

# SURGICAL OUTCOME OF MYCETOMA: ELEVEN YEARS EXPERIENCE IN A HIGH VOLUME CENTER

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## ABSTRACT

**OBJECTIVE:** to know the demographics, clinical presentation, underlying lung disease, surgical procedure performed and its perioperative outcome in patients with Mycetoma.

**METHODOLOGY:** This retrospective study was carried out in the department of thoracic surgery Lady Reading Hospital Peshawar from June 2002 to June 2013. Computerized medical records of patients operated upon for Mycetoma, were retrieved and reviewed for demographics, clinical presentation, underlying lung disease, investigations performed, operative procedure & outcome.

**RESULTS:** Out of 294 total patients, 176 (60%) were males and 118 (40%) females with a mean age of  $38.4 \pm 11.30$  years (range 14 to 71 years). Hemoptysis was present in 92% patients while Tuberculosis was the most frequent underlying lung disease (92.5%). Lobectomy, bi-lobectomy, wedge resection and pneumonectomy were performed in 86.3%, 2.3%, 5.4% and 4% patients respectively. Cavernostomy was performed in 5 patients (1.7%). Supplementary procedure (thoracoplasty or myoplasty) was performed in 5.1% patients. The operative time averaged  $91 \pm 9.37$  minutes (range 78-190 minutes). Operative mortality was 2.3% while morbidity was 14.6% including hemorrhage (4.4%), pleural space problems (6.4%) and others (3.7%).

**CONCLUSION:** Mycetoma is not uncommon in our set up and occurs more frequently in young male patients having underlying tuberculosis. Lobectomy was the commonest surgical procedure for Mycetoma in our set up. Surgery can be performed safely though with increased morbidity including hemorrhage and pleural space problems. Operative mortality rate was 2.3%.

**KEY WORDS:** Mycetoma, Aspergilloma, Lobectomy, Thoracoplasty, Air leak, Bronchopleural fistula.

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Mycetoma or Aspergilloma is the formation of fungal ball when fungi colonize the pre-existing pulmonary cavities of any etiology<sup>5,6</sup>; the term was coined by Deve in 1938<sup>7</sup>. There is no effective medical treatment<sup>8,9</sup> and surgery is the mainstay of treatment<sup>8-12</sup>. The most common symptom is hemoptysis<sup>13,14</sup> which may be of varying degrees and because of fear of life threatening fatal hemoptysis<sup>11,15</sup>, surgery is advised in all patients.

The operation for Aspergillosis is technically challenging<sup>8-12</sup> because of dense pleural symphysis with thickened pleura and secondary blood supply from chest wall; and adhesions & inflammatory changes with lymphadenopathy in & around hilum. As mycetoma forms in previously diseased lungs, the reduced compliance of the remaining lung adds to complexity of procedure (gravity of outcome). Perioperative morbidity as well as mortality is high<sup>8-12</sup> concerning which some controversy still exists regarding routine surgery for mycetoma. All these facts are addressed in multiple publications in international literature in past 30 years while nationally there is a paucity of literature on this topic. Only one publication could be found on this topic in Pakmedinet, which was published by Ahmad et al in 2010.<sup>16</sup> So there is a strong need to study the disease in our population, the response to surgery and perioperative outcome.

The purpose of the present study was to review the demographics, clinical pre-

## INTRODUCTION

Pier Antonio Mitcheli, an Italian priest and biologist, discovered *Aspergillus* in 1729<sup>1</sup>. The first description of aspergillosis in man was reported by Bennet in 1842<sup>2</sup>. Since then different spectrums

of the disease is known in man including invasive aspergillosis, aspergilloma and allergic bronchopulmonary aspergillosis. Sluyter was first to report pulmonary Aspergillosis in 1847<sup>3</sup> and Gerstly reported the first lung resection for it in 1948<sup>4</sup>.

sentation, underlying lung abnormalities in patients with Mycetoma in our population and share our surgical experience and perioperative outcome in Peshawar.

## METHODOLOGY

It was a retrospective, descriptive case series carried out in Cardiothoracic Surgery Unit Postgraduate Medical Institute Lady Reading Hospital (PGMI, LRH) Peshawar Pakistan. Computerized medical records of patients operated upon for Mycetoma, were reviewed from June 2002 to June 2013. Patients of all ages, both male & female, diagnosed as Mycetoma were included in the study. Patients with Mycetoma along with empyema, carcinoma and active Tuberculosis were excluded from the study. Demographic data, clinical presentation, underlying lung disease, investigations performed, site of Mycetoma, operative procedure & outcome were noted for each patient.

Mycetoma was classified into simple & complex types according to Belcher & Plummer criteria.<sup>17</sup>

All the patients underwent Chest x-ray (CXR), CT scan, Bronchoscopy, Serology and pulmonary function testing. CT guided Transthoracic needle aspiration biopsy (TTNA) was performed in patients, where diagnosis was in doubt.

All surgeries were performed through standard posterolateral thoracotomy using one lung ventilation. All resected specimens were sent for histopathology.

Hemoptysis was defined as blood with cough or sputum, ranging from blood streaked sputum to fatal hemoptysis episode. Massive hemoptysis was defined as 300ml or more blood per 24 hours while life-threatening hemoptysis was defined as 600ml or more blood per 24 hours or the episode with choking of the patients. Morbidity was defined as significant intra-operative (> 1000ml) & postoperative (> 1000ml/first 24 hours) hemorrhage, injury to surrounding structures, wound

infection & wound dehiscence, respiratory failure and pleural space problems including prolonged air leak (>7 days), bronchopleural fistula (BPF), empyema and residual pleural space.

Mortality was defined as any death within first 30 postoperative days. All the data was analyzed using SPSS version 16. Continuous data was presented as mean +/- SD while categorical data was presented as frequencies and percentages. All the data was presented as tables.

## RESULTS

A total of 294 patients underwent surgery for Mycetoma during the study time period. There were 176 (60%) males and 118 (40%) females with mean age of 38.4 years ± 11.30 SD (range 14 to 71 years).

The most common symptom was recurrent hemoptysis (92%) while 8

(2.7%) patients were asymptomatic. Sixty two patients had massive hemoptysis out of which 29 patients had complex aspergilloma. Underlying pulmonary diseases were present in 288 (97.9%) patients while 6 (2%) patients had no evidence of previous disease, 3 were diagnosed on CT guide TTNA and 3 were operated upon for indeterminate mass, only to be diagnosed as Mycetoma on histopathology of the resected specimen. The most common pre-existing pulmonary disease was Tuberculosis (TB) (272 patients - 92.5%). Two hundred and twelve (72.1%) patients were having simple Mycetoma. The most common location of Mycetoma was left upper lobe (57.4%) while 6 patients had Mycetoma in multiple lobes and one had bilateral mycetoma. All the above information is summarized in Table I.

All surgical procedures were performed electively except 8 patients

**TABLE I: BASELINE CLINICAL CHARACTERISTICS (N=294)**

Variable	Frequency (Percentage)
<b>Presentation</b>	
Hemoptysis	271 (92%)
Chest pain	42 (14.2%)
Recurrent RTIs	31 (10.5%)
Asymptomatic	8 (2.72%)
<b>Underlying Lung disease</b>	
Pulmonary TB	272 (92.5%)
Bonchiectasis	12 (4%)
Bullous lung disease	2 (0.68%)
Carcinoma lung	1 (0.34%)
Lung abscess	1 (0.34%)
Indeterminate	6 (2%)
<b>Location of Mycetoma</b>	
Left upper lobe	169 (57.4%)
Right upper lobe	113 (38.4%)
Right middle lobe	2 (0.68%)
Right lower lobe	2 (0.68%)
Left lower lobe	1 (0.34%)
Multiple	6 (2%)
Bilateral	1 (0.34%)
<b>Type of Aspergilloma</b>	
Simple	212 (72.1%)
Complex	82 (27.9%)

**TABLE II: OPERATIVE PROCEDURES PERFORMED (N=294)**

Surgical procedure	Frequency (Percentage)
<b>Primary procedure</b>	
Lobectomy	254 (86.3%)
Bi-lobectomy	7 (2.38%)
Wedge resection	16 (5.4%)
pneumonectomy	12 (4%)
cavernostomy	5 (1.7%)
<b>Supplementary procedure<sup>a</sup></b>	
Thoracoplasty	11 (3.7%)
Myoplasty <sup>b</sup>	4 (1.36%)
<b>Subsequent procedure<sup>c</sup></b>	
Thoracoplasty	7
Myoplasty <sup>b</sup>	1
<b>Completion Pneumonectomy</b>	1
Exploration for hemorrhage	3
Resuturing of wound	1

- a- Same stage procedure in addition to primary surgical resection
- b- Performed with already harvested Latissimus Dorsi flap
- c- Redo surgery for complications

**TABLE III: POSTOPERATIVE COMPLICATIONS (N=294)**

Complication	Frequency
<b>Significant hemorrhage</b>	
Intraoperative <sup>a</sup>	8
Postoperative <sup>b</sup>	5
<b>Pleural space problems</b>	
Residual pleural space	5
Prolonged air leak <sup>c</sup>	9
Bronchopleural fistula	2
Empyema	3
<b>Wound complications</b>	
Infection	5
Dehiscence	1
<b>Others</b>	
Respiratory insufficiency	1
Injury to structures	1
Azygus vein Intercostals vessel	3

- a- > 1000ml
- b- > 1000ml/first 24 hours
- c- Persistent air leak for >7days

who underwent emergency surgery for massive hemoptysis and could not be managed conservatively. The operative time averaged  $91 \pm 9.37$  minutes (range 78-190 minutes).

The most common procedure was Lobectomy (86.3%). Thirty eight Lobectomies and 3 pneumonectomies were performed by clamping and mass suturing with Prolene because of massive adhe-

sions in & around hilum. Cavernostomy/ Cavernoplasty was performed in 12 (4%) patients because of prohibitive risk for lung resection. In 15 (5.1%) patients supplementary procedure like Thoracoplasty, Myoplasty; was performed along with primary lung resection for gross residual pleural space. (Table II)

Intraoperative blood loss averaged  $280 \pm 25.35$  ml (range 100-2200 ml). In 8 (2.7%) patients a higher intercostal space was needed to open in order to control hemorrhage from chest wall. Intraoperatively azygus vein was injured in one case and intercostal vessel in three cases. All were ligated.

Postoperative complications occurred in 43 (14.6%) patients (Table III). Postoperative blood loss averaged  $190 \pm 22.45$ ml (range 100 to 2100ml). Blood loss exceeded 1000ml in 5 (1.7%) patients, two were managed conservatively with blood and blood products, out of which one was HCV positive while 3 (1%) patients needed re-exploration to control hemorrhage. Second stage Thoracoplasty was performed in 7 (2.38%) patients (5 for residual postoperative pleural space after upper lobectomies, 1 for post-pneumonectomy empyema, 1 for prolonged air leak). Mean postoperative hospital stay was 9 days (range 5 to 27days).

Overall mortality was 7 (2.3%). There was no intraoperative death. One patient died of post-pneumonectomy respiratory failure, one of pulmonary embolism, 2 of acute respiratory distress syndrom (ARDS), one of myocardial infarction, one after Cavernostomy while one patient had sudden death, cause of which could not be determined.

## DISCUSSION

Two hundred and ninety four patients underwent surgery for Mycetoma in past 11 years, which is a large cohort of patients as compared to most other

studies.<sup>8,10,11</sup> This could be explained by the fact that Cardiothoracic surgery unit, LRH is a high volume Thoracic surgery unit<sup>18</sup> and has a large catchment area as it is the only tertiary care Thoracic surgery unit in the whole province. We also receive patients from other provinces and many patients from neighboring country Afghanistan as well.

Any chronic cavitary lung disease can harbor Mycetoma<sup>5,6</sup>. In our study the most common underlying pulmonary disease was TB (92.5%), also reported by many others.<sup>5,6,11,13,14</sup> The range in the literature varies from 13 to 92%<sup>4,19</sup>. The reason is that the incidence of TB in Pakistan is very high, being the fifth highest burden TB area in the world, as reported by WHO<sup>20</sup>. Mostly poor people living in rural areas with limited access to healthcare facilities are the victims and because of ignorance, lack of education and noncompliance to drugs, timely diagnosis and treatment is delayed leading to cavitary lung disease. These cavities are colonized by *Aspergillus* leading to Mycetoma. The British Thoracic and Tuberculosis association reported 6% chance of developing aspergilloma in patients with cavitary tuberculous lung disease in 3 years<sup>21</sup>. Also Babatasi et al reported that cavitary tuberculous lung disease of more than 7 years duration has increased chance of Mycetoma.<sup>6</sup> Thirteen patients had active disease showing caseous granulomatous inflammation on histopathologies who were referred to pulmonologist for anti-tuberculus treatment.

Recurrent hemoptysis was the most frequent presentation (92%) in our study as reported by others as well.<sup>13,14,22</sup> Range of hemoptysis in literature is from 50 to 83% in different series.<sup>13,14</sup> It was massive in 62 cases and life-threatening in 8 patients making one additional mortality in a preoperative patient (not included in the study) who could not survive after all resuscitation measures and being 2 days

on ventilatory support as well, while in other patients emergency lobectomy was performed. Major hemoptysis episodes were seen in 29 (35%) out of 82 complex and 33 (15%) out of 212 simple Mycetomas. There's no predictability for major hemoptysis and it could be life threatening as well.<sup>15</sup> Though complex aspergilloma has higher rate for major hemoptysis but still simple mycetoma is no exception. Life threatening hemoptysis episodes were estimated to be 20% by Henderson et al<sup>23</sup> and 30% by Garvey et al.<sup>24</sup> There was no recurrence of hemoptysis after surgery in different series.<sup>18,25</sup>

The number of asymptomatic patients quoted in literature is up to 36%.<sup>5,10,11,13,19,23,26</sup> In our study asymptomatic patients were 8, three were diagnosed as mycetoma on CT scan, 3 were diagnosed on CT guided Trans-thoracic needle aspiration (TTNA) and 3 patients were operated upon for indeterminate upper lobe mass and were diagnosed as Mycetoma by histopathology of the resected specimen. Surgery in asymptomatic and patients with minor symptoms, has very few postoperative complications, is essentially very safe and the chance of limited resection is also more in these patients.<sup>8,10,11</sup> There is rare chance of regression if any<sup>8,11</sup>, while there's a fair risk of these patients to become symptomatic which is estimated to be up to 20%.<sup>6,25</sup> Disseminated form of Aspergillosis can develop if the patient harboring asymptomatic mycetoma becomes immunocompromised because of any reason.

As reported in most series,<sup>5,11,15,16,22</sup> diagnosis of the mycetoma was done on the basis of symptoms, past history of cavitary lung disease mostly TB, CXR, CT scan, bronchoscopy to show the source of hemoptysis to be the same lobe harboring the lesion and serology. TTNA was found useful in 6 patients where the diagnosis of Mycetoma was

in doubt, which revealed diagnosis in 3 patients. This technique is mentioned by Demir et al.<sup>11</sup>

Surgery for aspergilloma is technically hazardous and challenging, with high morbidity and mortality. However there is no effective medical treatment available<sup>8,9,22</sup> and surgery is the only chance of cure.<sup>8-12</sup> Medical treatment may only occasionally cause the lesion to shrink but is usually unsuccessful and the lesion still progresses.<sup>8</sup> Surgical morbidity and mortality is especially significant in high risk patients with complex mycetoma. But even in these patients, if surgery is not performed, most patients will have poor quality of life and many will succumb to fatal hemoptysis.<sup>11</sup> One of our patients died preoperatively of massive hemoptysis with respiratory tree soiling & cardiopulmonary arrest, was resuscitated, kept on ventilator for 2 days but was ultimately expired (excluded from the study).

The most common and preferred procedure was Lobectomy in our study which was performed in 254 (86.3%) patients. It is in agreement with previous studies.<sup>8,10,11,13,14</sup> Pneumonectomy was the most high risk procedure in the present study because of the technical difficulties (strong pleural adhesions with indurated inflamed hilar structures with enlarged adherent lymph nodes) which is in agreement with previous studies.<sup>10,11,13,25</sup> Emergency lobectomy was performed in 8 patients for massive hemoptysis, not resolving with conservative treatment. Chen et al had 15 emergency procedures in his series of 256 patients.<sup>8</sup>

Primary procedure of lung resection was supplemented by same stage thoracoplasty or myoplasty in 15 patients for residual pleural space in present study, also reported by Borrelli and others.<sup>8,22</sup> Cavernostomy with thoracoplasty or myoplasty is an acceptable alternative in patients not fit for lung resection surgery, reported by some other authors.<sup>6,10,11,22,25</sup>

In 5 patients cavernostomy with thoracoplasty or myoplasty was performed in present study.

Reported morbidity varies from 15.6% to 70% in different series,<sup>5,6,10,11,13,14</sup> while in one study 67 complications were noted among 51 patients.<sup>26</sup> In our study morbidity was 14.6% including significant bleeding, pleural space problems and bronchopleural fistula (Table III).

There are more chances of intraoperative blood loss in mycetoma surgery as compared to other thoracic surgery procedures because of dense pleural adhesions and secondary collateral vascular supply from the chest wall, also reported by other authors.<sup>5,6,8,9</sup> Intraoperative blood loss exceeded 1000ml in 8 of our patients while postoperatively in 5 cases. We try to avoid excessive intraoperative hemorrhage by the technique that we dissect an area, pack it, work in other area and return to the initial area after interval. We use intra and extrapleural plane of dissection interchangeably where necessary. Also we are using harmonic scalpel for dissection for the last 3 years. Re-exploration for significant postoperative hemorrhage was done in 3 patients in present study which is in acceptable range with some other studies.<sup>9</sup>

Results of pleural space problems are variable in literature but largely are the more common ones, ranging up to 29%.<sup>10,11</sup> In our study pleural space complications occurred in 19 (6.5%) patients. Reduced occurrence is because, along with other routine measures, we strictly avoid spillage of mycetoma contents during surgery and do irrigation with antiseptic (diluted povidone) at the end of the procedure to limit postoperative infectious complications. Along with the primary resectional surgery, additional supplementary procedures are performed for grossly residual pleural space, as agreed by some authors,<sup>3,8,22,25</sup> while

opposed by others.<sup>10,11</sup> Continuous low pressure suction is applied to all patients postoperatively to help the remaining partially compliant lung to fill the pleural space. Subsequent second stage thoracoplasty was needed in 5 patients for residual pleural space.

The incidence of BPF is reported to be from 1.6 to 15.8% in different series.<sup>8,16,27,28</sup> We had a very low incidence of BPF (0.68%) as, in agreement with Chen et al,<sup>8</sup> we routinely cover the bronchial stump with vascularised tissue flap (mostly intercostals muscle flap).

Mortality for mycetoma surgery is reported in different studies in literature to be upto 22%.<sup>8,10,13,16,19,24,25</sup> In our study mortality was 2.3%. Some authors experienced increased mortality with complex Mycetomas and avoid operating upon complex mycetomas, and operate upon simple mycetomas to reduce their operative mortality.<sup>10</sup> Present study contradicts with it and it's shown complex mycetomas can be operated upon with acceptable mortality, also shown by some other authors.<sup>8</sup>

## CONCLUSION

Pulmonary mycetoma, a benign but potentially life-threatening condition is not uncommon in our set up. It's more common in young male patients having underlying tuberculosis and the most common symptom is hemoptysis. Lobectomy was the commonest surgical procedure for Mycetoma in our set up. Surgery can be performed safely though with increased morbidity including hemorrhage and pleural space problems. Operative mortality rate was 2.3% in our study. Surgery is recommended for all patients with simple and complex mycetomas surgically fit patients. Lung resection in the form of Lobectomy is the most common procedure sometimes supplemented with additional procedures like Thoracoplasty and muscle flaps to avoid residual pleural space. In

patients with difficult dissection in and around hilum clamp and suture method can be used for lung resection. In high risk patients, otherwise acceptable for general anesthesia but not fit for lung resection, cavernostomy with or without supplementary procedure is a viable alternative.

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### AUTHOR'S CONTRIBUTION

Following authors have made substantial contributions to the manuscript as under:

**SZAS:** Study design, acquisition of data, drafting the manuscript, final approval of the version to be published

**AB:** Concept, acquisition of data, critical revision, final approval of the version to be published

**VA:** Acquisition and analysis of data, drafting the manuscript, final approval of the version to be published

Authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

### CONFLICT OF INTEREST

Authors declare no conflict of interest

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