A CASE OF BRONCHIAL MUCOEPIDERMOID CARCINOMA WITH CUTANEOUS METASTAS: LOW-GRADE HISTOLOGY BUT AGGRESSIVE BEHAVIOUR

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SUMMARY

Mucoepidermoid carcinomas are malignant neoplasms that rarely arise in the tracheobronchial tree. Primary or metastatic cutaneous involvement is unusual. We describe a bronchial mucoepidermoid carcinoma that presented with cutaneous metastases and showed aggressive biological behavior. The microscopic and ultrastructural features of the cutaneous and bronchial tumors were compatible with low-grade mucoepidermoid carcinoma. However, there was widespread metastasis to the skin, subcutaneous and intraabdominal fat tissue, and several bony structures, and the patient died 5 months after the initial diagnosis. We emphasize that benign histological features of primary mucoepidermoid carcinomas are not always associated with low-grade behaviour.

Key Words: Bronchial Carcinoma, Cutaneous Metastases, Mucoepidermoid Carcinoma

ÖZET

Kütanöz Metastazlarla Tanı Alan Bronşiyal Mukoepidermoid Karsinoma Olguğu: Benign Histolojiye rağmen Kötti Klinik Seyir


Anahtar Kelimeler: Bronşiyal Karsinoma, Kütanöz Metastazlar, Mukoepidermoid Karsinoma

Mucoepidermoid carcinomas (MECs) are malignant tumors that usually arise in the salivary glands(1), but occasionally develop in the bronchus(2), esophagus(3), lacrimal glands(4), pancreas(5), thymus(6), and thyroid gland(7). These tumors are composed of mucus-secreting, epidermoid, and intermediate cells arranged in sheets and glandular formations(8-10). The history of MECs has been the subject of much discussion in recent years. In particular, efforts have been made to correlate morphological characteristics with biological behavior. In the most widely accepted approach to predicting clinical aggressiveness and prognosis, MECs are morphologically classified as low-grade and high-grade according to gross, microscopic, and ultrastructural features(8-11).

We report a patient with bronchial MEC who presented with cutaneous metastases. Although the bronchial tumor and the cutaneous lesions exhibited low-grade histologic features, the patient died due to widespread metastases within months of the initial diagnosis.

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Case Report

A 55-year-old man presented with asymptomatic nodules on his face and chest. The lesions had been present for 1 month, and were slowly enlarging. The patient was otherwise in good health, and coronary angioplasty was the only noteworthy item in his medical history. Dermatologic examination revealed a firm erythematous nodule inferior to the left nostril that was 1.3 cm in diameter and had a poorly defined, indurated border (Figure 1). Three other cutaneous nodules ranging from 0.5 cm to 1 cm in diameter were detected on the lateral aspect of the left lip commissure, in the right preauricular region, and on the presternum. Excisional biopsies were performed on one face lesion and the chest lesion. Histological examination of both specimens showed a dermal tumor composed of islands of mucus-secreting, intermediate, and epidermoid cells with no pleomorphism, mitotic figures, or cellular necrosis (Figure 2a). The biopsy tissues stained positive with mucicarmine (Figure 2b), alcian-blue, and periodic acid-Schiff, and also for cytokeratin on immunoperoxidase staining.

Electron-microscopic examination revealed that many of the tumor cells contained large numbers of mucous droplets in their cytoplasm. Other cells showed squamous differentiation and contained cytoplasmic tonofilaments. Scattered intermediate cells were also observed. Based on these findings, the morphologic diagnosis was low-grade MEC.

A chest radiograph was normal, but thoracic computed tomography (CT) revealed a lobulated

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**Figure 1.** Cutaneous metastatic MEC. An erythematous nodule inferior to the left nostril.

**Figure 2.** (a) Histology of a cutaneous metastatic tumor indicating low-grade MEC: Squamoid cells (thin arrow) admixed with rare mucinous cells (thick arrow) (haematoxylin and eosin; original magnification × 230); (b) Intracytoplasmic mucin (arrows) (mucicarmine; original magnification × 230).
soft-tissue mass in the right middle-lobe bronchus. Bronchoscopy showed stenosis of the same bronchus, but no apparent endobronchial tumor. A transbronchoscopic biopsy specimen of the stenotic bronchial wall exhibited similar microscopic features to those noted in the cutaneous lesions (Figure 3a and 3b). Further work-up with abdominal CT revealed multiple nodular lesions in the subcutaneous and intraabdominal fat tissue that were thought to be metastases. Lytic bone lesions in several of the patient’s ribs, the vertebral column, and the right pelvis were also demonstrated on Tc-99m MDP total-body bone scanning. When the diagnosis of metastatic bronchial MEC was established, the patient was started on chemotherapy with cisplatin and gemcitabine. Two months later, the protocol had to be switched to a cisplatin-docetaxel combination due to the appearance of new cutaneous lesions. However, 3 weeks after this change, the patient developed hepatosplenic and cranial metastases, and palliative radiation therapy was applied to the cranium. Despite all treatments, the patient’s condition continued to deteriorate, and he died 5 months after the initial diagnosis.

**Discussion**

MECs are malignant tumors that are composed of mucus-secreting, epidermoid, and intermediate cells(8-10). The history of these neoplasms is interesting. Initially, they were known to occur only in salivary glands, the site that has the highest frequency(12). However, with the first account of bronchial MEC by Smetana et al. in 1952(13) as well as other reports of bronchial involvement(14), it was recognized that these tumors occasionally arise in the tracheobronchial tree. Subsequently, various other sites, including the esophagus(3), thymus(6), and thyroid(7), have also been noted in rare cases. The skin has also been reported as a primary site of MEC(15,16) but this is extremely unusual. MECs presenting with cutaneous metastases are rare, with only three cases published to date (9,11,17). The first of these was reported by Metcalf et al(11). in 1986, and was a bronchial MEC that had metastasized to the skin. The other two cases presented with cutaneous metastases, originated from the sublingual gland(17) and parotid gland(9), respectively. Early reports of MECs indicated that these tumors were essentially benign neoplasms(14,18). However, later studies documented a malignant form that tended to produce widespread metastasis. As mentioned above, in attempt to predict malignant potential from histologic appearance, a classification system was proposed that divided MECs into low-grade and high-grade types according to their histological and ultrastructural characteristics(2,10). Low-grade MECs show minimal pleomorphism, rare mitoses, and minimal or no necrosis. The metastatic potential of this type is reported to be extremely

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**Figure 3.** (a) A group of tumor cells (arrow) in the bronchoscopic biopsy specimen (haematoxylin and eosin; original magnification x 45); (b) Higher magnification reveals mucinous (thick arrow) and intermediate cells (thin arrow) (haematoxylin and eosin; original magnification x 230).
low, and death due to this form is very unusual. In contrast, the histologic features of high-grade MECs are marked nuclear pleomorphism, increased mitotic rate, and cellular necrosis. Widespread metastasis is common with this type(19), and the histology of the metastases may indicate even more aggressive malignant features than the primary tumor(8).

Although this classification system is accurate in the majority of MEC cases, a few reports have noted low-grade histologic features in association with fatal outcome. Barsky and colleagues(8) reported a case of bronchial MEC in which the primary tumor showed low-grade histologic features but exhibited aggressive biological behavior. The findings were similar in a case described by Metcalf et al(11), that detailed a bronchial MEC presenting with cutaneous metastases. The morphologic features of our patient’s tumor on light- and electron microscopy were compatible with low-grade MEC. However, the clinical course, namely, death due to widespread metastases, was obviously that associated with high-grade tumors.

It is true that some high-grade MECs contain areas of low-grade growth, and that these areas may be missed if the tumor is not sufficiently sampled. In our case, total excision of the bronchial tumor was not performed due to widespread metastasis. Because of this, we were unable to investigate the primary tumor extensively. However, the features of the transbronchial biopsy specimen indicated low-grade MEC, as did the histologic examination of the two cutaneous (face and chest) lesions that were sectioned extensively. Furthermore, in cases of mixed low- and high-grade MEC, the metastases are expected to be less differentiated (higher grade) than the original tumor(8).

To the best of our knowledge, this is only the second reported case of a MEC diagnosed by cutaneous metastases that behaved aggressively despite having low-grade morphology. Although classification of MECs based on the histologic features has high prognostic value in the majority of cases, it does not always accurately predict the potential for metastases.
REFERENCES

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