

Surgical Experience of Thoracic Hydatidosis - A Retrospective Study

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Abstract:

Introduction: the aim of the study was to retrospectively evaluate all patients with thoracic hydatid cysts, treated surgically in our department between July 2010 To July 2016.

Materials and methods: retrospective analysis was carried out in 350 cases of thoracic hydatidosis, in whom some surgical interventions were carried out at RIMS, Ranchi between 2010 to 2016. There were 204 male patients in the age group from 10 to 81 years (average 32 years). The majority were symptomatic with cough with expectoration (45%).

Results: The surgical access was through posterolateral thoracotomy in 269 patients, limited thoracotomy in 56 patients, and various others in the rest. 60.19% of intrapulmonary cystic lesions were right sided. Associated extrathoracic cysts were found in 70 (20%) patients. Most patients were managed with Conservative surgical procedures (91.42%). 5 patients needed complex procedures. There were 5 postoperative mortalities. Postoperative complications were seen in 30 cases. 11 patients developed prolonged air leaks.

Conclusions: Surgery is the main stay of management for thoracic hydatidosis. Limited thoracotomy and conservative surgical procedures should be the first line of therapy. Radical procedures should be avoided as far as possible. Transdiaphragmatic approach is preferred for hydatid cysts involving right lung and liver.

Keywords: Echinococcus granulosus, Hydatid Cyst, Lobectomy, Pneumonectomy, Thoracotomy.

I. Introduction

Hydatidosis is a parasitic zoonosis caused by larval stage of the tapeworm Echinococcus granulosus. Hydatid disease is known since the time of Galen and Hippocrates, and was described by Thebesius in the 17th century [1]. Rudolphi (1808) first used the term hydatid cyst to describe echinococcosis in humans [2]. Infestation is predominantly in endemic areas in which there is continuous contact between humans and domestic carnivores like dogs and ungulates like sheep[3]. The lung is the second most commonly affected organ (10–40%) after the liver [4,8]. Extrapulmonary thoracic cysts are rare and may be located in fissures, pleural cavity, chest wall, mediastinum, myocardium, and diaphragm [5]. This study was carried out to evaluate retrospectively all thoracic hydatid cysts treated surgically in our department between July 2010 and July 2016.

II. Material And Methods

350 patients with thoracic hydatidosis underwent surgery in our department between July 2010 and July 2016. The majority were male 204 out of 350(58.28%). Age range was from 10 years to 81 years (average 32 years). 175 patients (50%) had a long history of close contact with dogs, or sheep farming. The main clinical presentations were recurrent chest infection (in the form of cough, sputum, and fever), hemoptysis, dyspnoea and chest pain (TABLE 1). However, 70 patients (20%) were symptom-free and were diagnosed as accidental chest x ray findings. (Fig.1).ultrasound chest was done to confirm fluid filled cyst. Chest computed tomography was always done for more clarification regarding size, location, number of cysts and relation with surrounding organs (Fig.2). Ultrasound abdomen was done to detect concomitant liver cysts. Pulmonary Function Tests were done to screen patients with bilateral disease and patients having dyspnoea. Hydatid serology was done with Enzyme Linked Immunosorbent Assay (ELISA) and titers more than 1:1600 were considered for positive hydatidosis.

Table 1. Major clinical manifestations in 280 symptomatic patients.

| Clinical finding | Number of patients (in percentage) |
|--------------------------|------------------------------------|
| Cough with expectoration | 126 (45%) |
| Hemoptysis | 102 (29.14%) |
| Fever | 87 (24.85%) |

| | |
|--------------|-------------|
| Dyspnoea | 40 (11.42%) |
| Chest pain | 33 (9.42%) |
| Asymptomatic | 70 (20%) |

The surgical access were standard posterolateral thoracotomy(Fig. 3) in 269 patients, limited thoracotomy in 56 patients, bilateral staged thoracotomies in 17 patients, combined thoracotomy with transdiaphragmatic hepatic access in 3 patients, median sternotomy in 3 patients and local incision over lesion in 2 patients over breast tissue(TABLE 2). 375 of the 623 intrapulmonary cystic lesions were found in the right lung (60.19%). Intrathoracic extrapulmonary cystic lesions were detected in 9 patients (2.57%). 70 (20%) patients also had cystic lesions in the liver or other abdominal organs.

Table 2. Surgical access

| Incision | Number of Patients (in percentage) |
|---|------------------------------------|
| Posterolateral thoracotomy | 269(76.85%) |
| Limited thoracotomy | 56(16.00%) |
| Bilateral staged thoracotomy | 17(4.85%) |
| Combined thoracotomy with transdiaphragmatic hepatic access | 3(0.85%) |
| Median sternotomy | 3(0.85%) |
| Local incision | 2(0.5%) |

The operative field surrounding the hydatid cyst was covered with towels moistened with povidone iodine to prevent inadvertent implantation of scolices or daughter cysts. Depending on the localization, size and intact state of the cyst we employed various surgical techniques for cyst removal (TABLE 3) such as cystotomy (incision in the lung parenchyma and removal of cyst), enucleation of the intact cyst (Fig. 4) and removal of the cyst after needle aspiration. Enucleation was performed for superficial and small cysts, cystotomy for those deep inside the parenchyma and needle aspiration for large cysts within the parenchyma to prevent implantation of cyst fluid prior to removal of cysts. The residual cavity and lung damage were treated by conservative methods (such as pericystectomy, which is always associated with suturing of bronchial fistulas and capitonnage of the cavity) or by lung resections (such as wedge resection, segmentectomy and lobectomy).

Table 3. Operative techniques employed for 623 intrapulmonary cystic lesions in 341 patients.

| Surgical procedures | Hydatid cyst (in percentage) |
|---|------------------------------|
| Removal of cyst after needle aspiration | 264(42.37%) |
| Enucleation | 263(42.25%) |
| Cystotomy | 62(9.95%) |
| Wedge resection | 20 (3.58%) |
| Segmentectomy | 7 (1.11%) |
| Lobectomy | 3(0.5%) |

Albendazole treatment was given to all patients postoperatively for 3 courses. The albendazole regimen was 400mg (10-20 mg/kg) twice daily for three one month courses with 14 days rest period in between.

III. Results

The descriptions of surgical procedures for intrapulmonary cystic lesions are given in TABLE 3. Our preferred surgical techniques were enucleation of the cyst or cystotomy. In most patients the cyst cavity was closed by capitonnage. 375 of the 623 intrapulmonary cystic lesions were found in the right lung and 248 in the left lung. (TABLE 4). The most common cyst localizations were the lower lobes of both lungs (366 out of 623 cysts).

Table 4. Localisations of 623 intrapulmonary cystic lesions.

| Right lung Lobe involved (in percentage) | Left lung Lobe involved (in percentage) |
|--|---|
| Upper lobe 125 (20.06%) | Upper lobe 63 (10.11%) |
| Middle lobe 63 (10.11%) | Lingula 6 (1%) |
| Lower lobe 187 (30.01%) | Lower lobe 179 (28.73%) |
| Total 375 (60.19%) | Total 248 (39.80%) |

Of the 623 intrapulmonary lesions, 180 were ruptured. The ruptures were directed towards the bronchus in 152 cysts, the pleural cavity in 23 and to the subphrenic area via the diaphragm in 5(TABLE 5).

Table 5. Direction of Rupture in 180 patients

| Site | Number (In Percentage) |
|----------|------------------------|
| Bronchus | 152 (84.44%) |

| | |
|--------------------|-------------|
| Pleural cavity | 23 (12.77%) |
| Transdiaphragmatic | 5 (2.77%) |

98 of 180 ruptured cysts had been infected. It was observed that among 443 intact and uncomplicated intrapulmonary cystic lesions 56 were smaller than 5 cm, 201 were between 5–10 cm and 186 larger than 10 cm. The size of the largest cyst was approximately 20 cm. Extra pulmonary intrathoracic lesions were removed by cyst extirpation from surrounding tissue or by pericystectomy. Of our 350 patients, 9 patients had extrapulmonary intrathoracic cysts of which two patients have myocardial cyst, 2 in breast of female and 5 in chest wall (TABLE 6). In those five patients two of them had rib destruction and partial rib resections were carried out.

Table 6. Operative technique for 9 extra pulmonary intra thoracic hydatid cyst

| | |
|-----------------------------|---------|
| Cyst extirpation | 5 (55%) |
| Myocardial cyst extirpation | 2 (22%) |
| Pericystectomy | 2 (22%) |



Fig. 1:- Chest X-ray showing large Hydatid cyst in Left Hemithorax



Fig.2 :- CT Scan showing multiple Intra and Extrapulmonary Hydatid cysts

70 (20%) patients also had cystic lesions in the liver or other abdominal organs. The etiological agent was identified as *E. granulosus* in all patients. 5 recurrences were observed in the follow-up period ranging from one month to 60 months. All of them needed long term albendazole therapy and repeat surgery. There were 3 deaths in this subset of patients. There were no intra Operative deaths and postoperative mortality was 1.42 % (5 out of 350) over the follow up period. The postoperative course in most patients was uneventful. Wound infection developed in 21 patients of whom 12 were diabetic. Prolonged air leak developed in 11 patients, all of which sealed with conservative measures. The mean postoperative hospital stay was 8.9days. After the operation all the patients' hydatid cyst-related symptoms were improved.





Fig. 3:- Intra operative image showing ruptured cyst Membrane

Fig.4 :- Intact and ruptured cysts removed by enucleation

IV. Discussion

Hydatid cyst disease is a zoonotic disease caused by the larval stage of *Echinococcus granulosus* (dog tapeworm), *E. multilocularis*, or *E. vogeli* [6]. This disease occurs when humans ingest the hexacanth embryos of the dog tapeworm. Infestation by hydatid disease in humans most commonly occurs in the liver (55–70%) followed by the lung (18–35%); the two organs can be affected simultaneously in about 5–13% of cases [7].

Thameur et al (2000) reported frequency of intrapulmonary hydatid cyst of 94.6% in 1,619 patients with thoracic hydatidosis [8]. Srinivasan et al (2010) showed a incidence of 10.23% intrathoracic and extrapulmonary cysts [5]. Al – amran (2008) reported 825 cases of thoracic hydatidosis of which 818(99.15%) had intrapulmonary cysts [9]. In our 350 patients with thoracic hydatidosis 333 (95.15%) had intrapulmonary cysts and 17 (4.85%) intrathoracic extrapulmonary cysts. The mean age of our patients was 32 years.

The World Health Organization published an excellent overview of treatment guidelines for Echinococcal disease in 1996 [10]. The treatment of choice for Hydatid cyst of the lung is complete excision of the cysts with maximum preservation of lung parenchyma [11]. The surgical procedure for treatment of thoracic hydatidosis may be conservative or radical. The conservative procedures are 1) enucleation of intact cysts, 2) removal of the cyst after needle aspiration, 3) cystotomy and 4) pericystectomy (capsule resection) [6,7]. Radical procedures are pulmonary resection such as 1) wedge resection (lung parenchyma resection of less than one segment), 2) segmentectomy, 3) lobectomy, and 4) pneumonectomy [7]. Peripherally located cysts of any size and small-to medium-sized centrally located cysts are amenable to conservative procedures [10]. In our series 91.42% (320 out of 350) of patients were treated with conservative procedures. Most commonly applied cyst removal techniques were removal of cyst after needle aspiration (42.37%) and enucleation (42.25%). VATS approach, although described in literature, were not employed in any of our patients [12]. Segmental resection is indicated for large simple cysts occupying the whole of the involved segment. Lobectomy should be performed when the size, number of cysts and the degree of infection exclude lesser procedures. The indications for lobectomy are large cysts involving more than 50% of the lobe, suppurative cysts unresponsive to preoperative treatment, multiple unilobar cysts and sequelae of hydatid disease such as pulmonary fibrosis, bronchiectasis, or severe haemorrhage. Pneumonectomy is rarely indicated for treatment of hydatid disease of the lung and should be used only when the whole lung is involved in the disease process and no salvageable pulmonary parenchyma remains [13]. In our series we did not need pneumonectomy in any patient.

Thameur et al. [8] reported a lung resection rate of 14.1%. Our indications of lung resection technique included destruction of lung parenchyma as a result of bronchiectatic changes and chronic inflammation in the lung tissue surrounding very large or multiple cysts. We performed lung resection in 30 patients (8.57%) of which 20 were wedge resections, 7 segmentectomy, and 3 were lobectomy. After the hydatid cyst is removed the bronchial openings should be closed by suturing and the pericystic cavity obliterated to prevent secondary infection [6]. The residual cavity is obliterated by capitonnage using multiple purse-string sutures from the deepest level to surface level [14]. We used the capitonnage technique after removal of 382 hydatid cysts (64.85%). Complex procedures involving thoracic and transphrenic approaches are needed for cysts involving both lungs and liver [15].

Although surgery remains the treatment of choice for thoracic hydatidosis, the usefulness of drug therapy has been reported in many studies. Mebendazole, albendazole and tinidazole have been used as primary drug therapy and as an adjunct to surgery to diminish recurrence and potential spread of the organism [16]. Horton (1997) reported that albendazole therapy in *E. granulosus* infection can result in apparent cure in up to

30% of cases, with a further 40–50% showing objective evidence of response when followed in the short term [17]. Oxfendazole, a promising newer benzimidazole, is being studied in animals [18]. According to World Health Organization guidelines chemotherapy is the preferred treatment when surgery is not available, when complete removal of the cyst is impossible, when cyst contents threaten to disseminate due to cyst rupture, or when cysts are multiple or multiple organ involvement [10,16]. Mawhorter et al. (1997) reported the use of percutaneous aspiration together with albendazole therapy for the treatment of recurrent pulmonary echinococcal infection in a patient [16]. We used albendazole (400 mg twice daily for three 1 month courses with 14 day rest periods between courses). There were 5 recurrences of which 2 died in the follow-up period ranging from 6 months to 72 months. The recurrences were due to multiple Hydatid cysts in inaccessible areas of brain in 2 cases, preoperative contamination during PAIR procedure in 2 cases and in 1 case due to uncontrolled intraoperative spillage. The surgical procedures for hydatid cyst carry a morbidity of 0 to 13% and mortality of 0 to 5% [17]. Qian (1988) reported operative mortality of 0.6% in 807 patients [11]. Dogan et al. (1989) reported postoperative complications in 37 patients (3.5%) and 30-day mortality was 1.7% in 1,055 patients treated surgically for pulmonary Hydatid disease [19]. Thameur et al. [8] reported operative mortality of 0.78% [8]. Al-amran (2008) reported a mortality of nil and postoperative complications in 23 patients (2.78%) in a series of 825 patients [11]. In our experience there were a total of 5 deaths (1.42%). In 3 cases, there were multiple liver cysts with suppuration and peritonitis preoperatively. Two patients had multiple recurrent brain Hydatid cysts with status epilepticus. Postoperative complications were encountered in 30 patients (8.57%) of which 21 had superficial wound infection and 11 had prolonged air leak. All patients responded to conservative measures.

V. Conclusion

Thoracic hydatidosis is involvement of intrathoracic structures by the larval stage of *Echinococcus granulosus*. Diagnosis is usually made by CT scan. Surgery is the preferred treatment and lung preserving procedures are appropriate for most patients, supplemented with albendazole therapy. Occasionally more radical approaches are needed to tackle extensive disease, which can cause increased morbidity and even mortality.

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