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Research Article

Role of VATS in Management of Pulmonary Hydatid and Follow Up: A Case Series

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Abstract

Introduction: Hydatid cyst caused by *Echinococcus granulosus* is endemic in India. Pulmonary hydatid does not have a uniform treatment recommendation. This case series presents the thoracoscopic management of hydatid disease from India.

Method: Retrospective multicentric case series of patients treated between 2003 and 2018 at three multispeciality centers are included in this study. 16 patients with an average follow-up of 17 months was done.

Results: Thoracoscopic surgery performed in all 16 patients with mean duration of hospital stay of 5 days and there was no recurrence noted at 6 months follow up.

Conclusion: Thoracoscopic surgery helps in faster recovery and needs to be offered in all patients with hydatid cyst.

Keywords: *Hydatid Cyst; Echinococcus granulosus; Pulmonary Hydatid Cyst; VATS; Video Assisted Thoracoscopic Surgery; Pulmonary Echinococcosis; Minimal Access Surgical Procedures*

Abbreviations

VATS: Video Assisted Thoracoscopic Surgery

Introduction

Human Echinococcal disease is mainly caused by the larval forms of two species i.e. *Echinococcus granulosus* and *Echinococcus multilocularis* [1]. Hydatid cyst is caused by *E. granulosus* and it is seen to be hyper endemic in the sheep grazing communities of Mediterranean, middle east countries, etc. and endemic in India [2]. The larval forms are seen in almost any organ in the human body but the most common sites are the filtering organs liver followed by lungs. Pulmonary hydatid disease varies in its presentation making a uniform treatment recommendation impossible [1]. Therefore we decided to share our experience in the management of pulmonary hydatid disease.

It is known that the surgical management of lung hydatids need to be as conservative as possible [1]. As pulmonary hydatids are seen more commonly in children there are studies done in this population where the advantages of Video assisted thoracoscopic surgery compared to thoracotomy are clearly demonstrated with respect to length of hospital stay, intraoperative blood loss and thoracic intubation dwelling time [3]. With the advances in minimal access thoracic surgery these techniques are being applied to the adult population to enable a faster postoperative recovery and there are case series reported from hydatid endemic areas like Turkey [4].

Hybrid Video assisted thoracoscopic surgery gives the dual advantage [5] of operating under maximal vision and delivery of the daughter hydatid's without spillage of the contents through the muscle sparing utility incision. Here we would therefore like to present the case series of Video assisted thoracoscopic surgery/hybrid Video assisted thoracoscopic surgery applied in cases of pulmonary hydatid disease.

Materials and Methods

Study design: The study is a retrospective multicenter case series where consecutive patients of hydatid disease involving the lung were included.

Setting: 16 patients of pulmonary hydatid operated from 2003 to 2018 at three centers by thoracic surgical team were included. All three centers were multispecialty and were catering to patients from India and also from countries in the Middle East. All the patients underwent the similar operative technique of Video assisted thoracoscopic surgery/hybrid Video assisted thoracoscopic surgery and were followed up for a minimum period of 3 months and maximum period of follow up in the first operated patient of the series was 15 years.

Data collection was done retrospectively from case records and follow up was done by telephonic conversations with the patient s and reviewing the digital imaging pictures i.e. chest X ray done at 1 and 3 months following the surgery.

All patients presenting with hydatid cyst involving the lungs were included irrespective of the age. After the diagnosis was established by imaging modalities i.e. Chest X ray and Computed tomography of the chest, all patients were worked up for surgical management. None of the patients in the series received preoperative antiparasitic agents like albendazole.

All patients were taken up for thoracoscopic surgery. All were subjected to general anaesthesia and intubated with Double lumen endotracheal tube for one lung ventilation. They were operated by experienced thoracic surgeons but there were variations in technique as three different surgeons were involved in the surgical management.

The data was interpreted by an independent researcher to decrease the element of interpretation bias. Surgical technique involved filling the thoracic cavity with 10%cetrimide or 10% povidone iodine for 15 minutes before proceeding to do the cystectomy. No Hypersensitivity reactions were noted during the surgery in any of the patients in the series. The daughter cysts were removed intact either using an endobag/through the utility incision which was created by extending the ports created for Video assisted thoracoscopy usually in the fifth intercostal space anteriorly. The residual peri-cyst cavity was examined for air leaks by filling the cavity with normal saline and asking the anaesthetist to give positive pressure. If there was air leak noted the area was identified and captionnage was performed.

Figure 1: *Hybrid VATS technique with thoracoscope at the prospective drain site.*

In the postoperative period the Intercostal drain was usually removed on the second post-operative day. And all patients were given antiparasitic treatment with Albendazole for a minimum period of 6 months. All the patients were followed up for a minimum of 1 and 3 months following the surgery.

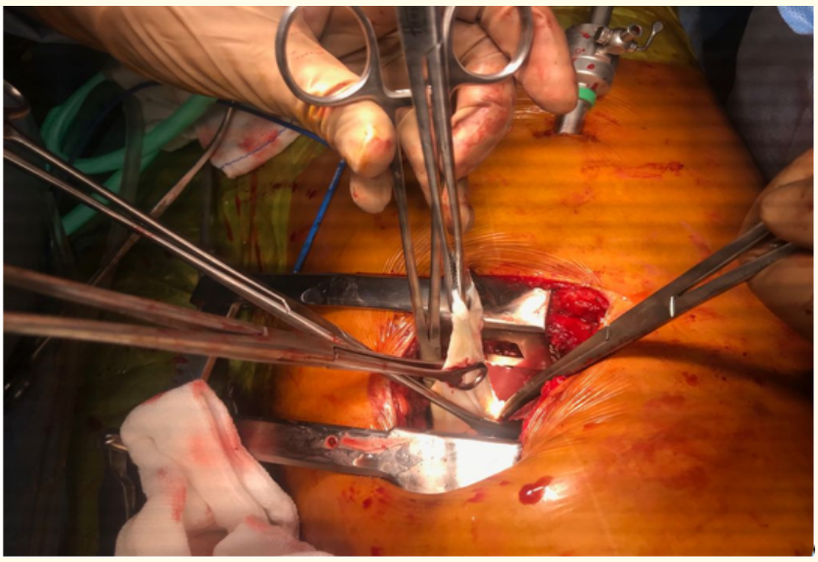

Figure 2: *Hybrid VATS used for delivery the daughter cyst through the utility incision and thoracoscope port in place to offer better magnification.*

Results and Discussion

Results

Patients diagnosed to have pulmonary hydatid cyst included in the study had a mean age of 33 years [range 9 - 55 years]. Male to female ratio of presentation was 1:1.28. The majority of the patients were from India and three of the patients were from Iraq. The demographics of the 16 patients included in the study are presented in the table 1 below.

Patient Number	Age	Sex	Place of residence	Location of hydatid cyst	Number of Hydatid Cyst	Surgery performed
1	17	Female	Iraq	Bilateral	Multiple	Robot assisted
2	55	Female	India	Right middle lobe	Single	VATS
3	9	Female	India	Right lower lobe	Single	VATS
4	39	Female	Iraq	Left lower lobe	Single	VATS
5	21	Female	India	Right lower lobe	Single	VATS
6	14	Male	India	Right lower lobe lung	Single	VATS
7	45	Male	India	Left lower lobe	Single	VATS
8	48	Male	India	Right lower lobe	Single	VATS
9	51	Male	Iraq	Right middle and lower lobe, left lower lobe	Multiple	Hybrid VATS Right partial cystectomy+ hepatic hydatid
10	18	Female	India	Right upper lobe	Single	VATS
11	32	Female	India	left lower lobe	Single	VATS
12	46	Male	India	Right lower lobe	Single	VATS
13	38	Female	India	Right upper lobe	Single	VATS
14	25	Male	India	Left lower lobe	Single	VATS
15	29	Female	India	Left upper lobe	Single	VATS
16	43	male	India	Right lower lobe, right middle and left lower lobe	Multiple	VATS

Table 1: *Demographics and patient characteristics of the 16 patients included in the case series.*

Most of the patients underwent unilateral surgeries except for one patient who underwent bilateral hydatid cyst excision. The Patient with bilateral hydatid cyst who underwent excision in the same sitting was young and fit and therefore taken up for bilateral surgery in the same sitting.

Most of the hydatid cysts were located peripherally except in one patient who presented with a centrally located hydatid cyst requiring a lobectomy for removal of the hydatid cyst.

Four of the patients were noted to have an air leak intraoperatively and this was addressed by captionnage using 2-0 vicryl.

Two of the patients in the series developed an air leak in the post-operative period which settled with conservative management and intercostal drain placement.

One of the patient diagnosed preoperatively with a single right lower lobe hydatid was found to be infected with *Echinococcus multilocularis* on the final histopathology.

The mean duration of hospital stay was 5 days in our series, only two patient who had bilateral disease had a prolonged hospital stay of 14 and 15 days. This value has skewed the mean duration of hospital stay.

Patient number	Follow up duration	Evidence of Recurrence on Imaging Outcome	
		1 Month	3 Months
1	1 year	No recurrence	No recurrence
2	6 months	No recurrence	No recurrence
3	6 months	No recurrence	No recurrence
4	6 months	No recurrence	No Recurrence
5	6 months	No recurrence	No Recurrence
6	6 months	No recurrence	No recurrence
7	6 months	No recurrence	No recurrence
8	3 months	No recurrence	No recurrence
9	1 month	No recurrence	Not available
10	15 years	No recurrence	No recurrence
11	3 months	No recurrence	No recurrence
12	2 months	No recurrence	No recurrence
13	5 months	No recurrence	No recurrence
14	1 year	No recurrence	No recurrence
15	3 months	No recurrence	No recurrence
16	6 months	No recurrence	No recurrence

Table 2: *Outcome of the patients at follow up as noted on imaging.*

All patients were followed up for a duration of 6 months except for patient 8 who has completed one month follow up till date. Patient 1 had a one year follow up and patient 10 had a 15 year follow up.

All of them were free of recurrence at follow up and this was confirmed by Chest X ray. Computed tomography was not done routinely during follow up. It was reserved for those cases with evidence of recurrence on Chest X ray. Images of the pre and post op follow up are presented here.

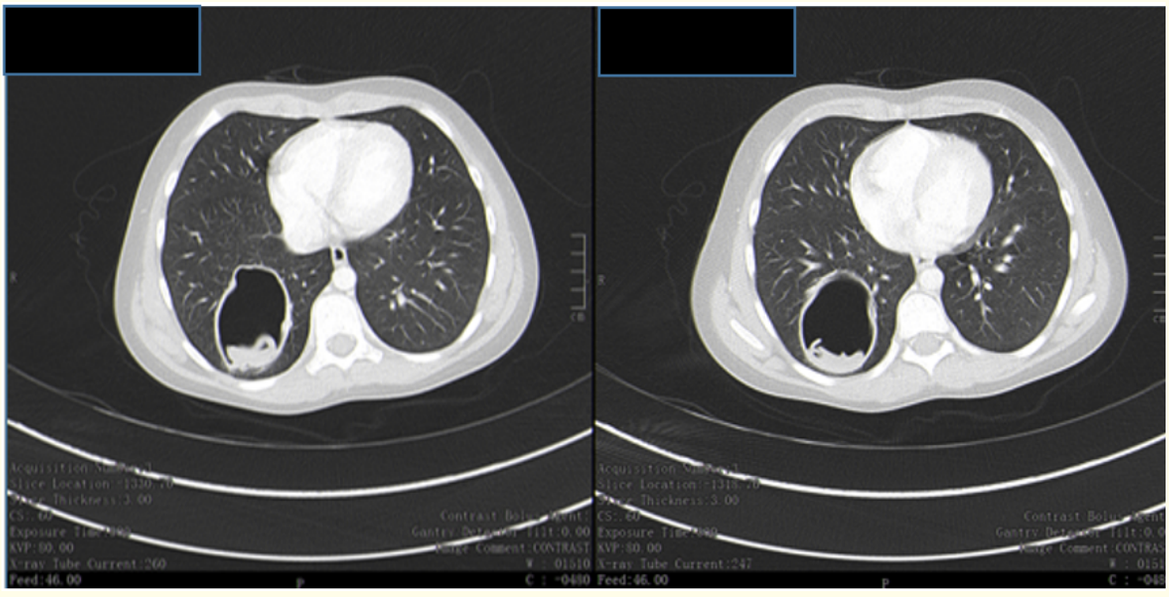

Figure 3: *Preoperative Computed tomography of patient 1 showing the hydatid cyst lung.*

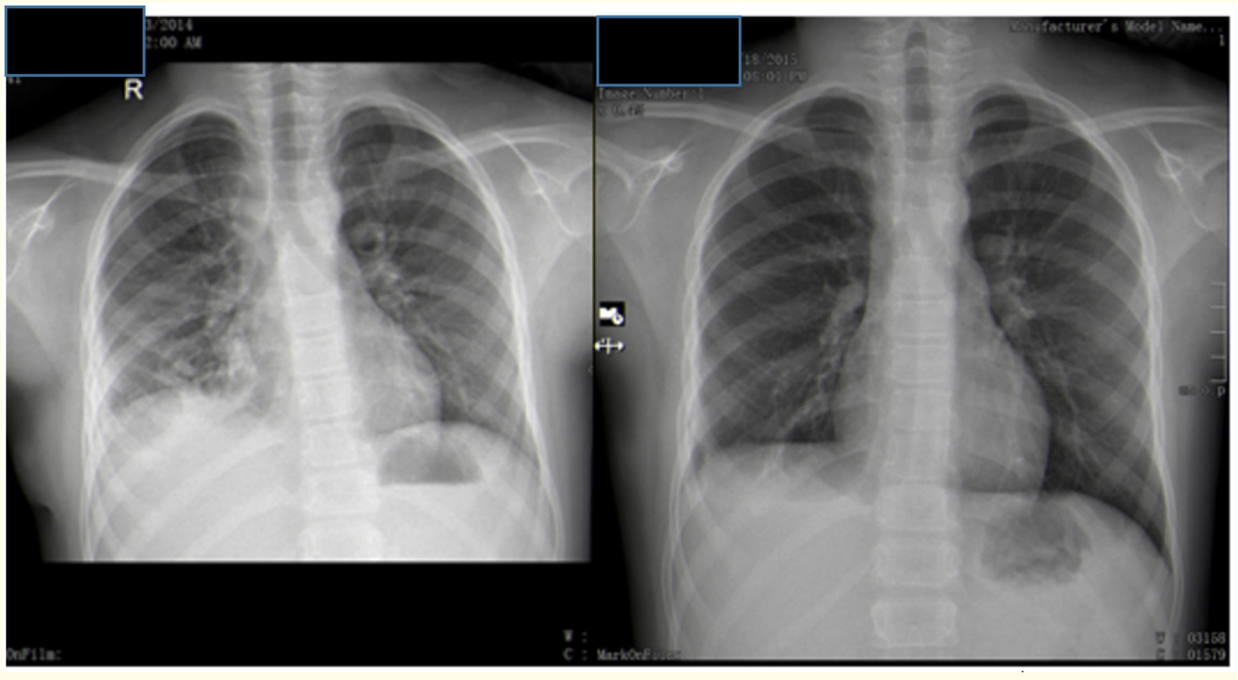

Figure 4: *Post-operative Chest X ray before discharge and at 3 months follow up of patient 1.*

Discussion

Pulmonary hydatid cyst studied in our series were in the age groups ranging from 9 to 55 with a mean age of presentation of 33. Studies done by Sadrizadeh., *et al*. from Iran also found a similar mean age at presentation. Their study also had a wide variation in age range from 7 to 70 years [6]. Lung hydatids are therefore not confined to a particular age group.

Middle east countries and India are endemic for the disease. The patients in our case series belonged to hydatid endemic areas. It's a common zoonotic disease endemic in the sheep grazing areas and leads to significant disease burden in humans. Human to human transmission does not occur. Therefore preventive measures ideally needs to be taken at the level of the animal hosts. But as these livestock are not symptomatic this preventive measure is not a priority for the livestock owners. To make the preventive and control programmes cost effective combining hydatid with other zoonosis for the programme is a solution, with simultaneous initiation and acceptance by the livestock owners [7].

7 of 16 cases presented with cyst involving the right lower lobe. Right lower lobe was the commonest area involved in a study done by Sehitogullari., et al. [6] in Turkey as well. The infective form of hydatid spreads through the blood stream and liver is the most common organ involved followed by Lungs. The lower lobes of the lungs with their lower ventilation perfusion ratio could explain the increased occurrence in lower lobes of the lung.

Once the lung is involved, transbronchial spread occurs and this explains the bilateral disease seen in two of our patients. Intraoperatively the method of single lung ventilation was adopted to prevent this transbronchial spread of the cyst.

In cases of bilateral hydatids usually unilateral surgery is performed first followed by one month interval and before operating on the other side.

The surgical approach used in our case series was video assisted thoracoscopic surgery mainly and where indicated hybrid video assisted thoracoscopic surgery was performed by creating a utility incision by extending once of the thoracoscopic ports anteriorly. Studies have been done comparing thoracoscopic surgery and thoracotomy and there is a definite reduction in postoperative pain and length of hospital stay noted in the thoracoscopic group. Hybrid VATS was performed depending on the size of the hydatid. It was performed in large hydatids as Hybrid Video assisted thoracoscopic surgery offers the added advantage of delivering the cyst without spillage and its associated complication. The utility incision created is a muscle sparing incision which enables faster postoperative recovery as it is not impeded by increased pain [4,5].

Intraoperatively the main step that has helped us prevent any anaphylactic reaction has been the use of 10% cetrimide or 10% povidone iodine that was instilled into the thoracic cavity and left in-situ for 10 - 15 minutes. This ensures that the scolicidal has reached all the corners of the cavity and if there is a spillage the daughter cysts are killed. None of the 16 patients in this series developed any anaphylactic reaction. This has been a novel approach practiced which has shown benefits.

The mean duration of hospital stay was 5 days. We would have expected to have a further reduced mean but 2 of the 16 patients had a prolonged stay and this increased the average mean. One of the patients had a bilateral disease operated in the same surgery and the other patient had comorbidities and this contributed to the increased length of stay. The other studies done using Video assisted thoracoscopic surgery also documents an average length of stay of 4 days [4]. In contrast, thoracotomy which was performed for hydatid cyst reports a hospital stay which is more than double the length of stay for VATS.

Follow up of the cases were done as per the recommendations of the expert consensus group [1] at 3 and 6 months with imaging modalities. One of the patients had a one year follow up and another patient was followed up for 15 years. None of the patients had a recurrence. Recurrence could be due to either a missed contralateral lung lesion or reinfection if the patient is from endemic area and does not take adequate precautions or due to intraoperative spillage of the cyst contents which usually presents as pleural hydatidosis [8].

There was no intraoperative spillage of the cyst contents in our series and particular attention was given to fill the thoracic cavity with the scolicidal agent and waiting for 15min before operating on the cyst. During the delivery of the cyst either endobag or making use of the utility incision could be contributory to the absence of recurrence in the series. All patients were put on albendazole post operatively and this also helps to control and prevent recurrence.

Conclusion

Pulmonary hydatid cyst can be effectively managed by either Video assisted or Hybrid Video assisted thoracoscopic surgery. All patients presenting with pulmonary hydatid should be offered the option of thoracoscopic surgery so that a shorter hospital stay, earlier recovery and faster return to normal activity can be achieved. But we would need larger studies to guide treatment recommendation.

Conflict of Interest

There is no conflict of interest.

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