Axial CT images on (a) lung and (b) soft tissue windows at the level of the aortic arch show a mass within the distal trachea (black arrow). Central calcification is present. There is extension through the right anterolateral aspect of the trachea and into the mediastinum (white arrow).

DISCUSSION

Aneurysmal bone cyst (ABC) is a benign tumor of the bone that most commonly involves the long bones, spine, and pelvis

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Figure 3. Histopathologic section demonstrates normal cartilage surrounded by blood-filled cystic spaces.

of adolescents and young adults. Four forms of ABC have been described in the literature. Primary ABCs are tumors in which no coexisting or precursor lesions can be identified. Secondary ABCs are tumors in which coexisting or precursor lesions can be identified. ABCs may be secondary to trauma and occur at the site of prior osseous injury. Finally, a solid variant of ABCs has been described (1). Identifiable lesions are associated with ABCs about half the time, the most common of which include giant cell tumor, osteoblastoma, chondroblastoma, and osteosarcoma (2). Coexisting lesions such as fibrous dysplasia, nonossifying fibroma, and chondromyxoid fibroma have been described.

Histologically, ABCs are composed of multiple blood-filled spaces separated by fibroblasts and myofibroblasts, osteoclast-like giant cells, osteoid, and bone. ABCs were primarily treated with curettage in the past. Autograft implantations and intercalary allografts were utilized in large lesions or tumors that compromised bone integrity. However, the current approach employs biopsy, curettage, and implantation of either allograft bone chips or polymethylmethacrylate. Recurrence rates range from 17% to 26% but have decreased over the past 20 years (3).

Extrasosseous aneurysmal bone cysts are rare. Case reports have described primary ABCs arising from the larynx, cricoid cartilage, thyroid cartilage, and soft tissues (3). However, to the best of our knowledge, this is the first reported case of tracheal ABC. The radiographic features of this entity are nonspecific, and the differential diagnosis primarily includes malignant and benign tumors of the trachea. While not seen in this case, postobstructive imaging findings such as atelectasis, pneumonitis, and bronchiectasis may be seen with both malignant and benign tumors.

Malignant neoplasms of the trachea comprise <1% of thoracic malignancies. Clinical symptoms at the time of presentation are typically nonspecific and include dyspnea, wheezing, stridor, and hemoptysis. The most common primary tracheal malignancies are squamous cell carcinoma and adenoid cystic carcinoma (4). Mucoepidermoid carcinoma and carcinoid are

less common and typically involve the main, lobar, and segmental bronchi. These tumors may manifest as intraluminal polypoid or sessile masses, eccentric narrowing of the airway, or circumferential wall thickening on CT. Carcinoid tumors are highly vascular and may demonstrate intense enhancement after the administration of intravenous contrast material. On 18-fluoro-2-deoxyglucose (FDG) positron emission tomography/CT, intense FDG uptake is typically seen in squamous cell carcinomas and high-grade adenoid cystic and mucoepidermoid carcinomas. FDG uptake within carcinoid tumors is more variable and, when present, is typically less intense than in other primary tracheal malignancies (5).

Papillomas are the most common benign neoplasm of the tracheobronchial tree and occur in multiple and solitary forms. The multiple form is more common in children and young adults and is known as juvenile laryngotracheal papillomatosis. The solitary form is more common in adults and appears as a focal polypoid nodule or mass on CT. Hamartomas of the tracheobronchial tree are composed of tissues normally present within the airways. Although less common than intrapulmonary hamartoma, these hamartomas are more likely to produce symptoms of airway obstruction. Intraluminal hamartomas typically appear as soft tissue masses on CT. The presence of fat or calcification is more specific for hamartoma. Leiomyomas are rare tumors that may arise from the smooth muscle of the tracheal wall and manifest as intraluminal soft tissue masses on CT. Cystic degeneration may be present secondary to poor vascularization of the tumor. Amyloidosis is a family of diseases that is characterized by extracellular deposition of amyloid. The most common form of pulmonary amyloidosis is diffuse tracheobronchial involvement, which may appear as multiple nodules or masses, diffuse tracheal narrowing, or circumferential thickening of the airways. Amyloidomas are focal collections of amyloid that present as solitary nodules or masses that may contain calcification (6).

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