The Journal of Laryngology & Otology, E4, 1 of 3.

doi:10.1017/S0022215107001326

Online Only Clinical Record

Complete recovery of an isolated left vocal fold palsy associated with a benign mediastinal thymic cyst

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Abstract

Objectives: To present a case of benign mediastinal thymic cyst, and to review the published information on these cysts, including their incidence, presentation, diagnosis and management.

Methods: We report the case of a 55-year-old man who presented with a unilateral vocal fold palsy subsequently found to be due to the presence of a benign mediastinal thymic cyst. A literature search was undertaken to identify the incidence, key features and management of this rare condition.

Results: Benign mediastinal thymic cysts are a rare cause of mediastinal masses. Usually diagnosed incidentally, their management is usually surgical. Vocal fold palsy in isolation has not previously been reported in association with mediastinal thymic cysts.

Conclusion: This report describes what we believe to be the first published case of a completely reversible vocal fold palsy presenting in association with a rare benign mediastinal thymic cyst.

Key words: Thymus; Vocal Cord Paralysis; Mediastinum

Introduction

Unilateral vocal fold palsy is a common presentation to otolaryngology clinics. Such a presentation is a symptom rather than a diagnosis, and necessitates a search for an underlying, treatable cause. The cause may be found anywhere in the distribution of the vagus nerve, and varies from invasive malignant disease to benign cystic lesions. Symptoms of vocal fold palsy vary between patients, with some patients having no symptoms and others having voice abnormalities (classically, a weak, breathy, husky voice) and problems with aspiration. Complete recovery of vocal fold function is rare, with the best outcome being an immobile or poorly mobile vocal fold with a satisfactory voice and airway.[1]

We report the case of a 55-year-old man who presented with a unilateral vocal fold palsy due to a rare benign mediastinal thymic cyst. His vocal fold function recovered completely post-operatively. We review the pathology, incidence, presentation, investigation and management of this rare condition.

Case report

A 55-year-old Caucasian man was referred by his general practitioner to the ENT clinic with a six-month history of intermittent 'rasping', hoarseness and dysphagia. He had no past medical history, was taking no medication and was a lifelong non-smoker.

Clinical examination of the patient's neck, ears, nose and oropharynx was normal. Flexible nasendoscopy demonstrated an isolated paralysis of the left vocal fold. There was no evidence of any other laryngeal pathology.

A computerised tomography (CT) scan from skull base to mediastinum was performed. The CT scan at the level of the larynx was entirely normal, but the post-contrast CT scan of the mediastinum demonstrated a bi-concave, hypodense, clearly defined soft tissue mass in the anterior mediastinum, together with fibrotic change within the lung parenchyma (see Figure 1).

The patient was referred urgently to the cardio-thoracic unit, where he underwent surgery to remove the mediastinal mass. He had an uneventful post-operative period and was discharged home five days after surgery.

Macroscopic examination of the mass revealed the presence of a unilocular, thin-walled, cystic lesion filled with a yellowish, cloudy fluid. Microscopic examination revealed squamous non-keratinised epithelium partially replaced by an inflammatory infiltrate containing cholesterol clefts. The presence of thymic tissue and Hassell's corpuscles in the cyst wall was pathognomonic of a benign thymic cyst.

Two months later, complete recovery of the left vocal fold palsy was demonstrated by flexible nasendoscopy in the ENT clinic.

Discussion

Unilateral vocal fold palsy is a common presentation in otolaryngology. This presentation, however, is not a diagnosis but a symptom of other pathology, both benign and malignant. In older patients, a search for malignancy is imperative, and in many centres the standard initial investigation is a CT scan from the skull base to the diaphragm. The differential diagnosis includes: skull base tumours (and iatrogenic paralysis secondary to surgery for such

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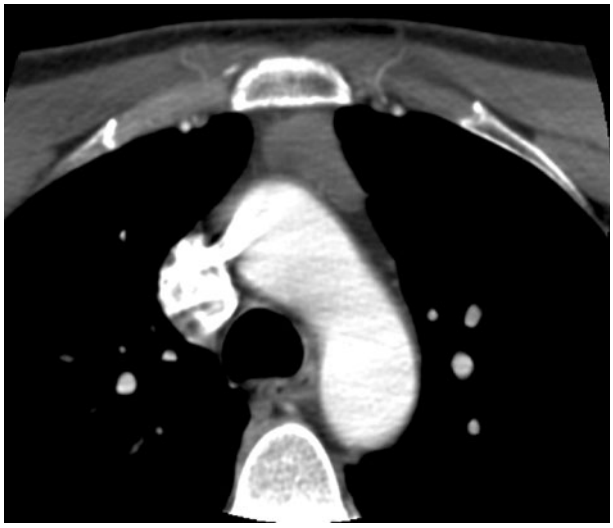

FIG. 1
Axial computerised tomography scan of the thorax with contrast, showing a well defined, bi-concave mass in the anterior mediastinum.

tumours); benign or malignant lesions of the larynx, neck or mediastinum; and a post-operative complication of thyroid, neck or cardiac surgery. In some patients, no cause is found and the unilateral palsy is labelled 'idiopathic'. In this 55-year-old, non-smoking man, the cause of his unilateral vocal fold palsy was rare: a mediastinal benign thymic cyst.

The thymus is the main organ of the lymphoid system during childhood, with regression occurring after puberty. Therefore, most thymic pathologies occur in the first two decades of life.[2] Embryologically, the thymus develops from the ventral aspect of the third and fourth pharyngeal pouches during the sixth week of embryonic life.[3] The ventral wing of the third pharyngeal pouch becomes the thymus, with only a small contribution from the fourth pharyngeal pouch.[4,5] The dorsal wing becomes the inferior parathyroid glands and elongates into a structure known as the thymopharyngeal duct.[4,5]

During the seventh to 10th weeks of embryological development, the primordial thymus migrates into the mediastinum, and the proximal portion of the thymopharyngeal duct should atrophy. Midline fusion of the thymic primordium is completed by week nine.[6] By the third embryonic month, the thymus develops a cortex and medulla, and Hassell's corpuscles (epithelial cells aggregated into concentric, 'onion-skin' layers of keratinised cells)[4] are evident.[3] The thymus increases in size until puberty, being at its relatively largest size at the age of four years. Post-puberty, the lymphoid tissue is replaced by fat or connective tissue, but Hassell's corpuscles remain.[5] Thymic tissue may be present anywhere along this route of descent.

Thymic cysts are rare,[7,8] benign lesions of the mediastinum (Takeda and colleagues identified only 30 patients over a 50-year period)[9] which comprise only 5 per cent of all mediastinal cysts and only 1 per cent of all mediastinal tumours.[10] Within the mediastinum, thymic cysts occur at any level from the base of the neck to the diaphragm.[11] In 1938, Speer[12] suggested five possible mechanisms for the development of thymic cysts: (1) from embryonal remnants of the thymopharyngeal ducts, branchial clefts or thymic tubules; (2) from sequestration products and pathological involution of the gland; (3) from degeneration of Hassell's corpuscles; (4) from lymphatics, blood vessels or connective tissue in various stages of thymic development, hyperplasia or involution; and (5) from neoplastic processes in the lymphoreticular system. Currently, there are two main schools of thought regarding the pathological origin of thymic cysts, both variants of Speer's mechanisms. Bieger and McAdams[8] believe these cysts to be congenital in origin, deriving from the thymopharyngeal duct. Conversely, Graeber *et al.*[13] divide thymic cystic lesions into congenital, neoplastic and degenerative. The latter may develop following chemotherapy for Hodgkin's disease.

Thymic cysts, which are usually located in the anterior mediastinum, are generally believed to be asymptomatic,[11,13] with most being discovered incidentally on a chest radiograph.[8] In Graeber and colleagues' series, only 13 per cent of patients were symptomatic.[13] However, in Takeda and colleagues' series, 40 per cent of patients were symptomatic, with chest pain and hoarseness being the two most common symptoms.[9] Other symptoms included dyspnoea, cough, fever and dysphagia. Presentations with respiratory distress,[11] tracheomalacia[14] and Horner's syndrome[15] have also been reported. Rapid enlargement may occur as a result of haemorrhage within the cyst cavity or secondary infection.[13] Symptoms such as dyspnoea, dysphagia, weight loss, cardiac tamponade and eventration of the diaphragm have all been reported,[13] and make it extremely difficult to differentiate thymic cysts from malignant mediastinal disease. Isolated vocal fold paralysis in association with a thymic cyst limited to the mediastinum has not previously been reported. One case report describing vocal fold paralysis in association with a cervico-mediastinal thymic cyst was identified.[16]

Thymic cysts may be uni- or multilocular. They are commonly elongated, with both ends tapered to a tract or fold.[6] The fluid may be clear, amber, brown, red or gelatinous.[3] The cyst may also contain semisolid material with necrotic debris and old blood.[17] The epithelial lining may be ciliated, nonciliated, stratified, pseudostratified, cuboidal or columnar.[3] The cyst wall thickness varies between a few millimetres and a centimetre[5] and contains thymic tissue with Hassell's corpuscles.[3] There may be evidence of cholesterol crystals and giant cell reaction.

The diagnosis of a thymic cyst may be a coincidental finding on a chest radiograph undertaken for other reasons. All but the smallest thymic cysts are likely to be visible on postero-anterior chest radiographs, with lateral radiographs determining the exact compartment of the mediastinum involved. Computerised tomography scans are an important part of the investigation of mediastinal masses. They are helpful in determining the exact location of the mass, its relationship to other structures and its tissue density. It may be useful to perform T2-weighted magnetic resonance imaging (MRI) in order to distinguish between solid and cystic masses and between mediastinal masses and vascular anomalies. However, small thymic cysts may not be detected on MRI because of the lack of characteristic signs. Computerised tomography guided fine needle aspiration and core needle biopsies are undertaken in some centres, and may be useful in differentiating cysts from solid tumours such as thymomas, germ-cell tumours and lymphomas.

The management of thymic cysts remains controversial. Whilst they are benign lesions, it is not always possible radiographically to distinguish them from malignant lesions that have undergone cystic change (particularly thymoma, teratoma, lymphoma and seminoma). Treatment options include careful observation, therapeutic or diagnostic CT-guided aspiration, and surgical resection. Most authors favour the latter, particularly in symptomatic patients. Surgical advances now allow complete resection with minimally invasive surgery.[18]

https://doi.org/10.1017/S0022215107001326 Published online by Cambridge University Press

Conclusion

Mediastinal thymic cysts are rare, representing only 1 per cent of all mediastinal tumours. Many are asymptomatic, being an incidental finding on a chest radiograph. Mediastinal thymic cyst symptoms, including weight loss, dysphagia, dyspnoea and cough, make it difficult to differentiate these cysts from malignant mediastinal tumours. The presentation of our 55-year-old patient with unilateral vocal fold paralysis was highly suspicious of a malignant lesion. Appropriate investigations may confirm or refute suspicions but surgical resection is the most suitable management strategy, as a definitive diagnosis can be made and most patients recover completely.

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Dr S L Woolley takes responsibility for the integrity of the content of the paper.
Competing interests: None declared