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Large cervical mass and head and neck cancer. First report of ectopic papillary adenoma of the lung

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ABSTRACT

Aim: We aim to report the first case of ectopic papillary adenoma of the lung (PAL), identified in the neck.

Methods: A 62-year-old woman with a known history of thyroid cancer treated several years ago, presented at our department because affected by an unpainful latero-cervical neck mass. The patient performed neck ultrasound echography, blood tests and computed tomography (CT) of head, neck and thorax to exclude the recurrence of the thyroid tumor or other unknown cancer of head and neck.

Results: Her CT with contrast identified a large mass in the left thyroid loggia, with well-defined margins without post-contrast enhancement; the mass laterally displaced the trachea, without reduction of airways caliber. The tumor was removed. The histologic analysis showed aspects typical of PAL. Because the mass was completely removed, and no lymph nodes were identified, additional treatments were excluded.

Conclusions: Despite the ectopic tissue can be rarely identified in the neck, it should be always considered because it might degenerate and cause, as in our case, a tumor like PAL. The diagnostic plan is fundamental to well manage latero-cervical mass avoiding risks for patients.

Dear Editor,

We present in this letter the first case of papillary adenoma of the lung (PAL) which arose from ectopic tissue in the neck; an asymptomatic, laterocervical mass was the only sign/symptom of the tumor. The mass was suspected to originate from a tumor in the head and neck, and the patient performed appropriate investigations. None of the examinations was supportive for a tumor presence/recurrence, but because of the unknown origin of the mass, we decided to remove it. Histologic analysis revealed PAL.

PAL is a rare lung tumor [1–3], described for the first time by Spencer [2] in 1980; the tumor arises in the pulmonary apex and is asymptomatic [3]. Because of absence of the symptoms, PAL is usually discovered as an incidental lesion [4]. To our knowledge, this is the first report that describes PAL arising out of the lung.

A 62-year-old woman, who has undergone total thyroidectomy three years ago for a massive goiter, presented to the Otolaryngology department of San Giovanni Addolorata Hospital (Rome, Italy) in March 2021 because she noted a mass on the left side of her neck. The lesion, which occurred three months ago, was clearly visible, unpainful, soft, and immobile on the surrounding plans. Her endoscopy showed a right-sided tracheal deviation without significant reduction of the airway space. The laboratory tests were normal. Her neck ultrasound (US) showed an anechoic mass (70x49x102 mm) in the thyroid loggia and, confirmed the deviation of the trachea on the right. A computer tomography (CT) was performed to exclude recurrence of thyroid disease and to identify any other occult cancers [5].

Contrast-enhanced neck and chest CT showed a large mass (77x52x107 mm) in the left thyroid loggia, with well-defined margins, without contrast enhancement; the mass caused the deviation of the trachea on the right (as previously identified). Moreover, the lesion invaded the anterior jugular triangle and the antero-superior mediastinum for a length of 45 mm (Figure 1 AB). No other abnormal findings were identified on the CT.

Based on the clinical, laboratory, and imaging findings, lymphocele was suspected. We planned the removal of the mass. Transcervical approach was performed for better viewing and managing of any adverse event during surgery, due to the unknown origin of the mass. After removal, the finding was sent to the pathologist for specific analysis. We did not observe any complications during or after surgery.

Pathological findings (Fig.2) showed cystic aspect with an endocystic papillary growth pattern; the lining epithelium presented few atypia and rare mitotic figures. Immunohistochemistry was performed to clarify the nature of the tumor. A positive reaction was identified for CK7 and strong positive for CKAE1AE3 and TTF1. The immunohistochemistry was negative for CK20, tireoglobulin, WT1, CK5 and calretinin. Based on the pathology findings the diagnosis PAL was made. Because of the complete removal of the mass and the absence of other pathologic findings, in agreement with the oncology team, we decided to avoid further treatments.

The patient was discharged 7 days after surgery with the definitive diagnosis of “PAL in the neck arising from ectopic pulmonary tissue.”

Her follow-up CT, performed six months after surgery showed no recurrence of the disease and normal anatomy of the neck and thoracic region.

PAL is a rare tumor that occurs in the apex of the lung; it is generally benign but has undetermined malignant potential [6], and in case of

Abbreviations: PAL, Papilalry Adenoma of Lung.

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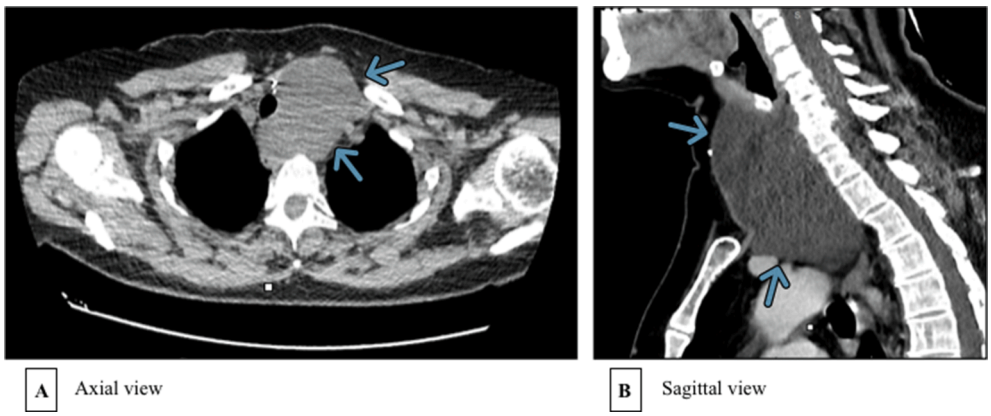

Fig. 1. (A) Axial section of contrast-enhanced CT showed a large mass (77x52x107 mm)(arrow) located in the left thyroid loggia, with well-defined margins, right deviation of the trachea and infiltration of the antero-superior mediastinum. (B) Sagittal section of contrast-enhanced CT.

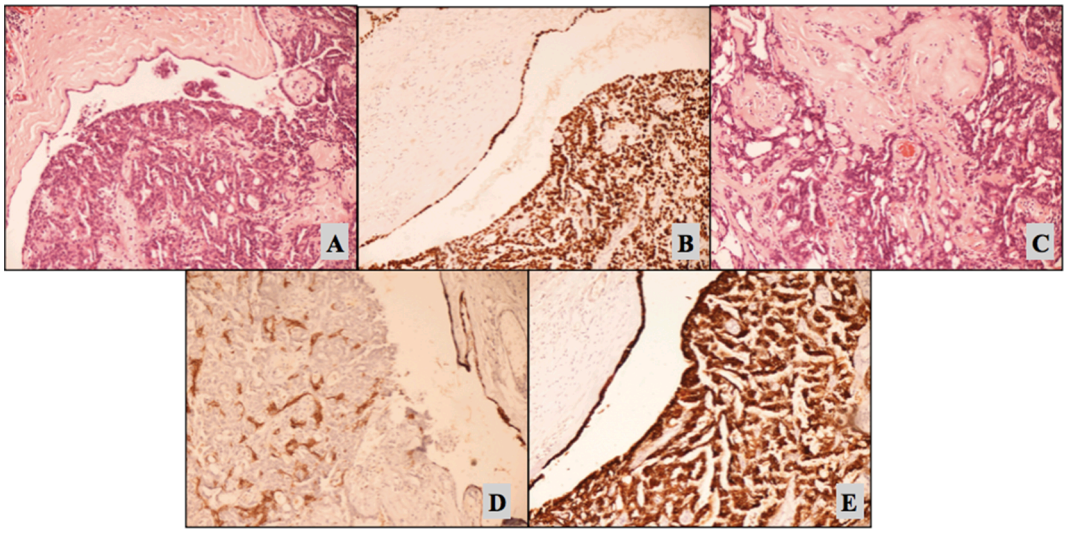

Fig. 2. Histologic features of present case. (A) papillary intracystic pattern delimited by cubic epithelium; (B) high magnification showing low grade atipica and rare mitosis; (C) positive thyroid transcription factor 1 (TTF-1) staining; (D) vascular axes positive for cytokeratin (CK) 5; (E) cytokeratin (CK) AE1/AE3 strongly positive staining.

tumor conversion, PAL can become a life-threating condition. In fact, the cancer, which is asymptomatic, can disseminate from the apex of the lung to the neck and mediastina, causing compression of the trachea, cough [4], dysphonia and airway obstruction.

In our case, the cervical mass was not caused by the dissemination of the tumor on the surrounding, but it was originated by the malignant transformation of ectopic lung tissue in PAL. The pathogenetic hypothesis for this unusual finding could be that the patient had an embryonic residue of lung tissue in her neck. The epithelium of the bronchi is entirely of endodermal origin. In the fourth week of the embryogenesis, the respiratory diverticulum originates from the ventral wall of the foregut (the anterior part of the alimentary canal); during its separation from the foregut, the pulmonary bud forms the trachea and bronchial buds. These structures dilate into the main bronchi (right and left) in the fifth week. In our patients, we hypothize that during this separation and subsequent expansion into the body cavity, the embryonic residual of lung remained into the neck. Then after years, probably under inflammatory and/or neoplastic stimuli [7] PAL originated from this tissue.

Because of our patient's cancer history, the clinical screening included US and CT to rule out recurrence of thyroid cancer. Because we identified PAL, we think that it has been the best clinical approach. Generally, laterocervical masses are studied by US and then by eco-guided biopsy to clarify their malign or benign nature. In our case, eco-guided biopsy, which is generally safe, would have been dangerous because the potential diffusion of the cancer cells in the surrounding tissue and, the invasiveness of the procedure. In fact, the insertion of a needle into the mass could trigger malignant transformation of the tumor [7]. CT scan, despite x-ray exposure, allowed to exclude positiveness for cancer (non-enhancing lymph node) and helped to define the margin and the diffusion of the mass. The latter facilitated the tumor removal.

All experts in head and neck disease and oral pathology, who find neck mass (es) during a clinical examination, should bear on mind the rare tumors. Although the lymph node bundle is generally the most likely mass in this area, other types of diseases can occur, especially in the absence of a primary tumor. Proper investigative management can reduce the number of tests and avoid the risk of invasive procedures.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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