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Original research

Surgical experience of primary salivary gland tumors of lung: A case series

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HIGHLIGHTS

- Primary salivary gland type tumors of the lung are rare neoplasms.
- Mucoepidermoid and adenoid cystic cancer are common histological subtypes.
- Surgery is the main stay of treatment.
- Regional node metastasis is rare.
- Complete surgical resection results in excellent overall survival.

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ABSTRACT

Introduction: Primary salivary gland type tumors of lung (PSGTTL) are rare intra-thoracic malignant neoplasm. Their description in literature is largely limited to a few case series/case reports. We herewith present our surgical experience of and review its clinical presentation, management options and survival outcomes.

Methods: This retrospective analysis of prospectively maintained computerized data-base of patients was conducted in a tertiary teaching oncology centre in North India. The case records of all the patients who underwent surgery for PSGTTL were reviewed. Details concerning the clinical presentation, pre-operative therapy, operative procedure, histopathological examination, postoperative complications and outcome were retrieved from the case records.

Results: There were seven patients who underwent surgery for PSGTTL during the period from January 2012 to December 2014. Hemoptysis (n = 6, 85.7%) and dyspnoea (n = 6, 85.7%) were common presenting clinical features. Fiber-optic bronchoscopy revealed endobronchial growth in all patients – five patients had growth in left main bronchus while one each had growth in right main bronchus and right intermediate bronchus. Biopsy confirmed adenoid cystic carcinoma in 4 (57.1%) and muco-epidermoid carcinoma in 3 (42.9%) patients. All but one had R'0' resection – pneumonectomy in five and bilobectomy in one patient; one patient was found to be unresectable in view of dense adhesions between lung and heart. Median pathological tumor size was 3.5 cm; none of the patient was found to have metastatic spread to lymph nodes. Overall, six patients are alive after a median follow up of 5 months (range 1–30).

Conclusion: Radical surgery to achieve R'0' resection is the main stay of treatment of PSGTTL to achieve prolonged survival.

1. Introduction

Salivary type neoplasms are known to occur at multiple organ sites in view of basic structural homology among exocrine glands in these anatomic sites [1]. Primary salivary gland type tumors of the

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lung (PSGTTL) are rare neoplasms; they account for less than 1% of all lung tumors [1]. They are histologically not indifferent from their counterparts of salivary origin; they are thought to arise from the sub-mucosal glands of the trachea-bronchial tree [2]. Majority of them is either adenoid cystic or mucoepidermoid carcinoma; rare types are epithelial-myoepithelial carcinoma, acinic cell carcinoma, and mixed tumors [1,3]. PSGTTL are slow growing indolent tumors and surgery is the main stay of treatment; there is limited role of chemoradiotherapy in the management of PSGTTL. Clinical importance of PSGTTL lies in the fact that their complete surgical resection R'0' resection) usually results in prolonged survival, unlike how the conventional pulmonary carcinomas behave. A greater awareness of PSGTTL is essential for accurate diagnosis and proper clinical management. We herewith present our experience of seven patients of PSGTTL and review its clinical presentation, management options and survival outcomes.

2. Materials and methods

2.1. Study design and settings

The study was a retrospective analysis of a prospectively maintained computerized data-base of patients who underwent oncological surgery at a tertiary teaching oncology centre in North India.

2.2. Study population

All the consecutive patients of intra-thoracic neoplasms who underwent oncological surgery for histopathologically confirmed PSGTTL between 1st of January 2012 to 31st December 2014. Details concerning the clinical presentation, management, histopathological diagnosis and follow-up of these patients were retrieved from the prospectively-filled case-records, and were analyzed using SPSS version 16.

2.3. Literature review

A pubmed search using MESH words "Salivary Gland Neoplasms/pathology"[Majr] and "Lung Neoplasms"[Mesh] was conducted on 08th February, 2015. A review of the case studies of PSGTTL was done to assess clinical profile, management options and survival outcomes.

3. Result

104 patients of lung cancer were operated during the period between January 2012 to December 2014. There were seven patients who underwent surgery for primary salivary gland type tumors of lung. Median age of the patients was 42.0 years (range 27–52); all patients were male. All of them admitted to be smokers. Hemoptysis (n = 6, 85.7%) and dyspnoea (n = 6, 85.7%) were common presenting clinical features, while chest pain and cough were present in 3 (42.8%) patients each. Four patients reported to have received anti-tubercular treatment for their symptoms. Fiberoptic bronchoscopy revealed endobronchial growth in all patients – five patients had growth in left main bronchus while one each had growth in right main bronchus and right intermediate bronchus. Biopsy confirmed adenoid cystic carcinoma in 4 (57.1%) and mucoepidermoid carcinoma in 3 (42.9%) patients. Two patients underwent rigid bronchoscopic debulking to relieve airway obstruction and to delineate the true extent of tumor. All but one had R'0' resection – pneumonectomy in five and bilobectomy in one patient; one patient was found to be unresectable in view of dense adhesions between lung and heart. He was given radical radiotherapy; he has been undergoing repeated bronchoscopic debulking of residual tumor. All the patients but one had uneventful postoperative recovery. One patient who had left carinal pneumonectomy developed pneumonia in postoperative period and succumbed to it. The other patient who could not undergo R'0' resection has been having repeated bronchoscopic mechanical debulking; he is alive and symptom free after 17 months of follow up. Histopathological examination of the resected specimen in the operable patients confirmed the preoperative diagnosis. Figs. 1 and 2 displays the microscopic pictures of MEC and ACC respectively. Median pathological tumor size was 3.5 cm (range 1.8–7 cm) and median number of harvested lymph nodes were 9 (range 4–18 cm); though none of the node in any patient was found to be metastatic. Overall, six patients are alive after a median follow up of 5 months (range 1–30). None of them were found to have salivary gland tumors at any other site in the follow-up evaluation.

A pubmed search using MESH words "Salivary Gland Neoplasms/pathology"[Majr] and "Lung Neoplasms"[Mesh] yielded 138 articles. Majority of the PSGTTL are either MEC or ACC; we focused our review on these histological types. Table 1 represents clinic-pathological characteristics of patients our series and previously published series; Table 2 displays the treatment and survival outcomes of these patients.

4. Discussion

Though different histological types of PSGTTL are known, MEC and ACC are the common types. In our series, they were almost equally distributed. Zhu et al. [4], in their experience of 88 cases, found MEC in 78.4%, ACC in 13.6% and EMC in 7.9%; while Moline et al. [2] reported ACC in 64.5% and MEC in 32.3% in their experience of 62 patients of PSGTTL. Kang et al. [5], in their experience of 48 cases of PSGTTL, found MEC in 54.1%, ACC in 41.7% and EMC in 4.2% Other published series focused on a particular histological type.

Though PSGTTL usually involve a wide age range from 3 years to 78 years, they mainly affect people in their middle age. Median age of our patients was 42 years. Previously published case series have also reported the similar age predilections. Kang et al. [5] and Molina et al. [2] showed median age of involvement in MEC was younger age group than ACC, while Zhu et al. [4] did not find any difference. Though all of our patients were male, previously reported case series reveal a conflicting gender predilection for PSGTTL. Zhu et al. [4], Molina et al. [2], and Maziak et al. [6] could not find any gender predilection for PSGTTL while Moran et al. [7], Mondal et al. [8] and Kang et al. [5] reported male predilection.

All of our patients had endobronchial growth in central airways. However, literature is not uniform regarding the location of PSGTTL: central airway involvement varies from 6.2% to 100% [2,4–8]. Centrally located endobronchial tumors cause obstructive symptoms like cough, dyspnea, hemoptysis and peripheral lung collapse. In tuberculosis endemic country like ours, these symptoms may be attributed to tuberculosis and the patients may be prescribed anti-tuberculosis treatment erroneously. More than half of our patients had received anti-tuberculosis treatment before they came to our hospital. One of our patients had received taxol-platinum based chemotherapy in view of T4 lesion in another hospital where bronchoscopy biopsy was misdiagnosed as adenocarcinoma. Greater awareness of PSGTTL is required to diagnose these lesions as complete resection of these tumors, even locally advanced, result in good survival outcomes in contrast to epithelial pulmonary neoplasms.

Previous literature clearly suggests that complete surgical resection (R'0' resection) provides the best chance of long term survival. Though attempts must be made to preserve maximum