



A case of Mullerian cyst resected by video assisted thoracoscopic surgery

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Abstract: Ever since Hattori reported first case of Mullerian cyst of mediastinum in 2005, there has been several new publications in the literature. We report a case of 53-year-old female patient with an incidentally found mass in right paravertebral location. The lesion was removed using video-assisted thoracic surgery (VATS) procedure. Histopathology examination revealed a ciliated cyst of Mullerian origin.

Keywords: Cyst; mediastinum; Mullerian cyst; video-assisted thoracic surgery (VATS)

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Introduction

Posterior mediastinum is common location of neurogenic tumours and neurenteric cysts. In 2005, Hattori described so far unknown cyst of this region, with probable Mullerian histogenesis (1).

We report a case of 53-year-old woman with Mullerian congenital cyst that was resected using video-assisted thoracic surgery (VATS) approach.

Case presentation

A 53-year-old, non-smoking woman was admitted to thoracic surgery clinic for suspicion of neuroma of posterior mediastinum. Tumour was accidentally detected in chest radiograph. After initial diagnostics in pulmonary clinic patient was qualified for surgery. Patient did not report any symptoms. Patient was taking hormone replacement therapy.

Physical examination and laboratory tests did not presented any abnormalities. The chest computed tomography (CT) demonstrated a cystic mass in right lateral part of the 4th vertebra, 22 mm × 30 mm × 15 mm in size (*Figure 1*). Magnetic resonance imaging of the chest was not performed. In radiological imaging neurenteric cyst was

diagnosed. The mass was removed using VATS two-portal approach (4th anterior axillary and 6th middle axillary intercostals space). Camera revealed lucent cyst with thin wall. During operation the cyst was opened and yellow fluid was evacuated. Rest of tissues was dissected and removed. Location of resected mass was coagulated using argon plasma coagulation. Chest drain was removed in second day after operation.

In the microscopic examination there were pieces of fibrous connective tissue with a single smooth muscle fiber. In the center there was cystic lesion lined by single layer of cuboidal epithelial cells without papillary structures or mucin secretion. Epithelium did not demonstrate cellular atypia and there were no mitosis. Differential diagnosis taken under consideration was Mullerian cyst, enteric cyst and unusual variant of bronchogenic cyst. Additional immunohistochemistry (IHC) tests had been processed. Pan-Cytokeratins (CK AE1/AE3), cytokeratin 7 (CK7) and estrogen receptor (ER) were homogeneously, strongly positive. Focal expression of transcription factor PAX8, progesterone receptor (PR) and cancer antigen 125 (CA125) were noticed. A few epithelial cells were also stained by anti-CD56 antibody. Staining for thyroid transcription factor (TTF-1), cytokeratin 20 (CK20), CDX2 were uniformly negative. Ki-67 mitotic index were less than

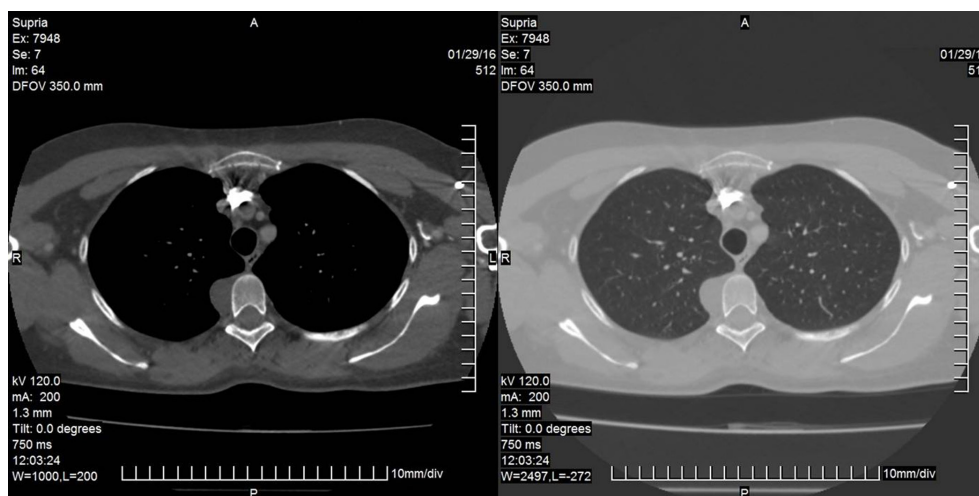


Figure 1 A cystic mass in right lateral part of the 4th vertebra, 22 mm × 30 mm × 15 mm in size.

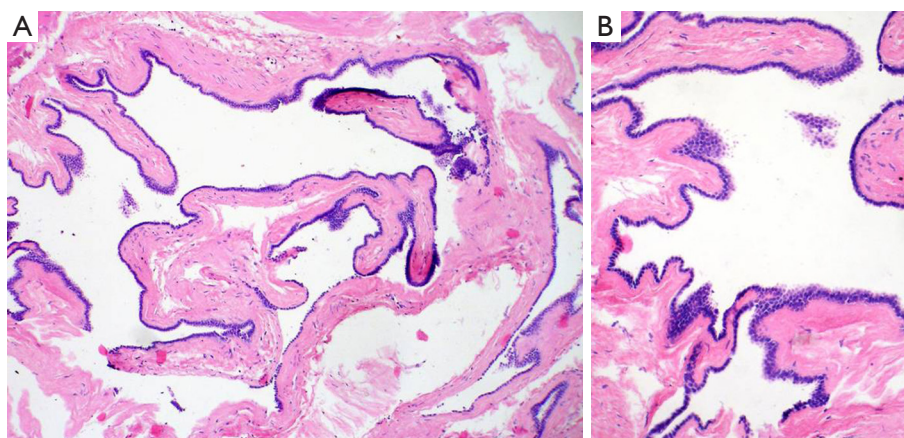


Figure 2 Hematoxylin and eosin staining (A, ×100; B, ×200) showing cystic lesion lined by single layer of cuboidal epithelial cells without atypia or mitosis.

1%. The final diagnosis was Mullerian cyst (Hattori's cyst) due the microscopic image and typical ER, PR, Ca125 and PAX8 staining (Figures 2,3).

Patient's postoperative course was uneventful. She was discharged on the third after surgery.

Discussion

Mullerian cysts are typically observed in genitourinary organs in pelvic region (2). Since first report in 2005 by Hattori two retrospective studies concerning mentioned cysts were performed. Hattori found this mass in 3 of 19 cases of mediastinal cysts (15.8%). However, in larger

patient group Thomas-de-Montarville confirmed only 7 cases of Mullerian cyst from 163 mediastinal cysts (4.3%) (2,3). In our clinical experience it was first incidence of these pathology.

The histogenesis of Mullerian cyst in the mediastinum is unknown. According to Ludwig's theory—lesion is a result of agenesis of primary Mullerian apparatus. In Mayer-Rokitansky-Küster-Hauser (MRKH) syndrome similar pathogenesis is observed (2,4). However, Hattori affirm that the cysts are misplaced mesothelium with Mullerian characteristics.

According to Kabayashi's study—Mullerian cysts usually occur in woman at the age of 40 to 60. Only Hattori in

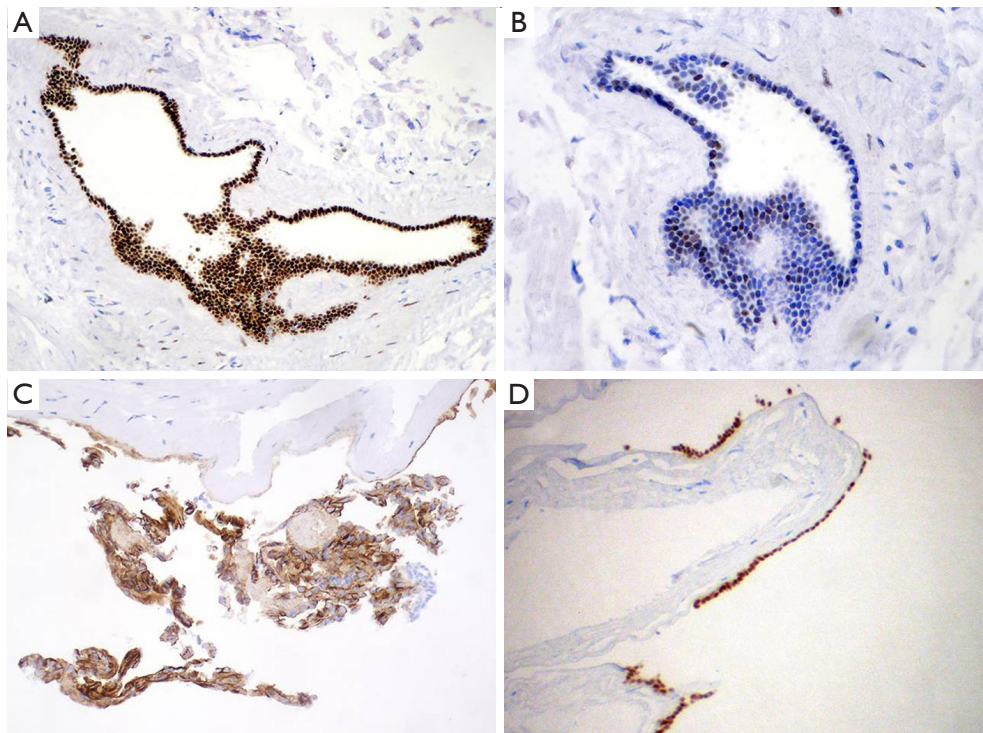


Figure 3 Immunohistochemistry. (A, $\times 200$) Strongly nuclear positive estrogen receptor (ER) staining; (B, $\times 200$) focal expression of progesterone receptor (PR); (C, $\times 100$) positive membranous staining for CA125; (D, $\times 100$) focal nuclear positive staining for transcription factor PAX8.

his original study reported this cyst in 18-year-old patient (2,4). Typical location of pathology is paravertebral space between 3rd and 8th thoracic vertebra (3,4). Our patient was a 53-year-old woman and cyst was located in paravertebral region of 4th vertebra. The most common symptoms are cough and chest pain. Dakak *et al.* reported a patient with dysphagia (5). Our patient did not notice any symptoms. Most Mullerian cysts of mediastinum occur in pre menopausal period. Obesity, hormone replacement therapy, a hysterectomy and an oophorectomy is also observed in many cases. Our patient was in menopausal period and was using hormonal replacement therapy. Differential diagnosis includes neurenteric, bronchogenic, gastroenteric and mesothelial and thoracic duct cysts. In microscope imaging typical neurogenic cysts are lined by columnar cells with neural or enteric differentiation, while mesothelial cysts are lined by a single mesothelial cells layer. Thoracic duct cyst may not communicate with the duct and is lined by respiratory or squamous epithelium. Enterogenous and bronchogenic cysts have lined by columnar epithelium with a smooth muscle bundles as well

as Mullerian cyst (5). IHC confirm negative expression or weakly positive of cytokeratin 5/6, and positive expression of ER and PR. ER and PR are considered to be the best markers of Mullerian cysts. In presence case ER was positive and PR focally positive. Recurrences were not reported.

In conclusion female patient in between 40 to 60 years of age with obesity or gynecological history and with cyst in paravertebral location is considered to differential diagnosis for Mullerian cyst.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <http://dx.doi.org/10.21037/vats.2017.05.03>). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

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