Article 6

PAMJ Clinical Medicine

Case report

Mandibular metastasis in a patient with undiscovered lung cancer: a case report

Raouaa Belkacem Chebil, Touil Dorsaf, Lamia Oualha, Zouha Mziou, Badreddine Sriha, Nabiha Douki

Corresponding author: Raouaa Belkacem Chebil, Department of Oral Medicine and Oral Surgery, Faculty of Dental Medicine, Monastir University, Monastir, Tunisia. raouaabelkacem@gmail.com

Received: 12 Aug 2020 - **Accepted:** 12 Nov 2020 - **Published:** 26 Nov 2020

Keywords: Oral metastasis, lung cancer; prognosis.

Cite this article: Raouaa Belkacem Chebil et al. Mandibular metastasis in a patient with undiscovered lung cancer: a case report. PAMJ Clinical Medicine. 2020;4(119). 10.11604/pamj-cm.2020.4.119.25514

Available online at: https://www.clinical-medicine.panafrican-med-journal.com//content/article/4/119/full

Mandibular metastasis in a patient with undiscovered lung cancer: a case report

Raouaa Belkacem Chebil[1,2,&], Touil Dorsaf[1,2], Lamia Oualha[1,2], Zouha Mziou[3], Badreddine Sriha[4], Nabiha Douki[1,2]

[1]Department of Oral Medicine and Oral Surgery, Faculty of Dental Medicine, Monastir University, Monastir, Tunisia, [2]Laboratory of Research on Oral Health and Oro-Facial Rehabilitation (LR12ES11), Faculty of Dental Medicine, Monastir University, Monastir, Tunisia, [3]Department of Stomatology and Maxillo Facial Surgery, Faculty of Medicine, Sousse University, Sousse, Tunisia, [4]Department of Cytology and Pathological Anatomy, Faculty of Medicine, Sousse University, Sousse, Tunisia

[&]**Corresponding author**

Raouaa Belkacem Chebil, Department of Oral Medicine and Oral Surgery, Faculty of Dental Medicine, Monastir University, Monastir, Tunisia

Abstract

Metastatic tumors account for 1% of all oral malignancies. Jawbone metastasis is uncommon. The mandible is the most frequent site. It is rare in the oral soft tissues, accounting for only 0.1% of oral malignancies. The majority of metastatic cases (70%) reported in the literature have primary tumors located in the lung, breast, kidney and colon. The clinical presentation of metastatic tumors is variable. This may create diagnostic dilemma and may lead to erroneous diagnoses. In this work, we present the radiological monitoring and the diagnostic dilemma of a mandibular metastasis resulting from a lung cancer. We report a case of a 57-year-old man who was referred to our department for the treatment of a left buccal cellulitis. On extra-oral examination, the patient presented a non-painful mass on the left mandibular ramus. The panoramic radiography showed a radiolucent lesion in the ramus with enlargement of the mandibular foramen. After excision of the jaw mass, histology revealed a metastatic adenocarcinoma having a lung origin. Mandibular metastasis may be the first presenting feature of an underlying occult malignancy of another primary site. Clinicians should always maintain a high index of suspicion for the possibility that a radiolucent lesion could be a metastatic tumor. Lip and chin hypoesthesia is an important symptom for malignancy detection.

Introduction

Metastatic tumors in the oral region are uncommon, they account for about 1% of all oral malignant neoplasms [1]. In approximately 20% of the cases, oral metastases are discovered before the detection of the distant primary tumor [1]. Most oral metastases involve the jawbones, especially the mandible which is the most common site. With regard to the oral soft tissues involvement, it is rare and it is mostly located in the gingiva and the tongue [2]. Jawbone metastasis mainly occurs in the posterior region of the mandible, the ramus and the condyle, which are rich in red bone marrow [3]. The most common primary tumor sites for jawbone metastases in women are the breast, adrenals, genital organs and thyroid. However, the most frequent sites of primary tumors in men are the lungs, prostate, kidney, bone and adrenals [4,5]. The most common histological type is adenocarcinoma followed by squamous cell carcinoma [1]. The dentist has an important role in these patients´ management and especially in making an early diagnosis. The present paper reports the radiological monitoring and the diagnostic dilemma of a mandibular metastasis in a patient with undiscovered lung cancer.

Patient and observation

A 57-year-old man was referred to the Department of Oral Medicine and Oral Surgery at Sahloul Hospital for a diffuse left buccal swelling evolving for a week before arriving to the hospital. A review of his medical history revealed a coronary syndrome with a history of myocardial infarction. The patient was a smoker with 40 packs a year. The initial clinical examination confirmed the diagnosis of cellulitis of dental origin. The patient was treated with Amoxicillin® (at a daily dose of 2g for one week) and the extraction of the decayed teeth (28 and 27). After one week, the patient showed improvement of the buccal swelling but the extra-oral examination revealed a persistent painless tough mass on the left mandibular ramus, measuring about 3*3 cm and associated with a trismus and a left side hypoesthesia of the lower lip and chin (Figure 1). The overlying skin was normal with presence of bilateral mandibular lymphadenopathy. The interrogatory revealed that the mass had been dating since one month.

The intra-oral examination revealed a poor oral hygiene without showing any mucosal or dental pathological changes. The panoramic radiography revealed a radiolucent lesion with ill-defined margins in the ramus and enlargement of the mandibular foramen (Figure 2). Compared to the

panoramic radiography performed one month before consulting at our department, we noticed a rapid evolution of an osteolytic process (Figure 3). The computed tomography scans showed a hypodense lesion with infiltration of soft tissues (Figure 4).

Based on these clinical examinations and imaging data, a malignant process was suspected, including osteosarcoma. The lesion was surgically excised with lymph node dissection and the histopathological examination revealed neoplastic cells with hyper-chromatic nuclei and acidophilic cytoplasm arranged in abortive glandular structures (Figure 5,Figure 6). Diagnosis of a metastatic adenocarcinoma was made. The specimen was strongly suggestive of metastatic adenocarcinoma from the lung. The patient underwent other examinations including chest X-ray and abdomen computed tomography (CT) scan. The chest X-ray showed a rounded mass, filling the right lower lung field. The pulmonary lesion was subjected to biopsy which revealed neoplastic cells arranged in glandular structures with high mitotic activity (Figure 7). Diagnosis of adenocarcinoma was therefore confirmed. The lung was discovered as the primary site. A whole body CT scan and a scintigraphy examination revealed distant bone metastases. The patient was referred to the regional Clinical Oncology Department for palliative treatment by chemotherapy (Navelbine® and Cisplatine®) but he died after 5 months.

Discussion

The mandible is a rare site for the development of metastases. Jawbone metastasis represents 1% of all malignant oral tumors [6]. Seventy per cent of metastases are located in the mandible, especially in the molar region. In our case, the mandibular ramus was affected. The abundant vascularization of this region and its richness in hematopoietic cells can explain this preferential location. However, some authors suggest that it could be due to the inferior alveolar vessels angulation leading to the slowing down of the blood stream [4]. For others, the bone marrow is an attractive site for metastasis because it contains growth factors, which promote tumor cell proliferation and survival. In addition, the vascular spaces are sinusoidal in nature, which facilitates the penetration and proliferation of cancer cells [7]. The pathogenesis is still unclear but it is thought to be a multistage process in which cells detach themselves from the primary tumor and get transported by lymphatic or blood vessels. Recent studies have stated that cancer cells metastasizing to bone have shown their ability to alter the physiological balance between both bone resorption and bone formation [8]. The peak incidence of oral metastasis is observed between the age of 50 and 70 years [6]. Our patient was 57 years old.

Most cases are diagnosed with reference to the primary tumor. However, in about 20% of cases, metastatic mandibular tumor allows, based on a thorough clinical examination and radiological investigations, the diagnosis of the primary tumor. In fact, in about 30% of patients with jawbone metastasis, the primary tumor is asymptomatic and not diagnosed [4]. For our patient, the reason for the referral to our department was swelling. The possibility of a tumor was not mentioned by his treating dentist. Lung cancer classification comprises two large groups: Non-Small Cell Lung Carcinoma (NSCLC) and Small Cell Lung Carcinoma (SCLC). NSCLC (80% of all lung cancers) comprises adenocarcinoma, squamous cell carcinoma and large cell carcinoma. On the other hand, SCLC (20% of all lung cancer cases), which is strongly associated with cigarette smoking, has an aggressive behavior [9]. So far, there are few reported cases of metastatic SCLC to the oral cavity. In the case presented herein, bronchial biopsy and mandibular excision showed a moderately differentiated adenocarcinoma. Thus, diagnosis of mandibular metastasis was established.

Clausen and Poulsen described the criteria for diagnosing metastatic lesions as: proved primary

tumor with histopathological confirmation and radiographic evidence, mandibular metastasis with histopathological evidence and radiographic evidence and histopathological correlation of the metastatic lesion with the primary site. In case of close proximity to the primary site, there must be a wide margin around the primary site to eliminate direct extension from the primary lesion [4]. In our reported case, there was similarity in the histopathological presentation of both the primary and the metastatic lesions. The diagnosis of mandibular metastasis constitutes a challenge because its clinical symptomatology is nonspecific and it is characterized by a high clinical latency causing a delay in diagnosis. The clinical presentations of metastatic lesions in the mandible include pain, rapidly progressing swelling, paresthesia, difficulty in chewing, dysphagia, disfigurement, cervical lymphadenopathies and rare pathologic fracture [10]. For our patient, a firm swelling with ill-defined borders, paresthesia and an opening mouth limitation were the only manifestations of an undiscovered disseminated malignancy.

People having jawbone metastasis often present vague or innocuous symptoms that can mimic dental infections [11]. For this reason, the dentist must perform a thorough clinical examination in order to avoid confusion and to gain time that can be very valuable. The radiological aspect of mandibular metastasis is also nonspecific and it varies according to the nature of the primary tumor. Usually, metastatic lesions present as osteolytic areas, poorly-defined with a “moth-eaten” appearance [8]. However, metastases from sites such as prostate present as a radiopaque or mixed radiopaque-radiolucent lesion [3]. Moraes RM *et al.* reported a rare case of mandibular metastasis of small cell lung carcinoma presenting unilocular radiolucency with well-defined sclerotic borders [12]. In about 5% of cases, X-ray shows no pathological pictures [13,14]. In the present case, the metastatic lesion appeared as a radiolucent area with ill-defined borders. For all cases, confirmation of the secondary location of the tumor requires a histopathological examination of the primary tumor, which must be identical to the suspected metastatic lesion.

Clinically and radiologically, confusion with osteosarcoma is possible although the elective site of osteosarcoma is the horizontal branch and the mandibular symphyseal region [15]. In addition to not having specific clinical symptomatology, osteosarcoma clinically and radiologically presents common signs with all maxillary tumors. Regardless of its location, osteosarcoma is manifested by the presence of bone lysis, a transition zone, an ossification zone, a periosteal reaction, as well as calcification and extension in the adjacent soft tissues. Classically, we find a characteristic, but not pathognomonic image; in "grass fire" or "sunlight", corresponding to a periosteal reaction with production of osteoid tissue on the tumor periphery [15]. This radiological semiology can also be a mandibular metastasis. The pathological examination along with the investigations needed to identify the primary site confirms the diagnosis. A clinical and radiological extension assessment is necessary in order to evaluate the loco-regional extension of metastasis and to direct it towards the site of the primary cancer. A minimum assessment is required. It should include a chest X-ray, abdominal ultrasound (or thoraco-abdominal CT-scan), bone scan and CT-scan or MRI of the facial fundus. Bone scintigraphy is a sensitive examination allowing the early detection of non-visible bone lesions on standard radiography, but it remains unspecific [16]. For our case, we noted a high accumulation of isotope in the ramus, the seventh rib and the tibia bone.

Conservative therapy and palliative treatment are the main treatment options of polymetastatic cancer to improve the patient´s life quality. In this case, the patient was subjected to surgical resection of the lesion and lymph node dissection combined with chemotherapy. Surgical excision is normally indicated for solitary metastasis while our patient had multiple lesions. In general, the treatment of oral metastasis depends upon its presentation and the stage during which it was

identified. Mandibular metastatic carcinoma is usually an evidence of a widespread disease and it is associated with a poor prognosis. An average survival time for lung cancer metastasis is from 4 months to 1 year with a maximum survival rate of 5 years. The majority of patients diagnosed with oral metastases succumb to the disease within 1 year, while the four-year survival rate is estimated to be 10% [15]. Our patient´s death, 5 months after the initial diagnosis, confirmed this severe prognosis.

Conclusion

Jawbone metastasis is rare and it occurs in the advanced stages of malignancy. Understanding the characteristics of oral metastasis can help the clinician in the early diagnosis of this disease. The practitioner should always maintain a high index of suspicion for the possibility that a radiolucent lesion, with either ill-defined or well-defined borders, may be a metastatic tumor, especially in patients with known-malignant diseases. Lip and chin hypoesthesia is an important symptom for malignancy detection. A multidisciplinary medical care is necessary in order not to worsen the patient´s health condition, mostly smokers who often have a poor prognosis.

Competing interests

The authors declare no competing interests.

Authors' contributions

All the authors have read and agreed to the final manuscript.

Acknowledgment

The authors thank Pr Samir Boukattaya for the proof-reading of this article.

Figures

Figure 1: extra-oral clinical presentation: a non-painful indurated mass with ill-defined borders measuring 3x3 cm on the left mandibular ramus; the overlying skin appeared normal

Figure 2: first panoramic examination: enlargement of the mandibular foramina one month before consultation at our department

Figure 3: second panoramic examination showing rapid evolution of the osteolytic process in one month: a radiolucent lesion with ill-defined margins in the ramus

Figure 4: coronal computed tomographic view (soft tissue window) showing a lump surrounding the left ramus with irregular bone destruction and muscle infiltration

Figure 5: histopathological examination: photomicrography of the biopsy specimen showing an infiltrative tumor composed of large epithelial cell blocks arranged in abortive glandular structures (H&E 40X)

Figure 6: histopathological examination: cells had scant cytoplasm and hyperchromatic nuclei with high mitotic activity (H&E, 200X)

Figure 7: lung biopsy: photomicrography of the biopsy specimen showing large epithelial cell blocks with irregular shapes arranged in glandular patterns (arrow, H&E 100X)

References

1. Matsuda S, Yoshimura H, Yoshida H, Umeda Y, Imamura Y, Sano K. Mandibular metastasis as the first clinical indication of occult lung adenocarcinoma with multiple metastases: a case report. Medicine (Baltimore). 2018 Apr;97(15): e0296. **PubMed**| **Google Scholar**

2. Aswath N, Balakrishnan C, Shyamsundar V, Thirrugnanamurthy S. Oral gingival metastasis: a diagnostic dilemma. Indian J Dent Res. 2017;28(3): 344. **PubMed**| **Google Scholar**
3. Irani S. Metastasis to the jawbones: a review of 453 cases. J Int Soc Prev Community Dent. 2017;7(2): 71-81. **PubMed**| **Google Scholar**
4. Adewale AO, Mofoluwake LA, Olamide OT, Yussuf SA. Two case reports on mandibular metastases. Ghana Med J. 2018;52(3): 168-172. **PubMed**| **Google Scholar**
5. Raouaa BC. Mandibular metastasis in a patient with undiscovered lung cancer: a case report. Dentistry. 2016;6: 9.
6. Watanabe M, Tada M, Satomi T, Chikazu D, Mizumoto M, Sakurai H. Metastatic rectal adenocarcinoma in the mandibular gingiva: a case report. World J Surg Oncol. 2016;14(1): 199. **PubMed**| **Google Scholar**
7. Patil S, Janardhan D, Suresh S, Thomas S. A rare case of metastasis to the mandible from the thyroid gland: a case report and literature review. Indian J Surg Oncol. 2019 Sep;10(3): 422-425. **PubMed**| **Google Scholar**
8. Hirshberg A, Shnaiderman-Shapiro A, Kaplan I, Berger R. Metastatic tumours to the oral cavity - pathogenesis and analysis of 673 cases. Oral Oncol. 2008;44(8): 743-52. **PubMed**| **Google Scholar**
9. D'Silva NJ, Summerlin DJ, Cordell KG, Abdelsayed RA, Tomich CE, Hanks CT *et al*. Metastatic tumors in the jaws: a retrospective study of 114 cases. J Am Dent Assoc. 2006;137(12): 1667-1672. **PubMed**| **Google Scholar**
10. Maschino F, Guillet J, Curien R, Dolivet G, Bravetti P. Oral metastasis: a report of 23 cases. Int J Oral Maxillofac Surg. 2013;42(2): 164-8. **PubMed**| **Google Scholar**
11. Hajji F, Meziane M, Boulaadas M, et Mohamed Kzadri HB. Ostéosarcome des maxillaires: présentation de deux cas. Médecine Buccale Chirurgie Buccale. 2011;17(4): 279-282. **Google Scholar**
12. Moraes RM, Alves FA, Carvalho BFC, Costa FDA, Lopes RN, Jaguar GC. Mandible metastasis of small cell lung cancer mimicking a residual cyst. Autopsy Case Rep. 2017;7(1): 37-41. **PubMed**| **Google Scholar**
13. Bouslama S, et Habib Khochtali MO. Métastases mandibulaires: présentation de 8 cas. Médecine Buccale Chirurgie Buccale. 2010;16(4): 247-253. **Google Scholar**
14. Aerden T, Grisar K, Neven P, Hauben E, Politis C. Numb chin syndrome as a sign of mandibular metastasis: a case report. Int J Surg Case Rep. 2017;31: 68-71. **PubMed**| **Google Scholar**
15. Cai Z, Zhu C, Wang L, Zhu L, Zhang Z, Zhu H *et al*. A retrospective study of six patients with mandibular metastatic carcinoma. Oncol Lett. 2016;11(6): 3650-3654. **PubMed**| **Google Scholar**
16. Glaser C, Lang S, Pruckmayer M, Millesi W, Rasse M, Marosi C *et al*. Clinical manifestations and diagnostic approach to metastatic cancer of the mandible. Int J Oral Maxillofac Surg. 1997;26(5): 365-368. **PubMed**| **Google Scholar**

Figure 1: extra-oral clinical presentation: a non-painful indurated mass with ill-defined borders measuring 3x3 cm on the left mandibular ramus; the overlying skin appeared normal

Figure 2: first panoramic examination: enlargement of the mandibular foramina one month before consultation at our department

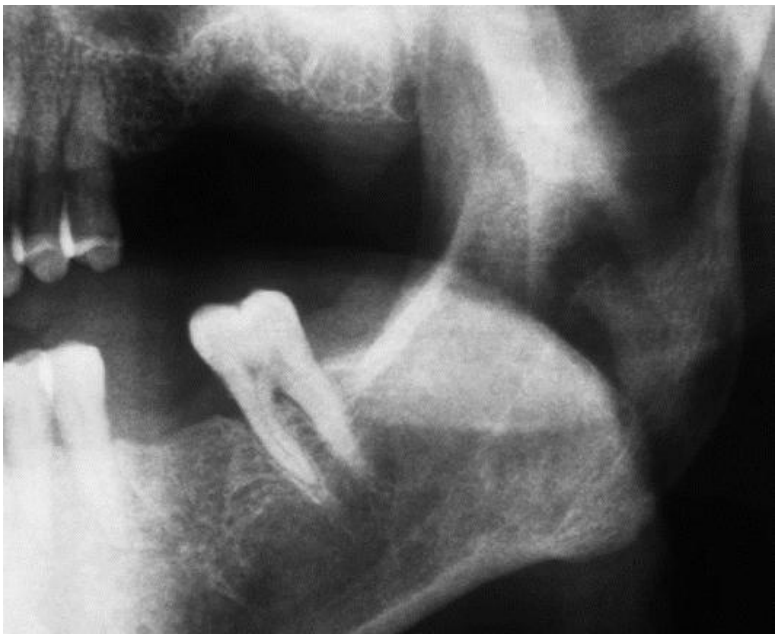

Figure 3: second panoramic examination showing rapid evolution of the osteolytic process in one month: a radiolucent lesion with ill-defined margins in the ramus

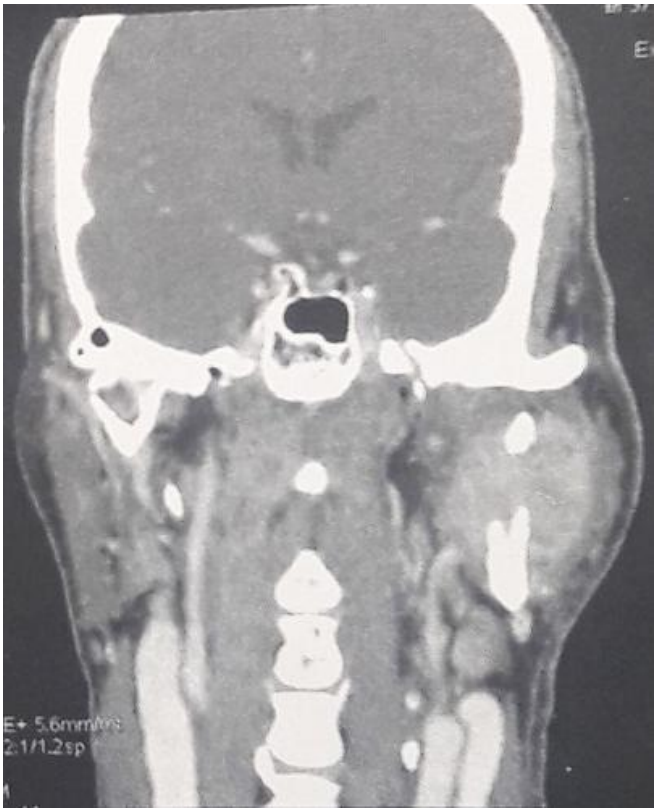

Figure 4: coronal computed tomographic view (soft tissue window) showing a lump surrounding the left ramus with irregular bone destruction and muscle infiltration

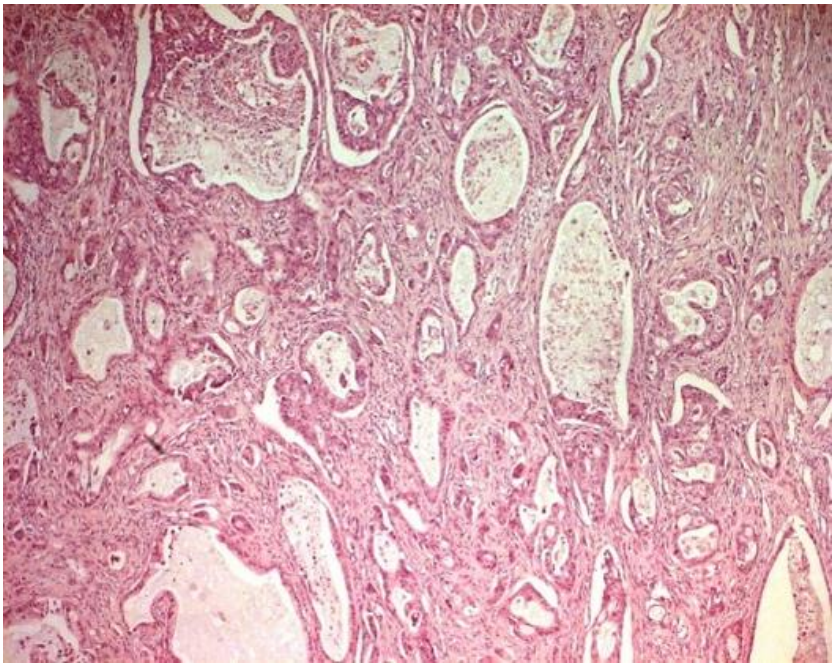

Figure 5: histopathological examination: photomicrography of the biopsy specimen showing an infiltrative tumor composed of large epithelial cell blocks arranged in abortive glandular structures (H&E 40X)

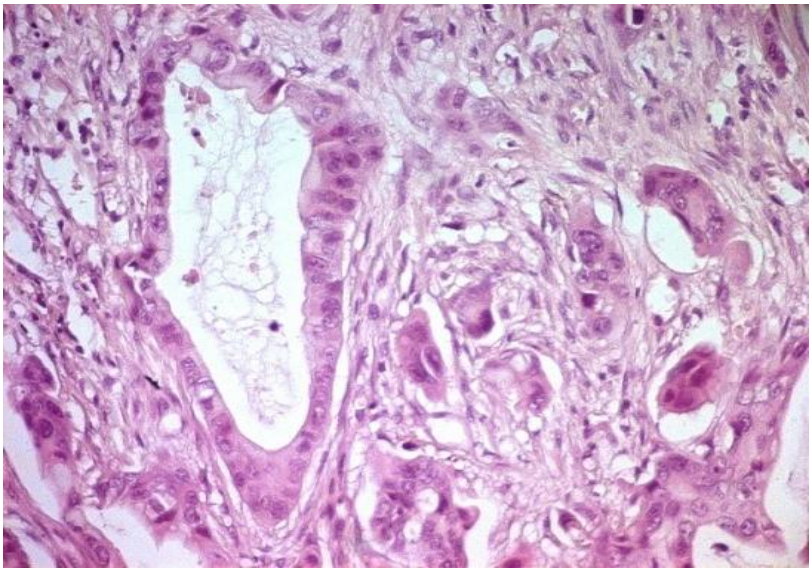

Figure 6: histopathological examination: cells had scant cytoplasm and hyperchromatic nuclei with high mitotic activity (H&E, 200X)

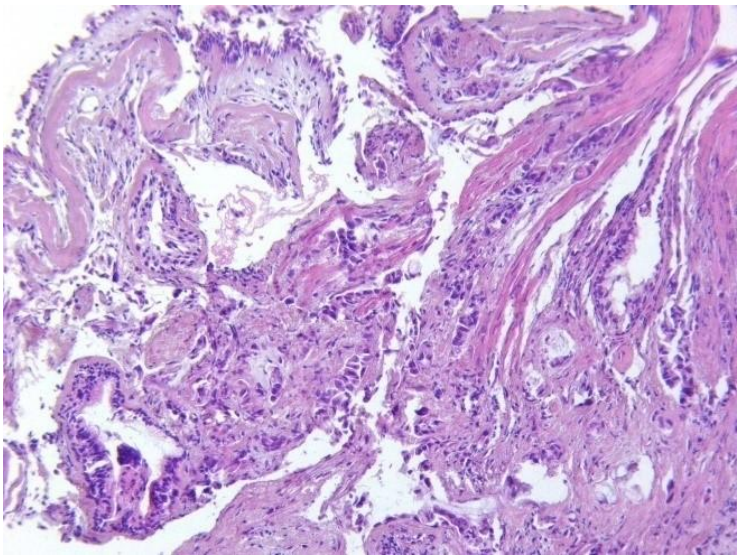

Figure 7: lung biopsy: photomicrography of the biopsy specimen showing large epithelial cell blocks with irregular shapes arranged in glandular patterns (arrow, H&E 100X)