

A case report of a swollen buccal fat pad revealing an aggressive lung tumor

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Case Report

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ABSTRACT

Large cell neuroendocrine carcinomas (LCNEC) are rare and aggressive tumors that often present significant diagnostic challenges due to their lack of distinctive clinical symptoms. This type of cancer is particularly difficult to diagnose because it does not typically present with specific or easily recognizable signs, leading to delays in diagnosis and treatment. The prognosis for patients with LCNEC is generally poor, and the optimal therapeutic approach remains a topic of ongoing debate within the medical community.

In this case, we report an unusual presentation of LCNEC in a 75-year-old man who initially sought medical attention for a swollen cheek, which had been gradually worsening over a three-month period. Upon further investigation, a mass was identified in the buccal fat pad, and a biopsy was performed, leading to a diagnosis of LCNEC based on histological findings. Subsequent evaluations revealed that the primary tumor originated in the lung and had metastasized extensively. Palliative chemotherapy was initiated in response to the advanced stage of the disease.

Key Words: Large cell neuroendocrine carcinoma, lung, buccal fat pad, metastasis

Received: 17 August 2024, **Accepted:** 24 January 2025.

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ISSN: 2090-097X, January 2025, Vol. 16, No. 1

INTRODUCTION

Large cell neuroendocrine carcinoma (LCNEC) is a recognized histologic entity whose clinical features have not yet been well defined. They are rare and aggressive tumors with a poor prognosis.

We present a case of a 75-year-old gentleman who first presented with a distant metastasis to the cheek of a large cell neuroendocrine carcinoma of the lung.

CASE PRESENTATION:

A 75-year-old patient presented to our department with a swelling of the left cheek evolving for 3 months. His medical history revealed a high blood pressure, a diabetes and a hypothyroidism all of which were balanced under medical treatment, plus a cataract surgery. However, no history of smoking was noted. Moreover, respiratory symptoms and other symptoms such as weight loss were denied.

Clinical examination objectified a painless hard lump on the left cheek. A face MRI scan was performed, showing a

mass of the buccal fat pad [Figures 1a & 1b].

The patient underwent an excisional biopsy [Figures 2a & 2b]. Histological examination reported an undifferentiated malignant tumoral process.

Subsequent immunohistochemistry revealed cells staining positive for pan cytokeratin, P63, synaptophysin, cytokeratin-7, but negative for P40, desmin, chromogranin and cytokeratin-20. Final report favored a diagnosis of large cell neuro endocrine carcinoma.

A CT scan was performed with the dual purpose to determine the primary or secondary nature of the tumor and to assess its extension, revealing: a 7 cm excavated right lung tumor mass with diffuse metastases to the anterior mediastinum, retroperitoneum, sigmoid, kidney, adrenal gland and a peritoneal carcinomatosis. [Figures 3a & 3b].

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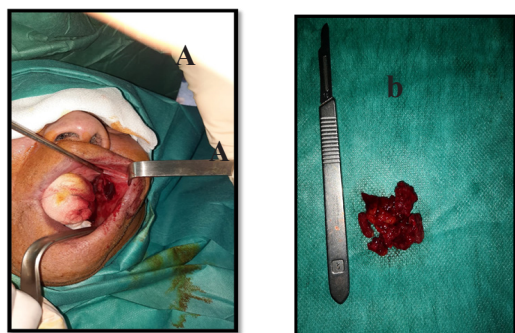


Figure 1. T1-weighted MRI with contrast showed a tissue nodule within the buccal fat pad:

a. Axial view; b. Coronal view

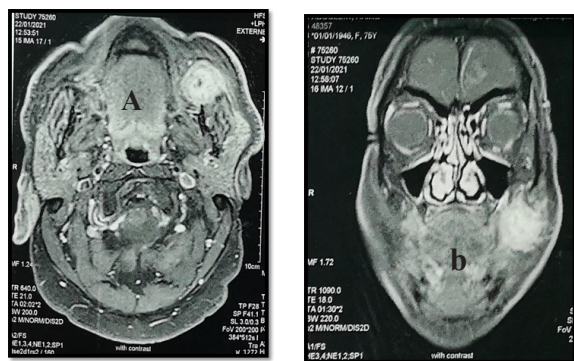


Figure 2a. Intra-operative image of the tumor excisional biopsy via transoral approach.

Figure 2b. Image of the operatory piece.

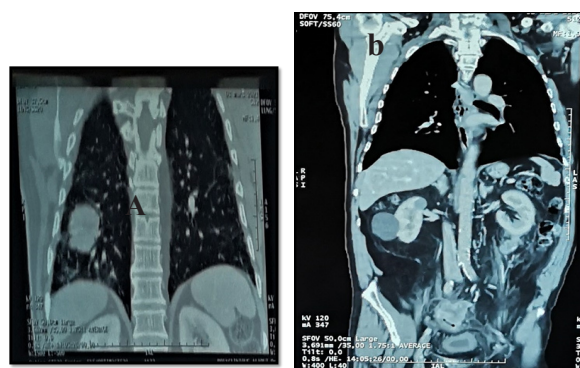


Figure 3. Coronal reconstruction of the CT scan showing:

a. an excavated tumor mass in the inferior lobe of the right lung.

b. a right kidney mass and a left adrenal gland mass.

DISCUSSION

LCNEC is a group of neuroendocrine carcinomas in the lungs and bronchial tubes, accounting around 1.6–3.1% of all pulmonary cancers [1]. According to the WHO, a LCNEC is a tumor with: a neuroendocrine architecture (nests, trabeculae, rosettes, palisades), large tumor cells with moderate to abundant cytoplasm, frequent nucleoli, a high mitotic index: ≥ 11 mitoses per 2 mm², average 70 per 2 mm², necrosis, one or more positive neuroendocrine markers on immunohistochemistry: chromogranin, synaptophysin and CD56. One marker is sufficient if $> 50\%$ of tumor cells [2].

One of the difficulties in diagnosis is the variety and lack of clinical specificity of symptoms. It has been reported that patients with LCNEC are less possibly to present with respiratory symptoms such as cough, hemoptysis or pneumonia [3]. This often leads to a delay in consultation and diagnosis at metastatic stages. This was the case of our patient who presented with a swollen cheek, without any other somatic complaint although he had a stage IV metastatic disease.

LCNEC is a biologically aggressive tumor and the poor prognosis is mainly related to early metastatic disease [2]. Furthermore, prognosis is poor even in patients with potentially resectable stage I lung cancer with 5-year survival rates ranging from 27% to 67% [4].

CONCLUSION:

Currently, there is no standard treatment of pulmonary LCNEC, and the knowledge base supports small cell neuroendocrine carcinoma (SCLC)-based regimes in managing patients with LCNEC, on this wise, chemotherapy is used for patients with stage IV disease [5,6]. Hence, our patient was referred to the department of oncology for palliative care.

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