

Lung preservation in mucoepidermoid carcinoma of tracheobronchial tree: A case series

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ABSTRACT

Introduction: Mucoepidermoid carcinoma (MEC) is a primary salivary gland tumor also arising from nonsalivary gland organs of the body such as submucosal glands of tracheobronchial tree. Surgical resection with negative margins is the treatment of choice. All efforts should be made to preserve as much lung parenchyma as possible, by various bronchoplastic procedures. We present our experience with mucoepidermoid tumors and review their management options including lung preservation techniques and outcome of surgery. **Materials and Methods:** This is a retrospective analysis of prospectively maintained data of 14 patients who underwent surgery for MEC. Their demographic data; clinical presentation; and preoperative, intraoperative, and postoperative details were recorded. All patients underwent contrast-enhanced computed tomography of chest and bronchoscopy as part of workup for diagnosis and to assess the location, size, and extent of tumor; extraluminal component; and status of distal lung parenchyma. **Results:** There were eight male and six female patients. The median age at the time of surgery was 28.36 years (range 22–45 years). The procedures performed included right upper lobectomy and right pneumonectomy in one patient each, left main bronchus sleeve resection in six patients, left upper sleeve lobectomy in three patients, and carinal resection and reconstruction of neo carina in three patients. Twelve (85.7%) of our patients underwent lung-preserving surgery. The median hospital stay and chest tube removal duration was 4 and 3 days, respectively. The median tumor size was 1.91 cm (range 1–8 cm). The median follow-up was 24 months (ranging from 6 to 78 months). **Conclusion:** Radical surgery to achieve R “0” resection with aggressive emphasis on lung preservation is the mainstay of treatment of MEC. Greater awareness of these tumors is necessary to avoid misdiagnosis and to prevent delaying of potential complete resection of MEC.

KEY WORDS: Lung preservation techniques, mucoepidermoid carcinoma, primary salivary gland tumor

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INTRODUCTION

Mucoepidermoid carcinoma (MEC) is a primary salivary gland tumor which can also arise from nonsalivary gland organs of the body such as submucosal glands of tracheobronchial tree and present with dyspnea, cough, and hemoptysis.^[1,2] Due to an overlap between these symptoms and those of asthma or tuberculosis, these

tumors are often misdiagnosed initially and treated conservatively. Surgical resection with negative margins is the treatment of choice. Because these tumors are mostly central and endobronchial in origin, efforts should be made to preserve as much lung parenchyma as possible, by various bronchoplastic procedures. We present our

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experience with mucoepidermoid tumors and review their management options including lung preservation techniques and outcome of surgery.

MATERIALS AND METHODS

This is a retrospective analysis of prospectively maintained data of patients who underwent surgery for MEC at a tertiary care center from March 2013 to September 2019. A total of 14 patients who underwent surgery for MEC were included in the study. Their demographic data; clinical presentation; and preoperative, intraoperative, and postoperative details were recorded. All patients underwent contrast-enhanced computed tomography (CECT) of chest as part of workup for diagnosis and to assess the location, size, and extent of tumor; extraluminal component; and status of distal lung parenchyma. Preoperative bronchoscopy was done in all the patients to assess the exact location and extent of tumor and take a biopsy. Histopathological diagnosis was achieved preoperatively in all the patients.

Surgical details

All the procedures were done under general anesthesia with lung isolation achieved by a double-lumen tube or an adequate-sized single lumen placed in the healthy bronchus under bronchoscopic guidance. All the 14 patients underwent posterolateral thoracotomy. Thereafter, lung parenchyma was assessed carefully. The procedure performed was guided by the location and extent of the tumor and the condition of the distal lung parenchyma. If lung parenchyma was completely destroyed, pneumonectomy was performed. In patients with healthy distal lung parenchyma and tumor in the main bronchus, bronchial sleeve resection was performed, after frozen section confirmation of the negative margins. In patients with tumor in the lobar bronchus, lobectomy and if it was extending proximally and involving secondary carina, a sleeve lobectomy was performed. If the tumor involved carina, carinal resection and neo carina formation was performed.

Basic surgical principles followed in all patients included minimal circumferential dissection, no energy source while dissecting near the bronchus, minimal tissue handling, and a tension-free anastomosis (with 3 0 PDS suture), maintaining correct orientation of the bronchial ends. Standard mediastinal lymphadenectomy was performed in all the cases. Bronchial stump of pneumonectomy was reinforced with pericardial fat/intercostal muscle flap. Air leak test of the bronchial stump was always done. Two chest tubes were placed in patients who underwent lung preservation surgery, one at the apex and another at the base and these were connected to Thopaz™ digital negative suction device. A single chest tube was placed in patients who underwent pneumonectomy. Patients who underwent bronchial sleeve resection had bronchoscopy performed on the table prior to extubating to confirm luminal patency. All the patients were extubated on the table, shifted to recovery room for overnight monitoring, and transferred to the ward the next morning.

Continuous epidural analgesia was administered through a pain pump for up to 3 days postoperatively for adequate pain relief. This ensured excellent patient cooperation for aggressive chest physiotherapy to maintain complete lung expansion. Chest tubes were removed when there was no air leak, and the drainage was nonpurulent/nonhemorrhagic and <100 mL in 24 h with complete lung expansion. In pneumonectomy patients, drain was removed on the 2nd day. Prolonged postoperative air leak (>7 days), chest tube duration, wound infection and other complications, and duration of hospital stay were analyzed. All patients were followed up in the outpatient department at weekly intervals for 4 weeks and then every 6th month for the first 2 years and then yearly for up to 5 years. A chest X-ray was done at 1 month and at all subsequent visits. All patients underwent check bronchoscopy at 6 months and then yearly for 2 years to rule out any recurrence.

Statistical methods

Statistical testing was conducted with the Statistical Package for the Social Science system version SPSS 17.0 (Statistical Package for the Social Sciences [SPSS]). Continuous variables were presented as mean ± standard deviation or median (interquartile range). Categorical variables were expressed as frequencies and percentages.

RESULTS

Patients who underwent surgery for MEC during the study period were included in the analysis. Demographic variables are shown in Table 1. There were eight male and six female patients. The median age at the time of surgery was 28.36 years (range: 22–45 years). Four patients had a history of smoking. The most common presentation was hemoptysis ($n = 6$, 42.85%) and dyspnea in five (35.7%), followed by chest pain and cough, which were present in three (21.42%) patients. Fiberoptic bronchoscopy was

Table 1: Demography and peri-operative variables

Variable	Frequency
Male	8
Female	6
Mean age	28.5
Symptoms	
Hemoptysis	6
Dyspnea	5
Cough	3
Comorbidities	
Diabetes mellitus	1
Others (CAD, HTN, and AF)	1
Surgical demographics (%)	
Left main bronchus sleeve resection	6 (42.85)
Left upper sleeve lobectomy	3 (21.42)
Right upper lobectomy	1 (7.1)
Right pneumonectomy	1 (7.1)
Carinal resection	3 (21.42)
Mean hospital stay time	4 days
Mean ICD duration	3 days
Tumor size	1.91

ICD: Intercostal Drainage, CAD: Coronary artery disease, AF: Atrial fibrillation, HTN: Hypertension

done in all patients, which showed growth in the left main bronchus in six patients, involvement of lower trachea and carina in three patients, tumor in the left upper lobe bronchus in three patients, and tumor in the right upper lobe bronchus and right main bronchus in one each patient. Biopsy was reported as MEC in all patients. One patient had presented with an acute central airway obstruction and underwent rigid bronchoscopic debulking to relieve symptoms as well as to delineate the true extent of the tumor. The procedures performed included right upper lobectomy and right pneumonectomy in one patient each, left main bronchus sleeve resection in six patients, left upper sleeve lobectomy in three patients, and carinal resection and reconstruction of neo carina in three patients. On-table frozen section was done in all patients to confirm negative resection margins. In patients with sleeve resection and sleeve lobectomy, on-table bronchoscopy was done to check the patency of the lumen prior to extubating. The median hospital stay and chest tube removal duration was 4 and 3 days, respectively. The postoperative recovery of these patients was uneventful. Histopathological examination of the resected specimen confirmed low-grade muco-epidermoid carcinoma in all patients. The median tumor size was 1.91 cm (range: 1–8 cm), and the median lymph node yield was 18 (range: 14–21). None of the resected lymph nodes was positive for cancer. Patients were followed up regularly by bronchoscopy at 6-monthly interval and then yearly till 2 years and then by chest X-ray for 5 years. There was no recurrence during follow-up. The median follow-up was 24 months (ranging from 6 to 78 months). All patients are alive and healthy on the last follow-up.

DISCUSSION

MEC has been classified as primary salivary gland tumor.^[3] These tumors constitute <1% of primary bronchial malignancy.^[4] MEC of the trachea and bronchi is classified as low grade or high grade based on nuclear pleomorphism, mitotic activity, and the presence or absence of necrosis.^[5] All cases in our series had low-grade MEC, which is an important favorable prognostic factor.

These tumors can occur at any age but are usually seen in middle age group. The median age of presentation in our study was 28 years (range: 22–45 years). Most of the studies report a slight male preponderance, which was observed in our study too.^[6]

No preferred site for occurrence of these tumors has been reported, though these are mostly present in the central airway.^[7] In our study too, the tumors mostly involved lobar bronchus, main bronchus, or carina. Most of the patients had only endobronchial component, with only one patient having an extrabronchial extension too.

Tumors present in the main airway cause obstructive symptoms such as cough, collapse of distal lung, dyspnea, and hemoptysis. Due to these symptoms, these patients are often misdiagnosed and treated as asthma or tuberculosis.

In our series, most patients presented with hemoptysis, cough, and dyspnea and one patient had complete collapse of the distal lung. Seven of our patients had received antitubercular treatment for varying durations before being diagnosed as endobronchial tumor. It requires a high index of suspicion to make a correct diagnosis in these cases. Persistent and progressive symptoms should alert a clinician to the possibility of such a tumor.

CECT thorax is important for diagnosis as well as defining the exact site and extent of tumor and the condition of the lung parenchyma. Bronchoscopy is always required which re-confirms the exact site and extent of tumor and allows biopsy to be taken for a definitive diagnosis, which helps in deciding the extent of surgical resection.^[8] Sometimes, these patients present with central airway obstruction and need immediate management, as was the case with one of our patient who underwent urgent rigid bronchoscopy and coring of tumor and later went for carinal resection and neo carina formation for achieving resection with clear margins.

Standard treatment of these tumors is complete surgical resection with negative margins with preservation of lung parenchyma, wherever possible. Chemotherapy and radiotherapy have not proved to be of much benefit.^[9] Every effort should be made to avoid pneumonectomy. However, it is unavoidable if the lung parenchyma is destroyed, as was the case in one of our patients. This happens in patients with large central tumors, left untreated for long time with collapse of the lung. The importance of high index of suspicion, early diagnosis, and timely referral for surgery cannot be overemphasized to avoid this eventuality. Irrespective of the kind of resection performed, it is vital to achieve microscopically clear margins, an important factor in reducing recurrence and enhancing survival. None of our patients had histologically positive margins and no recurrence during follow-up.

The term “lung-preserving surgery” does not have a standard definition and has been variously defined in literature. In this article, we have defined it as a procedure wherein the complete lung or a lobe could be saved as a result of complex bronchoplastic procedures such as bronchial sleeve resection, sleeve lobectomy, or carinal resection and reconstruction. By this definition, 12 (85.7%) of our patients underwent lung-preserving surgery. Their tumors were central in location and involved the main bronchus and major lobar bronchus with involvement of secondary carina or the main carina. Most of them had been offered pneumonectomy at their initial treatment centers, as those centers could not perform complex bronchoplastic procedures. Experience at these techniques allowed us to do lung preservation in these 12 patients. Thorough preoperative evaluation for exact location and size of the tumor and meticulous intraoperative lung preservation protocols learned and developed over years, help us to perform these complex procedures.

Table 2: Major surgical series on mucoepidermoid carcinoma

Author	Duration	Number of patients	Types of surgery (%)	Tumor size	Lymph node positivity (%)	Central location (%)
Molina <i>et al.</i> ^[7] Total n=62 MEC - 20	1972-2002	20	Tracheal resection (44.2) Lobectomy (25.6) Sleeve lobectomy (9.3) Pneumonectomy (18.6)	1.9	11.8	44.4
Kang <i>et al.</i> ^[4] Total: n=48 MEC - 26	1995-2009	26	Tracheal resection (16.7) Sleeve lobectomy (27.1) Bilobectomy (31.2) Pneumonectomy (14.6) Carinal resection (10.4)	3.5	19	37.5
Zhu <i>et al.</i> ^[9] Total: n=88 MEC - 69	2001-2013	69	Lobectomy (55.7) Sleeve lobectomy (18.2) Pneumonectomy (26.1)	2.65	12	35.9
Pandey <i>et al.</i> ^[13] Total: n=7 MEC - 3	2012-2014	3	Pneumonectomy (66.6) Lobectomy (33.3)	3.5	0	100
Current series MEC -w 14	2012-2019	14	Lobectomy (7.1) Sleeve lobectomy (21.4) Main bronchial sleeve (42.85) Carinal resection (21.42) Pneumonectomy (7.1)	1.91	0	100

MEC: Mucoepidermoid carcinoma

Prognosis of these patients depends on the grade of the tumor, completeness of resection, and status of lymph nodes.^[10] Garg *et al.* stated in their study that lymph node involvement is to the tune of 20% in these patients and emphasized the role of mediastinal lymph node dissection/sampling in all patients.^[11] We routinely perform complete mediastinal lymph node dissection in all cases as it is an important prognostic factor in the long-term survival of these patients. Our average lymph node yield was 18. All lymph nodes were free of tumor.

We routinely practice the principles of enhanced recovery after surgery (ERAS). These included preoperative counseling and nutritional screening and correction, smoking cessation, prehabilitation for high-risk patients, avoidance of long period of fasting and preoperative sedatives, venous thromboembolism prophylaxis, prevention of intraoperative hypothermia, use of short-acting anesthetic agents to facilitate early recovery from anesthesia, euvolemic fluid management during and after surgery, aggressive control of nausea and vomiting, aggressive postoperative analgesia by routine use of epidural catheters with continuous infusion through patient-controlled pain pump, same-day removal of urinary catheters, early mobilization after surgery, and early removal of chest drains.^[12] By uniform and strict adherence to these principles, we were able to keep a mean hospital stay of 4 days even after such major resections.

The major series on MEC in English literature are few and are listed in Table 2. The present series is the largest from India, with lung preservation achieved in majority of the patients. We wish to reiterate the importance of a high index of suspicion for early diagnosis, accurate anatomical localization by bronchoscopy and CECT chest, and early surgery at a center experienced in complex bronchoplastic procedures. Lung-preserving surgery with histologically negative margins will provide excellent long-term survival.

Practice of ERAS principles will allow early discharge and quick return to normal life.

CONCLUSION

Radical surgery to achieve R “0” resection with aggressive emphasis on lung preservation is the mainstay of treatment of MEC. Greater awareness of these tumors is necessary to avoid misdiagnosis and to prevent delaying of potential complete resection of MEC.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Leonardi HK, Jung-Legg Y, Legg MA, Neptune WB. Tracheobronchial mucoepidermoid carcinoma. Clinicopathological features and results of treatment. *J Thorac Cardiovasc Surg* 1978;76:431-8.
- Falk N, Weissferdt A, Kalhor N, Moran CA. Primary pulmonary salivary gland-type tumors: A review and update. *Adv Anat Pathol* 2016;23:13-23.
- Stewart FW, Foote FW Jr., Becker WF. Muco-epidermoid tumors of salivary glands. *Ann Surg* 1945;122:820-44.
- Kang DY, Yoon YS, Kim HK, Choi YS, Kim K, Shim YM, et al. Primary salivary gland-type lung cancer: Surgical outcomes. *Lung Cancer* 2011;72:250-4.
- Brandwein MS, Ivanov K, Wallace DI, Hille JJ, Wang B, Fahmy A, et al. Mucoepidermoid carcinoma: A clinicopathologic study of 80 patients with special reference to histological grading. *Am J Surg Pathol* 2001;25:835-45.
- Yousem SA, Hochholzer L. Mucoepidermoid tumors of the lung. *Cancer* 1987;60:1346-52.
- Molina JR, Aubry MC, Lewis JE, Wampfler JA, Williams BA, Midthun DE, et al. Primary salivary gland-type lung cancer: Spectrum of clinical presentation, histopathologic and prognostic factors. *Cancer* 2007;110:2253-9.
- Li X, Zhang W, Wu X, Sun C, Chen M, Zeng Q. Mucoepidermoid carcinoma of the lung: Common findings and unusual appearances on

- CT. Clin Imaging 2012;36:8-13.
9. Zhu F, Liu Z, Hou Y, He D, Ge X, Bai C, *et al.* Primary salivary gland-type lung cancer: Clinicopathological analysis of 88 cases from China. J Thorac Oncol 2013;8:1578-84.
 10. Chin CH, Huang CC, Lin MC, Chao TY, Liu SF. Prognostic factors of tracheobronchial mucoepidermoid carcinoma-15 years experience. Respirology 2008;13:275-80.
 11. Garg PK, Sharma G, Rai S, Jakhetiya A. Primary salivary gland-type tumors of the lung: A systematic review and pooled analysis. Lung India 2019;36:118-22.
 12. Batchelor TJ, Rasburn NJ, Abdelnour-Berchtold E, Brunelli A, Cerfolio RJ, Gonzalez M, *et al.* Guidelines for enhanced recovery after lung surgery: Recommendations of the enhanced recovery after surgery (ERAS[®]) society and the European society of thoracic surgeons (ESTS). Eur J Cardiothorac Surg 2019;55:91-115.
 13. Pandey D, Garg PK, Jakhetiya A, Pandey R, Bhorawal S, Nath D, *et al.* Surgical experience of primary salivary gland tumors of lung: A case series. Int J Surg 2015;21:92-6.