Journal of Oral Science, Vol. 50, No. 1, 95-98, 2008

Case Report

Primary intraosseous adenoid cystic carcinoma of the mandible with lung metastasis: a case report

Thorakkal Shamim, Vengal I. Varghese, Pallikandi M. Shameena and Sivasankar Sudha

Department of Oral Pathology and Microbiology, Government Dental College, Calicut, South India

(Received 4 June and accepted 20 November 2007)

Abstract: Intraosseous adenoid cystic carcinoma is an extremely rare neoplasm with only 17 cases reported previously. A case of primary intraosseous adenoid cystic carcinoma of the mandible with lung metastasis in a 45-year-old man is reported, together with a brief review of the literature. (J. Oral Sci. 50, 95-98, 2008)

Keywords: intraosseous adenoid cystic carcinoma; salivary gland tumor; mandible; lung metastasis.

Introduction

Adenoid cystic carcinoma (ACC) represents 7.5% of all carcinomas and 4% of both benign and malignant epithelial salivary gland tumors (1). It most commonly affects adults, with a peak incidence in the 4th to 6th decades (1). The most commonly involved sites are the parotid gland, submandibular gland and palate, whereas the lower lip, retromolar-tonsillar pillar region and sublingual gland are affected less frequently (1). Very rarely, ACC may arise centrally within the jawbones, usually in the posterior mandible of adults, causing pain due to perineural invasion (2,3). About 40-60% of patients develop distant metastases (lung, bone and soft tissues) despite local control of the tumor (1). The present paper reports a case of primary intraosseous ACC of the mandible with lung metastasis in a 45-year-old man.

Case Report

A male patient aged 45 years presented at Government Dental College, Calicut, with a 2-month history of swelling on the right lower half of the face. Extraoral examination revealed a firm tender swelling measuring 2 × 2 cm below the angle of the mandible. The overlying skin was normal with a smooth surface. The swelling was nonfluctuant and nonpulsatile. Regional lymphadenopathy was not evident.

Intraoral examination revealed a nodular swelling lingual to the right permanent mandibular third molar region (Fig. 1). The overlying mucosa was normal but slightly erythematous. The teeth present in the region were normal (neither carious nor periodontally diseased). Panoramic radiography revealed a radiolucent lesion in the area of teeth 45, 46 and 47 involving the angle (Fig. 2).

The possibility of a malignant tumor was considered on the basis of the clinical findings. Fine-needle aspiration cytology (FNAC) revealed closely packed round to ovoid basaloid cells with hyperchromatic nuclei (Fig. 3). The lesion was surgically excised and the specimen was subjected to histopathological examination. Microscopically the lesion showed a solid pattern with basaloid-type tumor cells arranged in cords and sheets (Figs. 4 and 5). In some areas, islands of epithelial cells containing numerous spherical spaces showing a classical "Swiss cheese" pattern were evident (Fig. 6). Based on the findings of FNAC and conventional microscopy, a diagnosis of intraosseous adenoid cystic carcinoma was made.

The patient was scheduled for further investigations including ultrasonography, CT scan, chest X-ray and bone scan. Chest X-ray revealed a coin lesion in the right lower lung field (Fig. 7). The pulmonary lesion was subjected to percutaneous FNAC and this revealed basaloid cells arranged in sheets (Fig. 8). Bronchoscopic biopsy confirmed the diagnosis of adenoid cystic carcinoma (Fig. 9).

Consequently, the final diagnosis was primary intraosseous adenoid cystic carcinoma of the mandible with lung metastasis. A whole-body scan and bone scan excluded distant metastasis to bone. The patient was referred to the

Correspondence to Dr. Thorakkal Shamim, Shangrila, Parappanangadi-676303, South India
Tel: +91-989-5447351
E-mail: shamu3duad@rediffmail.com

Regional Cancer Center for treatment, but unfortunately he died before the treatment had been completed.

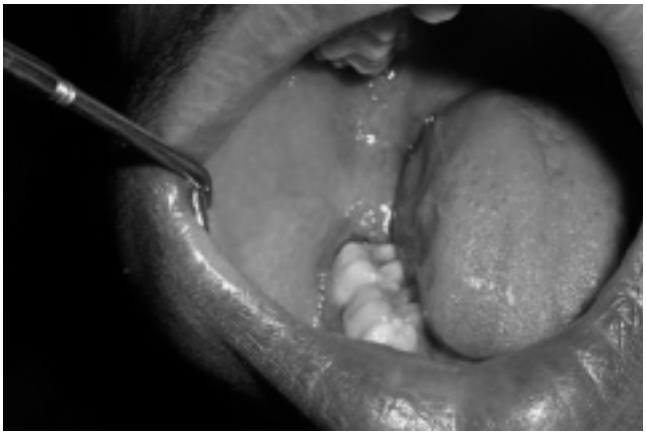

Fig. 1 Intraoral view demonstrating a nodular swelling lingual to the right permanent mandibular third molar region.

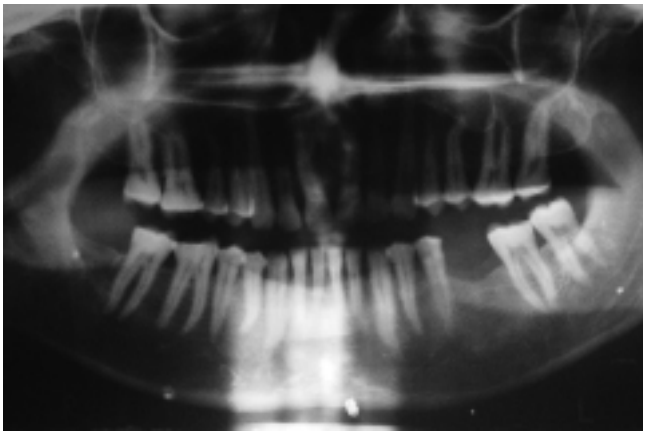

Fig. 2 Panoramic radiograph showing a radiolucent lesion in the region of teeth 45, 46 and 47 involving the angle.

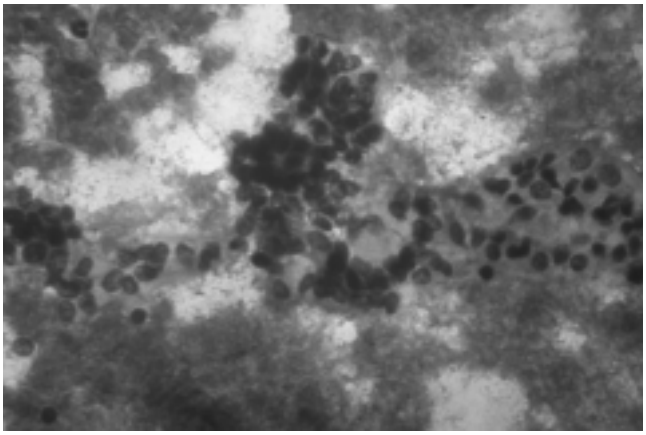

Fig. 3 Fine-needle aspiration cytology of the oral lesion, showing closely packed round to ovoid basaloid cells with hyperchromatic nuclei.

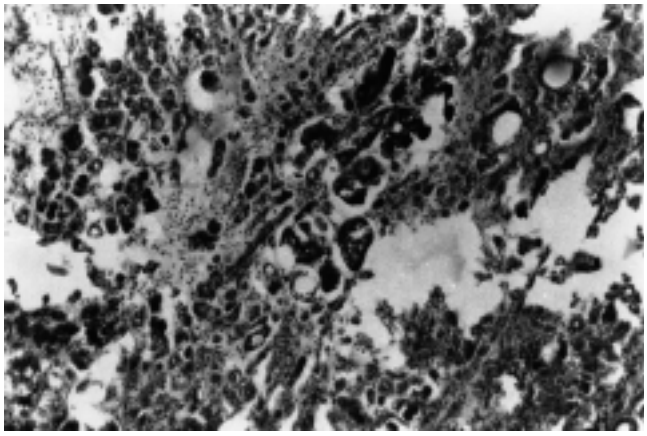

Fig. 4 Photomicrograph showing a solid pattern with basaloid-type tumor cells arranged in cords and sheets (H-E × 10).

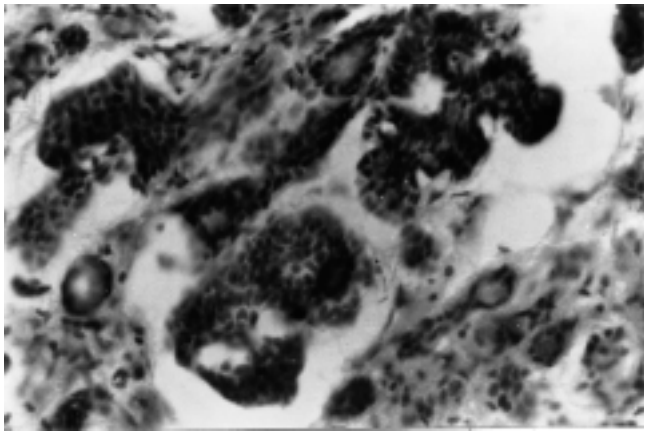

Fig. 5 Photomicrograph showing a solid pattern with basaloid-type tumor cells arranged in cords and sheets (H-E × 40).

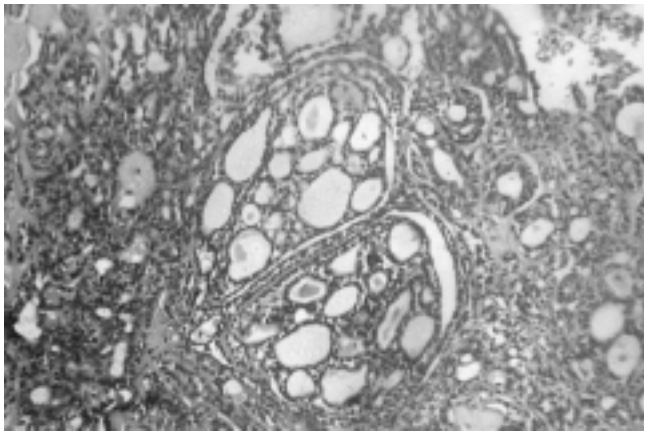

Fig. 6 Photomicrograph showing the classical "Swiss cheese" appearance with a cribriform pattern (H-E × 40).

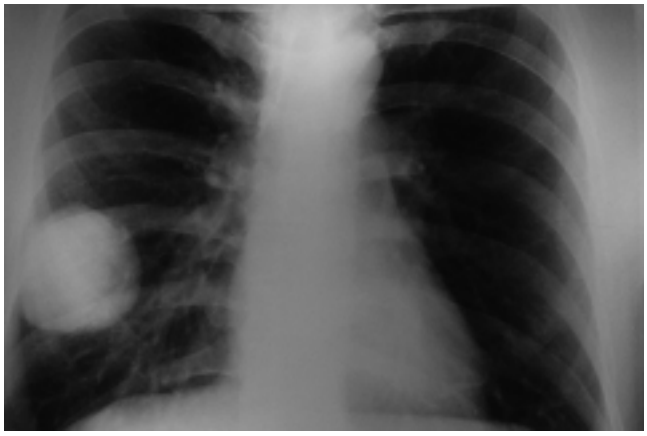

Fig. 7 Chest X-ray view showing a coin lesion in the right lower lung field.

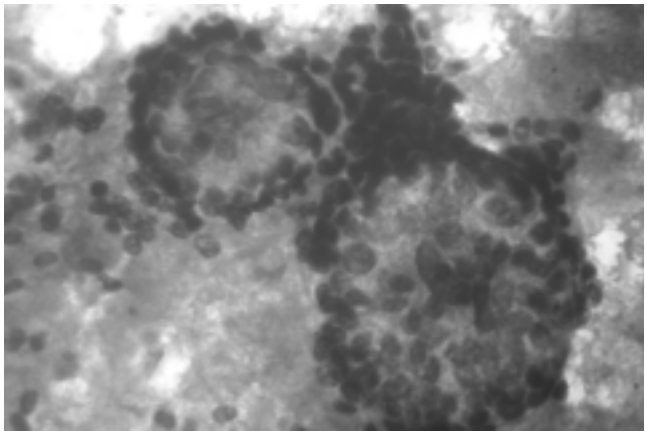

Fig. 8 Fine-needle aspiration cytology of the pulmonary lesion, showing basaloid cells arranged in sheets.

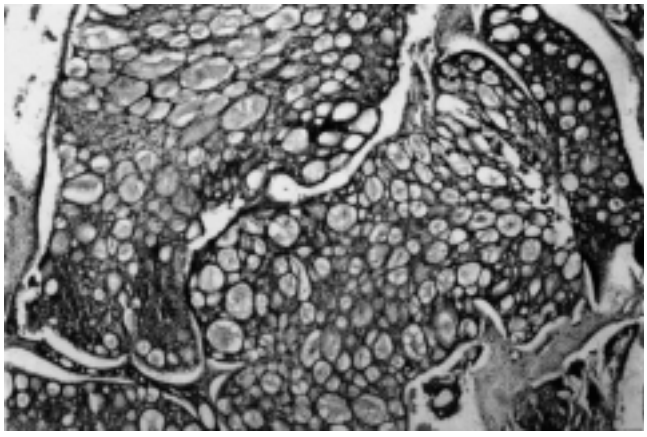

Fig. 9 Microscopy of the pulmonary lesion, showing cuboidal tumor cells arranged in a cribriform pattern (×40).

Discussion

ACC is a malignant epithelial tumor characterized by slow growth, late onset of metastasis and poor prognosis. In the present case, the simultaneous detection of ACC within the mandible and lung suggested possible metastasis from a still undiscovered major salivary gland neoplasm. In view of the fact that the tumor located in the right mandible was larger than that in the lung, we speculate that the former was the primary neoplasm from which subsequent hematogenous metastasis to the right lung developed.

ACC was first described as “cylindroma” by Theodor Billroth in 1856. He studied its histologic features and described the long amorphous compartments as cylinders. Spies in 1930 was the first to use the term adenoid cystic carcinoma (4). In 1943, Dockerty and Mayo emphasized the malignant nature of this tumor (4).

ACC arising centrally within the mandible is extremely rare. Swelling and pain are the most common clinical features (5). The age of affected patients ranges from 24 to 82 years, with no gender predilection (5). The most commonly affected site is the posterior body or angle of the mandible (5). These features are consistent with those of the present case.

If intraosseous ACC is suspected, a multidisciplinary diagnostic approach should always be adopted. The diagnosis of ACC rests on several criteria, including radiographic evidence of osteolysis, presence of intact cortical plates, absence of any primary tumor within the major or minor salivary glands, and histological confirmation of the typical architectural and morphologic features of ACC (6). All these diagnostic criteria were satisfied in the present case.

The histologic features of the present case were confirmed to be those of ACC by both FNAC and conventional histology of the mandible and lung, respectively. Histologically, three patterns of growth have been described (1,7). Typical ACC has a cribriform pattern with nests and columns of cells arranged concentrically. Some have a predominantly tubular pattern and a few others have a solid pattern. The present tumor belonged to the latter category with predominance of a solid pattern with basaloid cells over the cribriform pattern.

The prevalence of cribriform or tubular growth patterns is associated with a better prognosis, in contrast to the presence of more than 50% solid areas, which indicates an aggressive clinical course (6-8). Although it was thought that such histologic grading offers valuable prognostic information, contradictory reports suggest that clinical staging is more reliable for assessing prognosis (9). In the present case, perineural invasion was evident and tumor cells were present in the surgical margins. Both the histopathological and clinical grading was indicative of poor prognosis.

Metastatic tumors in the lung with a primary in the oral cavity will usually present as multiple radiopaque lesions on chest X-ray plates. However, in the present case, we encountered only a single tumor in the right lower lung field, and we inferred that the lesion evident on chest X-ray may have been the initial stage of ACC metastasis to the lung. This was consistent with the findings of Grillet et al. (10).

In conclusion, the present case calls attention to the possible occurrence of primary intraosseous ACC in the mandible with lung metastasis showing a very aggressive course. Since the prognosis of intraosseous ACC is poor, the same clinicohistopathological parameters as those of salivary gland ACC must be applied to intraosseous ACC. Long-term follow-up is mandatory to rule out regional and distant metastases in patients with intraosseous ACC.

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