ORIGINAL ARTICLE

CYSTIC ECHINOCOCCOSIS IN CHILDREN: A 5 YEAR RETROSPECTIVE ANALYSIS AT TIKUR ANBESSA SPECIALIZED TEACHING HOSPITAL, ADDIS ABABA, ETHIOPIA

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ABSTRACT

Background*: Cystic echinococcosis (hydatid cyst) is one of the neglected tropical diseases diagnosed in Ethiopian children. The aim of our study was to describe characteristics of pediatric hydatid cyst admissions to Tikur Anbessa Specialized Teaching Hospital.*

Methods*: We reviewed records of 17 pediatric admissions with a diagnosis of hydatid cyst over a 5 year period (September 2010 – September 2015). We analyzed the socio-demographic characteristics, clinical presentations, and diagnostic features of the children and also studied their treatment outcomes and associated complications.*

Results*: The average age of the children was 7 years and 2 months. The most common location identified for the cysts were the lungs (13 patients). The most common chief complaints were cough and chest pain. The average size of cysts was 6.35 cm in diameter. The most common complications were super-infected cysts. All recovered with a combination of Albendazole and surgical treatment.*

Conclusions*: Cysts were commonly diagnosed in the lungs and male children outnumber females. Early presentation and diagnosis of CE prevents associated complications. This study also raises awareness of this neglected illness as a cause of chronic respiratory or abdominal complaints in Ethiopian children.*

Key words*: Hydatid cyst, Echinococcus granulosus, Lung cyst*

INTRODUCTION

Echinococcusgranulosus is a cestode infection responsible for cystic echinococcosis (CE).Cystic and other domestic animals are at riskechinococcosis is one of the major neglected tropical diseases in Ethiopia. Children dwelling around dogs of acquiring the illness. Humans get infected by ingesting eggs passed in the feces of dogs[1].Though usually asymptomatic, it can lead to symptomatic cysts leading to compression of structures. Hydatid cysts can involve the liver, lungs and rarely in other parts of the body.The lung is the most common anatomic location for pediatric CE while adults mostly have hepatic cysts.Some studies attribute this due to the high elasticity and compressibility of pulmonary tissue and thus a faster growth of hydatid cystsin lungs than liver[2-5].

Cystic echinococcosis can complicate by super-infections, metastases, rupture with anaphylaxis and recurrence. Diagnosis of the

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larvae in the intermediate hosts, especially in humans, is mainly by imaging and immunologic techniques[5].For smaller cysts (less than 5 cm), a 1 – 6 month regimen of benzimidazoles can suffice. A combination of PAIR (percutaneous aspiration followed by instillation of a scolicidal agent and re-aspiration) or surgery; along with benzimidazoles is the recommended treatment for larger cysts [6].

Since untreated hydatidosis can have disastrous complications, it is necessary to understand the pattern of the illness in Ethiopian children. Hence, this study was performed with the aim of presenting our 5 years clinical experience in the diagnosis and management of cystic echinococcosis in children.

METHODS

In this retrospective study, we studied all children diagnosed and treated for cystic echinococcosis in TikurAnbessa specialized hospital from September, 2010 to September, 2015. There were 17 patients in total (10 males and 7 females) enrolled in the study. Medical record books and discharge summaries of patients admitted in the pediatric wards over the specified period of time were reviewed. Information concerning socio-demographic data, epidemiologic risk factors, clinical presentation, results from investigative modalities (including size & location of cysts and associated complications) and outcomes of medical and surgical treatment were recorded accordingly. Descriptive statistics were used to analyze results.

RESULTS

Demographic data

In this study, we reviewed the records of all children diagnosed with hydatid cyst from September 2010 up to September 2015. Totally 17 children, out of which10 males and 7 females, with hydatid cysts were referred to this centre in a period of 5 years. The average age of patients was 7 years and 2 months, which in males was 6 years and 8 months and in females was 7 years and 10 months. Half of the children were from the Oromo ethnic group. The religion of the children's families was Muslim 4 (23.5%), Orthodox Christians 3 (17.7%) and religion was undocumented in 10 (58.8%).

Out of 17 patients 4 were from Addis Ababa, 4 from Oromia region, 3 from Amhara, 2 from Southern nations nationalities and peoples region (SNNPR) and 1 from Ethio-Somali region. Residences of the rest were undocumented.

Clinical presentation and risk factors

The most common anatomic location observed in the patients was isolated lung involvement in 11/17(64.6%) of children while in 2/17 children the location was the liver. 10/17 children were males and 7/17 were females. Multi-organ involvement was seen in 2 cases (cysts were found in the lung and the liver. The gender distribution and location of the cysts in the lobes of the lungs is as shown

in table 1.

The chief complaints of 7 patients with lung cysts were cough while 5 complained of chest pain. A right upper quadrant abdominal pain was seen in 3 of the 5 children having abdominal cysts (liver, mesenteric or splenic). The mean duration of symptoms at time of presentation of all children was 7 months and 6 days. Major complaints are shown in Table 2. A history of animal contact was observed in 4 children. 3 of them had contact with dogs but 1 child had contact with cats and another with cattle.

Table 1: gender distribution and location of lung cysts in children who were treated at Tikur Anbessa Specialized Teaching Hospital, Addis Ababa, Ethiopia

Location of cysts in the lungs	Males (%)	Females (%)	Total (%)
Right middle & lower lobes	1 (11.1%)	0	1
Right hilum	1 (11.1%)	1 (25%)	2
Left upper lobe	1 (11.1%)	1 (25%)	2
Left lower lobe	1 (11.1%)	0	1
Left upper & lower lobes	1 (11.1%)	0	1
Both right and left lungs	0	1 (25%)	1
Undocumented location in lungs	4 (44.4%)	1 (25%)	5
Total	9	4	13

Table 2: symptoms at presentation of children with cystic echynococcosis in children who were treated at Tikur Anbessa Specialized Teaching Hospital, Addis Ababa, Ethiopia

Symptoms	No of patients	Percentage
Cough	3	23.1%
Chest pain	1	7.7%
Cough and chest pain	5	38.4%
Cough and fever	3	23.1%
Hemoptysis	1	7.7%
Abdominal pain	3	42.8%
Abdominal swelling	1	14.3%
Sweating	2	28.6%
Palpitation & exercise intolerance	1	14.3%

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Diagnosis: Laboratory & imaging features

Overall, the disease was diagnosed before surgery in 9 patients (52.94%). The pre-surgery diagnosis was pulmonary tuberculosis in 27.3% of cases of pulmonary cystic echinococcosis. Other pre-surgical diagnoses considered for the lung hydatid cysts were a mediastinal mass (1 patient), bronchogenic cyst (1), and pulmonary cyst (1).

Laboratory examinations showed anemia and an elevated erythrocyte sedimentation rate (ESR) in 5 patients each; leukocytosis in 3 and neutrophilia in a further 3 children. Eosinophilia was seen in none of the cases. Elevated liver enzymes were seen in all children with abdominal cysts.

Radiological examinations showed 23 cysts in total in 17 patients. 5 of the pulmonary cysts were found in the left lung; of which 2 were on the upper lobe, 2 in the lower lobe and 1 involving both lobes. In the rightlung, there were 4 cysts (2 in the right hilum, 1 in the right lower lobe, and 1 involving both right middle and upper lobes). Bilateral cysts occurred in 1 patient. Chest x-ray descriptions of the lung hydatid cysts included parenchymal masses, loculated effusions and hilar densities. But chest X-rays could diagnose only 7.7% of all pulmonary hydatid cysts. Ultrasonography and CT scans proved more accurate forms of diagnosis of CE, with 80% and 100% accuracy respectively. Of the hydatid cysts identified by a chest ultrasound, 6 were noted to have an irregular, double wall. Daughter cysts were noted in 1 child. Chest CT showed a ruptured cyst in 1 child and confirmed an infected hydatid cyst in another. Abdominal ultrasounds performed on cysts diagnosed in the liver, spleen and mesentery showed double walled cysts in 2 patients with a further 2 having daughter cysts.

The number of cysts in patients was varying from the minimum of 1 to the maximum of 3 in each patient and the average was 1.35. The average number of cysts in males was 1.5 and in females were 1.14 in each person. Multiple cysts were detected in 4 children (23.5%). The biggest dimension of cyst was 11.8 cm in its maximum diameter and the smallest was 1.8cm in its maximum span. The average maximum diameter of the cysts was 6.35 cm (Among males it was 6.36 cm and among females it was 6.69 cm). Pulmonary cysts were found to have a larger average size (6.06 cm) than liver cysts (5.15 cm).

TREATMENT AND COMPLICATIONS

Pre-surgery albendazole treatment was given for 88.2% of the patients; for a mean duration of 4 months and 1 day with no toxicities detected. Follow-up ultrasound was not done to assess changes in size of cyst but none disappeared. Following surgery, 64.7% of patients received albendazole treatment for a mean duration of 4 month and 7 days.

Surgery was done in 13 patients with no mortalities; with the remaining being lost to follow-up before being assessed for surgical treatment. Pre-operative complications in relation to the hydatid cysts were seen in 10 children. All 5 children with isolated lung cysts were assessed to have super-infected cysts. The presentation of 3 children with isolated pulmonary cysts was that of recurrent respiratory tract infections. Recurrence was noted in one child who had been diagnosed with a right lung CE and given albendazole (duration: undocumented). A ruptured cyst was documented in one patient while 3 had metastases to other organs.

DISCUSSION

The clinical characteristics and prevalence of CE in Ethiopian children is virtually unstudied. In this study, we evaluated all pediatric CE cases admitted to TikurAnbessa specialized hospital from September, 2010 to September, 2015. Most children in our study were males. In contrast, a female predominance was seen inKhalif et al's study. This was also shown in the study conducted by Assefa et al in Addis Ababa [7,8]. We postulate the male predominance in our study to be related to behavioral risks among boys concerning herding livestock and increased contact with domestic animals like dogs and cattle.

In contrast to adult reports from Ethiopia as well as African and Asian studies, the lungs are more affected in our hospital's pediatric admissions. 76.4% of our cases had pulmonary cysts; either isolated or in conjunction with extra-pulmonary CE. This is also reflected in Iranian and South African pediatric admissions (9,10). This might be due to earlier symptom onset and early presentation of the space-occupying cysts in lung tissue which has less resistance to the more compact organs like the liver, which

oppose cyst growth. The same explanation can be given to the larger size of pulmonary cysts in our patients as compared to hepatic cysts.

Multi-organ involvement was seen in 11.8% of cases in our study. This is in agreement with pediatric admissions' report from Iran (15.2%) but much lower than corresponding figures in South Africa (38.1%). Hydatid cysts can rarely be found in organs likethe kidney, heart, brain, peritoneum, lessersac and extremities (9,10). In our study, we identified cysts located in the mesentery, spleen and the heart (inter-

ventricular septum).

We found that the major presenting symptom of pulmonary cysts was cough followed by chest pain. Other symptoms were hemoptysis and fever in pulmonary CE and abdominal pain and swelling in extra-pulmonary CE.This is in accordance with findings in Aslanabadi's study from Tabriz, Iran and Celebi's study from Turkey (10,11). Among our children, 29.4% had secondarily infected hydatid cysts. This is a high incidence in comparison with reports from Iran (15.3%). Only 1 (5.88%) had a recurrence, much lower than observed in Sjostrand and Olsson's report from China (20%) and Andronikou et al's figures from South Africa (61.9%)(9,10,12). Multiple cysts were identified in 23.5% of our patients which is higher than Tantawy's description of Yemeni pediatric cases (6.7%). This might be due to a more delayed presentation to our hospital; which was on average 7 months and 6 days (12). Diagnosis of CE is significantly aided by imaging. Ultrasonography and computed topographyscans had a high level of accuracyin the diagnosis of our cases. This is echoed in a review from China (with almost all of the cysts in 56 patients being diagnosed by an ultrasound)(13). All of our patients recovered with a combination of Albendazole and surgical treatment.

Limitations: this is a retrospective chart review and incomplete documentation of information and missed patient records are the drawbacks of this study.

CONCLUSION

As exhibited in pediatric case summaries reported from elsewhere, we observed a predominance of lung hydatid cysts among our patients; with cough being the dominant presenting symptom. More males were diagnosed with cystic echinococcosis. Diagnosis using ultrasound and CT scan proved highly accurate. More than half of our cases involved complicated cysts and close to a quarter had multiple cysts. Delayed presentations may have contributed for both and as such, we hope that this study will raise awareness of this neglected illness as a cause of chronic respiratory or abdominal complaints in Ethiopian children.

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