

# Clinical Profile and Outcome of Surgery of Patients with Hydatid Cysts at the National Referral Hospital, Bhutan: An Observational Study

Kelzang S,<sup>1</sup> Dorji T,<sup>2,3</sup> Tenzin T<sup>1,3</sup>

<sup>1</sup>Faculty of Postgraduate Medicine, Khesar Gyalpo University of Medical Sciences Thimphu, Bhutan.

<sup>2</sup>Kidu Medical Unit, His Majesty's People's Project, Thimphu, Bhutan.

<sup>3</sup>Jigme Dorji Wangchuck National Referral Hospital, Thimphu, Bhutan

## Corresponding Author

Thinley Dorji

Jigme Dorji Wangchuck National Referral Hospital,

Thimphu, Bhutan.

E-mail: dorji.thinleydr@gmail.com

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## ABSTRACT

### Background

Hydatid disease a zoonosis caused by tapeworms (cestodes) of genus *Echinococcus*. Humans acquire this infection by ingestion of eggs of *Echinococcus* from infected dogs, sheep, goat, cattle or pigs. Hydatid cyst causes significant morbidity, mortality and socioeconomic loss.

### Objective

The objective of this study was to describe the clinical profile and short-term outcome of surgical treatment of hydatidosis at Bhutan's largest hospital.

### Method

This was a descriptive study conducted among patients  $\geq 18$  years at the National Referral Hospital, Thimphu, Bhutan from 01 January to 31 December 2017. Data on pre-surgery treatment, type and outcome of surgery and histopathology were collected. Data was entered analysed in EpiData. All variables are presented using descriptive statistics. Ethics approval was obtained from the Research Ethics Board of Health, Ministry of Health, Bhutan.

### Result

There were 38 patients who underwent surgeries. The mean age of the sample was 36 ( $\pm 17$ ) years; cases were reported from almost all districts in the country. The most common symptoms at presentation was abdominal pain (27, 71.4%), nausea (21, 55.3%), abdominal distension (16, 41.1%). The median duration of symptoms was 6.0 months (IQR 3.0, 12.0). Liver was involved in 36 patients (94.7%). All underwent de-roofing and drainage through laparotomy (35; 92.1%), laparoscopy (2, 5.3%) or thoracotomy (1, 2.6%). Histopathologic confirmation was obtained in 28 patients (73.7%). Twenty-three patients (60.5%) suffered from minor post-operative complications.

### Conclusion

Patients with hydatid cyst present with abdominal pain, nausea abdominal distension. They suffer with symptoms for long periods till surgical treatment. The majority had favourable outcomes of surgery.

## KEY WORDS

*Infection, Neglected tropical disease, Parasites, Surgery Bhutan*

## INTRODUCTION

Hydatid disease is one of the neglected tropical diseases and a zoonosis of worldwide distribution.<sup>1</sup> It is caused by the adult or larval stages of tapeworms (cestodes) belonging to the genus *Echinococcus* (family Taeniidae).<sup>2</sup> Domestic dogs are the most frequent definitive host while other domestic animals such as sheep, goat, cattle or pigs are intermediate hosts.<sup>3</sup> Humans are accidental dead-end hosts who acquire this infection by ingestion of eggs of *Echinococcus* excreted by the infected definitive hosts.<sup>1</sup>

Infection with hydatid diseases causes significant morbidity, mortality and socioeconomic loss.<sup>4,5</sup> The major sources of morbidity results from pressure effects from the hydatid cyst (cystic hydatidosis) in liver, brain, reproductive tract or bone; or from rupture of the cyst causing anaphylaxis or dissemination of the infection.<sup>2,6</sup>

Human infection in Bhutan related to the socioeconomic factors such as farming and animal rearing that bring humans in close contact with parasite eggs.<sup>7</sup> This is the first study from Bhutan to report on the clinical profile and outcome of surgical treatment of hydatid cysts at the National Referral Hospital, Thimphu.

## METHODS

This was a descriptive study conducted among patients receiving surgical treatment for hydatid cysts at the National Referral Hospital in Thimphu, Bhutan. Bhutan is a small Himalayan country with 62.2% of its population of 681,720 residing in rural areas.<sup>8</sup> Bhutan has a three-tiered health system where surgical facilities are available at the three tertiary hospitals located in Thimphu, Gelephu or Monggar.<sup>9</sup> The National Referral Hospital receives almost all of the cases of hydatid cysts from across the country because of the availability of post-operative intensive care services and histopathology services.

This study was conducted among patients aged  $\geq 18$  years receiving surgical treatment for hydatid disease at the National Referral Hospital from 01 January to 31 December 2017. Data on pre-surgery medical treatment was collected using a standard interviewer administered questionnaire. The type and outcome of surgery and histopathological assessment of the hydatid specimen were extracted from patient record. Data was double entered and validated in EpiData Entry 3.1 and analysed in EpiData Analysis 2.2.2.182. All variables are presented using descriptive statistics.

Ethics approval was obtained from the Research Ethics Board of Health, Ministry of Health, Royal Government of Bhutan. Approval was also obtained from the National Referral Hospital to conduct the study. Informed written consent was obtained from each patient to provide interview and allow extraction of their clinical data from patient records.

## RESULTS

There were 38 patients with hydatid cyst that were provided surgical treatment during the year 2017. The mean age of the sample was 36 ( $\pm 17$ ) years. The patients came from thirteen of Bhutan's twenty districts. The details are given in Table 1.

**Table 1. Socio-demographic characteristics of patients with hydatid cyst who underwent surgical treatment at the National Referral Hospital, Thimphu, Bhutan, 2017**

Variables	n	%
<b>Sex</b>		
Female	26	68.4
Male	12	31.6
<b>Occupation</b>		
Farmer	19	50
Civil servant	5	13.2
Student	10	26.3
Monk	1	2.6
Others	3	7.9
<b>Geographic distribution (districts with top five burden)</b>		
Bumthang	9	23.7
Wangdue Phodrang	6	15.8
Chukha	5	13.1
Paro	3	7.9
Trongsa	3	7.9
<b>Marital Status</b>		
Married	26	68.4
Unmarried	12	31.6

### Clinical profile

The most common symptoms at presentation and the organs involved by hydatid cysts are described in Table 2. The median duration of symptoms was 6.0 months (IQR 3.0, 12.0). *Echinococcus* Ig was positive in 26 patients (68.4%) and negative in seven patients (18.4%) and the test was not done in the rest. All patients had undergone pre-operative treatment with albendazole pre-operatively for a mean duration of 40.5 days ( $\pm 14.0$ ).

### Surgical treatment

Thirty-five patients underwent surgery with laparotomy, de-roofing and drainage method (97.4%). Additional procedures were done wherever applicable: omentopexy in 28 patients (73.7%), cystectomy of pelvic hydatid in one (2.6%), cystectomy of splenic hydatid in one (2.6%), thoracotomy in one (2.6%). Two patients underwent laparoscopy deroofing and drainage. Povidone iodine (10%) was used as an intra-operative scolicidal agent in all cases. All patients were prescribed albendazole for a duration of 30 days. Histopathologic confirmation was obtained in 28 patients (73.7%). The post-operative events are described in Table 3.

**Table 2.** Clinical profile of patients with hydatid cysts who underwent surgery at the National Referral Hospital, Thimphu, Bhutan, 2017

Variables	n	%
<b>Common symptoms at presentation*</b>		
Pain in upper abdomen	27	71.4
Nausea	21	55.3
Abdominal distension	16	41.1
Abdominal mass	12	31.6
Dyspeptic symptoms	2	5.2
Breathing difficulty	1	2.6
<b>Organs involved by hydatidosis reported by CT scan**</b>		
Liver	36	94.7
Lungs	1	2.6
Pelvic cavity	1	2.6
Spleen	1	2.6
<b>WHO classification of cysts by ultrasound scan***</b>		
Type I	31	81.6
Type II	5	13.2
Type III	1	2.6
Type IV	1	2.6
Type V	0	0

\*Multiple responses were allowed

\*\*One patient had involvement of two organs

\*\*\*World Health Organization (WHO) classification of hydatid cysts: Type 1 = unilocular cyst + wall; Type 2 = detached membrane; Type 3 = multivesicular, multiseptated cyst with daughter cysts; Type 4 = heterogeneous cyst, no daughter vesicles; Type 5 = cyst with wall calcification

**Table 3.** Type of surgery undertaken and its outcome among with hydatid cysts at the National Referral Hospital, Thimphu, Bhutan, 2017

Variables	n	%
<b>Surgical interventions</b>		
Non-radical de-roofing and drainage	37	97.4
Radical surgery	1	2.6
<b>Post-operative clinical events**</b>		
Atelectasis of alveoli	14	36.8
Surgical site infection	7	18.4
Biliary leak	1	2.6
Broncho-pleural fistula	1	2.6
Death	0	0

Clavien-Dindo classification of postoperative complications\*\*

## DISCUSSION

This study showed that surgeries for hydatid disease constituted 1.2% of the surgical volume conducted at the National Referral Hospital in 2017.<sup>10</sup> Almost half of the caseload (52.7%) came from semi urban districts of Bhutan where there is an overall high rate of migration of

population from the rural areas.<sup>8</sup> The highest number of patients came from Bumthang which is known for rearing of sheep. The cases were reported higher among female with a ratio of 2.1:1 representing the fact that females predominantly take up household and farming chores in Bhutan.<sup>6</sup>

Patients with hydatid cyst commonly reported abdominal distension and pain in the right upper quadrant of abdomen similar to reports from Middle East and Africa.<sup>11,12</sup> Similar to other findings, the liver was the most common organ that was affected.<sup>13</sup> These symptoms and findings are reported in number of other conditions and physical examinations are not helpful in diagnosis of the disease. All except one case had active hydatid disease at the time of presentation. However, up to 60% of patients with hydatid cysts can remain asymptomatic.<sup>14</sup> Therefore, investigations and imaging are essential to diagnose abdominal hydatid cysts.

In our study, latency from symptoms to correct diagnosis and treatment was 8.5 months. The cases do not get timely diagnoses due to the lack of ultrasound facilities at smaller centres located in rural areas. It might also because of lack of awareness about the endemicity of hydatidosis in Bhutan.

All patients were given pre- and post-operative albendazole to prevent recurrence of the disease.<sup>15</sup> These patients were offered surgical treatment with an aim to eradicate the parasite from the body and to obliterate the residual cavity.<sup>16</sup> We performed de-roofing and drainage of the hydatid cyst through laparotomy, laparoscopy or thoracotomy. Puncture-aspiration-injection-reaspiration (PAIR) was not performed in our setting. Povidone iodine (10%) was used as the scolicidal agent to deactivate the active cyst and careful attention was paid to prevent the formation of cysto-biliary communications.<sup>12</sup>

Surgical procedures for the removal of hydatid cyst has reported post-operative mortality rates of 0.7%.<sup>17</sup> Pre-operative planning for such surgeries require availability of intensive care facilities. In Bhutan, the National Referral Hospital is the only hospital where specialist intensive care facilities are available. While no death occurred following surgery, almost a third of patients had prolonged stay beyond five days and one patient was referred to India for the treatment of persistent broncho-pleural fistula following thoracotomy.

This study included very short follow up period until the discharge of the patient from the hospital. We therefore could not study the rate of recurrence. In addition, the outcome of treatment through only medications is unknown as our sample included only those patients from the Surgical Ward of the hospital.

## CONCLUSION

Patients with hydatid cyst commonly report abdominal pain and patients suffer for long periods till surgical treatment. The cases were reported from almost all districts in the country. The majority of those patients who underwent surgical treatment had favourable outcomes.

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