

# Hydatid Cysts in Unusual Sites: A Retrospective Imaging Study in Assam

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## Abstract

**Background:** Hydatid disease is a worldwide zoonosis produced by the larval stage of the *Echinococcus* tapeworm. It can reach any organ or tissue in the body and develops into small hydatid cysts (HCs). Most commonly, it occurs in the liver and lungs.

**Aim:** To study the cases of HCs in unusual sites.

**Materials and Methods:** A retrospective radiological study of HCs other than liver and pulmonary HCs for 1 year in a tertiary care hospital in Assam.

**Results:** Altogether, 11 cases of HCs in unusual sites were included in the study, and different imaging findings were noted.

**Conclusion:** With this study, effort was made to bring some light over the rare HC cases in this part of North-East India.

**Key words:** *Echinococcus*, Hydatid cyst, North-East, Unusual sites

## INTRODUCTION

Hydatid disease (HD) is one of the most frequent parasitosis, caused by the larval stage of *Echinococcus granulosus*, especially in countries with a warm climate such as India, African countries, Turkey, South American countries, and Middle Eastern countries.<sup>1</sup> In India, the highest prevalence is reported in Andhra Pradesh, Tamil Nadu, and Jammu and Kashmir.<sup>2</sup>

Infestation by *E. granulosus* in humans most commonly occurs in the liver (55-70%) followed by the lung (18-35%). Incidence of unusual sites is about 8-10%.<sup>2</sup> There are two types of *Echinococcus* infections. *E. granulosus* is the more common type, whereas *Echinococcus multilocularis* is less common but more invasive, mimicking a malignancy. Dogs or other carnivores are definitive hosts, whereas

sheep or other ruminants are intermediate hosts. Humans are accidental intermediate host infected by the ingestion of food or water that has been contaminated by feces of definitive hosts containing the eggs of the parasite. After the outer capsule of the egg has been ingested, the freed embryo (oncosphere) enters a branch of the portal vein by passing through the duodenal mucosa. Most of these embryos become lodged in the hepatic capillaries, where they either die or to grow into hydatid cysts (HCs).<sup>3,4</sup>

Here, a study was conducted on HCs occurring in unusual sites in human in a tertiary care hospital in Assam, and to the best of our knowledge, very few such studies in human have been published from this part of the country.

## MATERIALS AND METHODS

This is a retrospective study based on reports, films and digital pictures of X-rays, ultrasonography (USG), computed tomography (CT), and magnetic resonance imaging (MRI) of the patients with suspected HC attending the Department of Radiology, Gauhati Medical College and Hospital, Guwahati. The study was conducted for 1 year from June 2015 to May 2016. Ethical clearance

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was obtained from the Ethical Committee of the Medical College. Some of the patients were followed up and post-operative HC was confirmed histopathologically. In some cases, Gadolinium-based contrast was used for imaging. Siemens (800 MA) for X-ray, Siemens Acuson Antares machine for ultrasound, 16 slice Philips MX16 machine for CT scan, and 1.5 T Siemens Somatom Tim Avanto machine for MRI were used in the study.

### Inclusion Criteria

Patients with radiologically diagnosed HC other than liver and pulmonary HC.

### Exclusion Criteria

1. Patients with radiologically diagnosed liver and pulmonary HC
2. Patients with HC other than liver and pulmonary HC but refused to take part in the study.

## RESULTS AND OBSERVATIONS

Altogether, 11 cases with HC in unusual sites were included in the study.

### Case 1 - Splenic HC

A 30-year-old male patient referred to Radiology Department with abdominal pain. USG images revealed hydatid matrix filling the cyst, dependent hydatid sand and floating membrane, and detached laminated membrane (Figure 1a and b). Contrast enhanced CT (CECT) revealed unilocular cyst arising from splenic parenchyma with

small pericyst calcification (Figure 1c). Noncontrast CT (NCCT) showed increase in calcification after 3 months of albendazole therapy (Figure 1d and e).

### Case 2 - Retroperitoneal HC

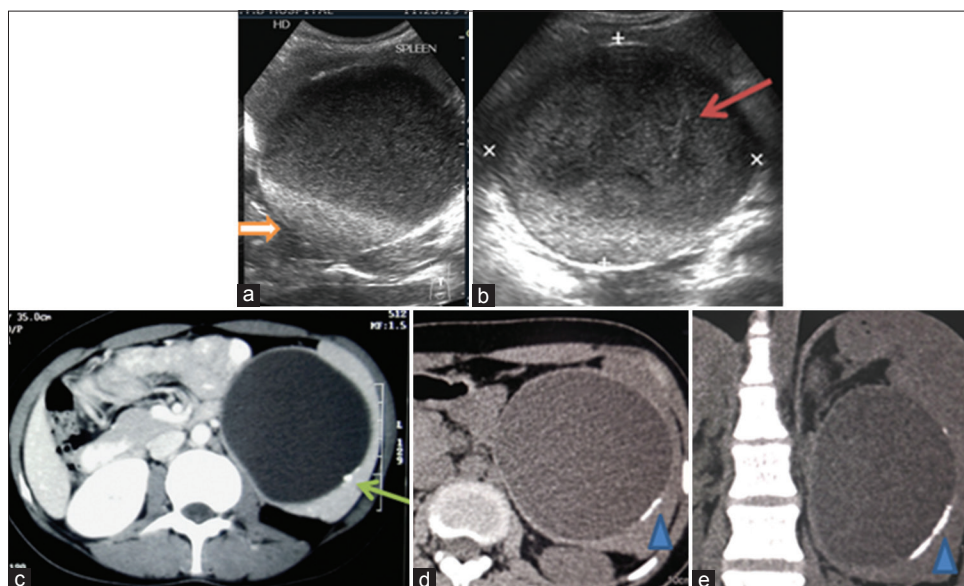
A 31-year-old male patient came with intermittent dull aching abdominal pain with vague abdominal symptoms. Axial, coronal, and sagittal CT (Figure 2a-d) sections revealed a well-defined, unilocular, homogeneous, spherical cyst in retroperitoneum, and arising from right psoas muscle. The lesion showed typical hyperdense wall on NCCT (Figure 2a) with no enhancement on CECT (Figure 2b-d).

### Case 3 - Intramuscular HC

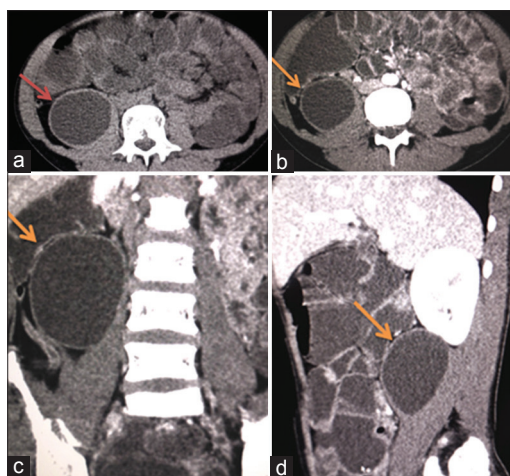
A 35-year-old male patient came with chief complain of swelling on the right thigh since 3 years. USG image shows linear serpentine structures and daughter cyst within the lesion (Figure 3a and b). Axial T1, T2W and short tau inversion recovery coronal MRI revealed multivesicular cyst in the right quadriceps femoris, with multiple daughter cysts appearing more hypointense on T1, with coiled floating membranes in cyst (Figure 3c-g).

### Case 4 - Intramuscular HC

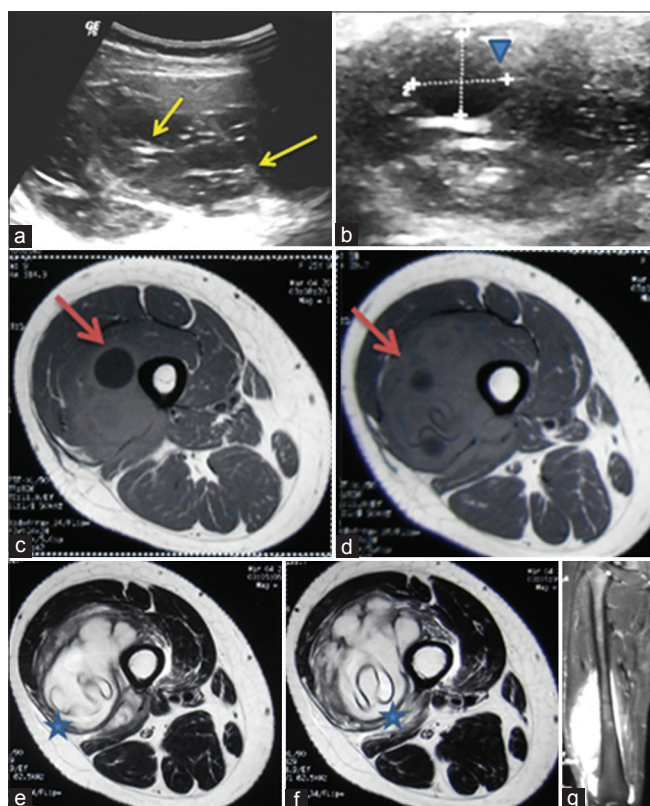
A 70-year-old male patient came with chief complaints of swelling in the left thigh since 10 years (Figure 4a). X-ray left thigh showed soft tissue swelling with no involvement of underlying bone (Figure 4b). USG revealed large heterogeneous lesion with multiple anechoic cysts within (Figure 4c). MRI revealed mixed signal intensity lesion with multiple small intralesional cysts, hypointense



**Figure 1:** Ultrasonography images show hydatid matrix filling the cyst, dependent hydatid sand (bold arrow - a) and floating membrane, detached laminated membrane (red arrow - b). Contrast enhanced computed tomography (CT) shows unilocular cyst arising from Splenic parenchyma with small pericyst calcification (green arrow - c). Non-contrast CT shows increase in calcification after 3 months of albendazole therapy (blue arrow - d and e)



**Figure 2:** Axial, coronal, and sagittal computed tomography (CT) (a-d) sections reveal a well-defined, unilocular, homogeneous, spherical cyst in retroperitoneum, arising from right psoas muscle. The lesion exhibits typical hyperdense wall on non-contrast CT (red arrow - a) with no enhancement on contrast enhanced CT (orange arrow - b-d)



**Figure 3:** Ultrasonography image shows linear serpentine structures (yellow arrow) and daughter cyst (blue arrow head) (a and b) within the lesion. Axial T1, T2W and short tau inversion recovery coronal magnetic resonance imaging show multivesicular cyst in right quadriceps femoris, with multiple daughter cysts (red arrow) appearing more hypointense on T1, with coiled floating membranes in cyst (blue star) (c-g)

on T1 (Figure 4e), and hyperintense on T2 (Figure 4d). Mild enhancement was present along walls of the cyst

(Figure 4g). The patient was operated (Figure 4f), and HC was confirmed histopathologically.

### Case 5 - Breast HC

A 35-year-old woman came with gradually progressive, painless lump in the left breast since 1½ year. USG of left breast revealed a solid appearing lesion with diffuse echoes. Posterior acoustic enhancement (Figure 5a) and floating echogenic membrane (Figure 5b) were clues to correct diagnosis. “Contained rupture” of lesion was noted from its posterior wall, with intact pericyst and focal breach in ectocyst (Figure 5c). Right breast was normal.

### Case 6 - Intracranial HC

A 40-year-old male patient came with chief complain of a severe headache and giddiness since 6 months. MRI revealed well-defined lesion in the right parietal region appears homogeneously hypointense on T1W (Figure 6a) and hyperintense on T2W (Figure 6b) sequences. MRI showing neither any enhancement (Figure 6c) nor blooming on susceptibility-weighted imaging and diffusion restriction (Figure 6d and e). The patient was operated (Figure 6f), and HC was removed.

### Case 7 - Right Myocardial HC with Splenic Hydatid

A 44-year-old female came with chief complain of chest pain and breathlessness. Cardiac CT and MRI revealed well-defined multiloculated non-enhancing cystic lesion in the right myocardium. Similar cystic lesion was present in spleen (Figure 7a-d). Coronal image showed cardiac and splenic cystic lesions (Figure 7e). Patient underwent surgical extirpation of cardiac HC followed by splenic cystectomy after 6 weeks (Figure 7f).

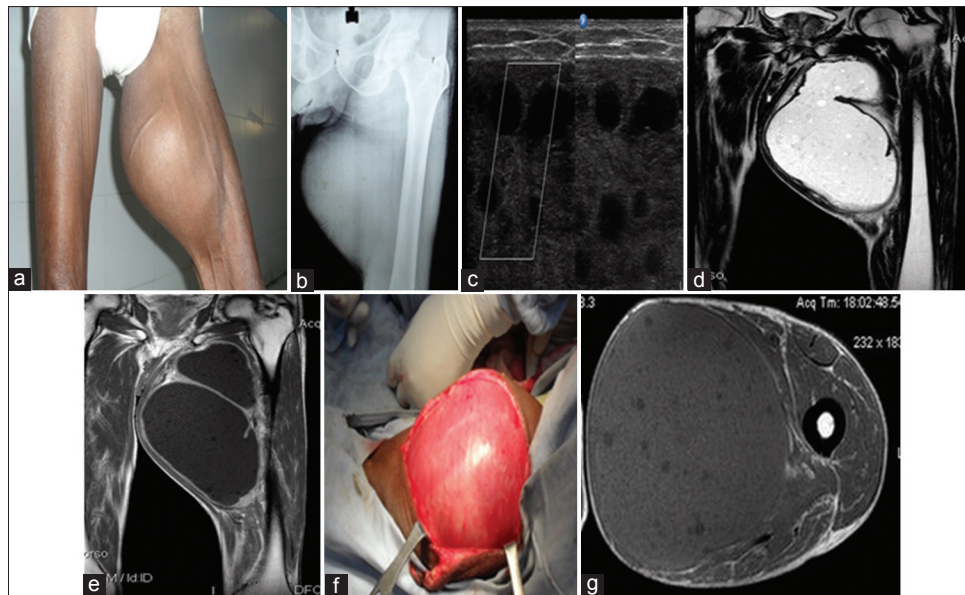
### Case 8 - Subcutaneous HC

A 50-year-old female came with chief complain of swelling over upper third of the right arm since 7 years (Figure 8a). X-ray of right shoulder showed soft tissue density lesion in upper 1/3<sup>rd</sup> of the right upper limb (Figure 8b). USG revealed heterogeneous lesion in subcutaneous plane with multiple rounded anechoic cysts within surrounded by double membrane (Figure 8c). MRI showed mixed signal intensity lesion with multiple intralesional cysts in subcutaneous plane (Figure 8d-g).

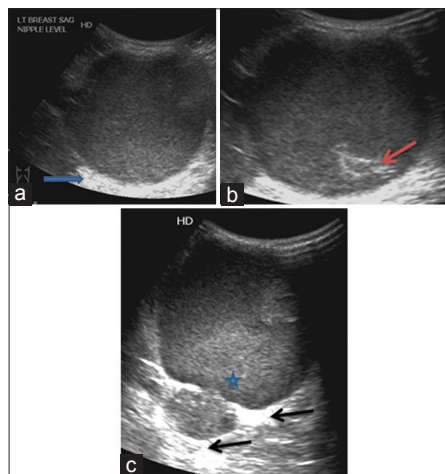
### Case 9 - Renal HC

A 35-year-old female came with lump in abdomen on the left side and hematuria. USG revealed enlarged and multicystic left kidney. Cysts showed double wall with membranous structures within (Figure 9a). CT revealed large cystic lesion with multiple daughter cysts and peripheral wall calcifications and enhancement (Figure 9b and c). MRI abdomen confirmed HCs (Figure 9d-g).





**Figure 4:** 70 years patient with left thigh swelling (a). X-ray left thigh shows soft tissue swelling with no involvement of underlying bone (b). Ultrasonography show large heterogeneous lesion with multiple anechoic cysts within (c). Magnetic resonance imaging shows mixed signal intensity lesion with multiple small intralesional cysts. Hypointense on T1 (e) and hyperintense on T2 (d). Mild enhancement along walls of the cyst (g). Intraoperative hydatid cyst (f)



**Figure 5:** Ultrasonography of left breast revealed a solid appearing lesion with diffuse echoes. Posterior acoustic enhancement (blue arrow - a) and floating echogenic membrane (red arrow - b) were clues to correct diagnosis. "Contained rupture" of lesion is noted from its posterior wall, with intact pericyst (black arrows) and focal breach in ectocyst (blue star - c)

### Case 10 - Intraperitoneal HC

A 65-year-old female came with distension of abdomen since 2 years. USG revealed multiple large well-defined intraperitoneal anechoic cystic lesions with crumpled membranes and small daughter cysts within walls of lesions (Figure 10a-c). CECT revealed multiple large ill-defined cystic lesions involving the peritoneal cavity and the right lobe of liver with peripherally arranged daughter cysts and calcifications in few lesions (Figure 10d-g).

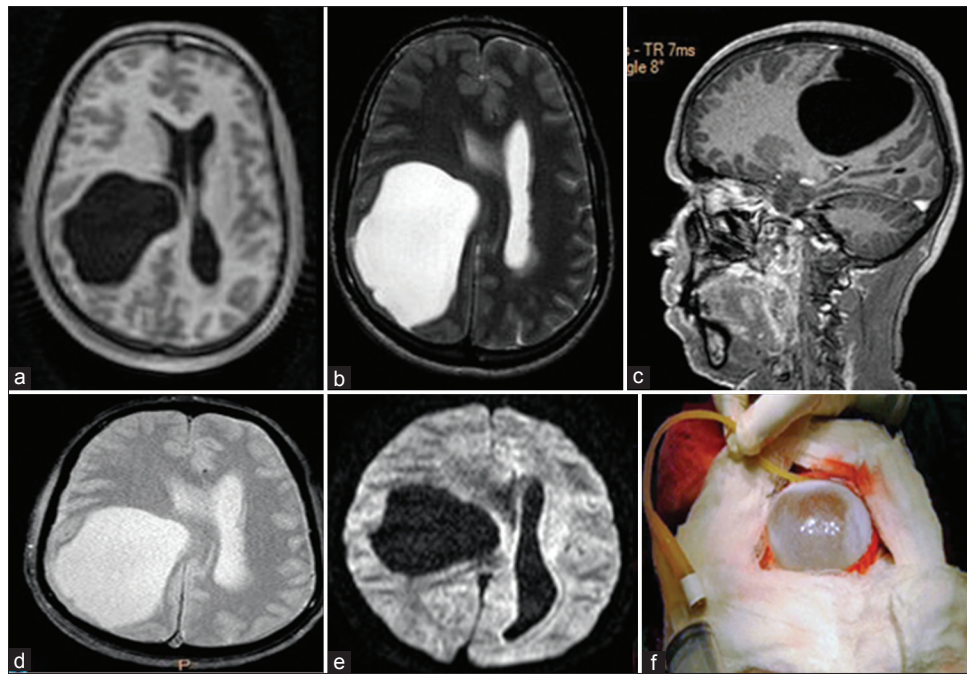
### Case 11 - Spinal HC

A 32-year-old male patient came with a history of back pain radiating to the right lower limb since 3 months. There was no history of trauma, fever, or any other last operative history. T1W plain and post contrast images showed evidence of well-defined multiloculated hypointense lesion seen involving the left hemipelvis with extension into the spinal canal and compressing the cauda equina. The lesion showed minimal peripheral enhancement on post contrast images (Figure 11a-c). T2W sagittal, axial, and coronal images revealed evidence of well-defined multiloculated hyperintense cystic lesions involving the left hemipelvis in presacral region. The lesion was extending into spinal canal and compressing the cauda equine (Figure 11d-f).

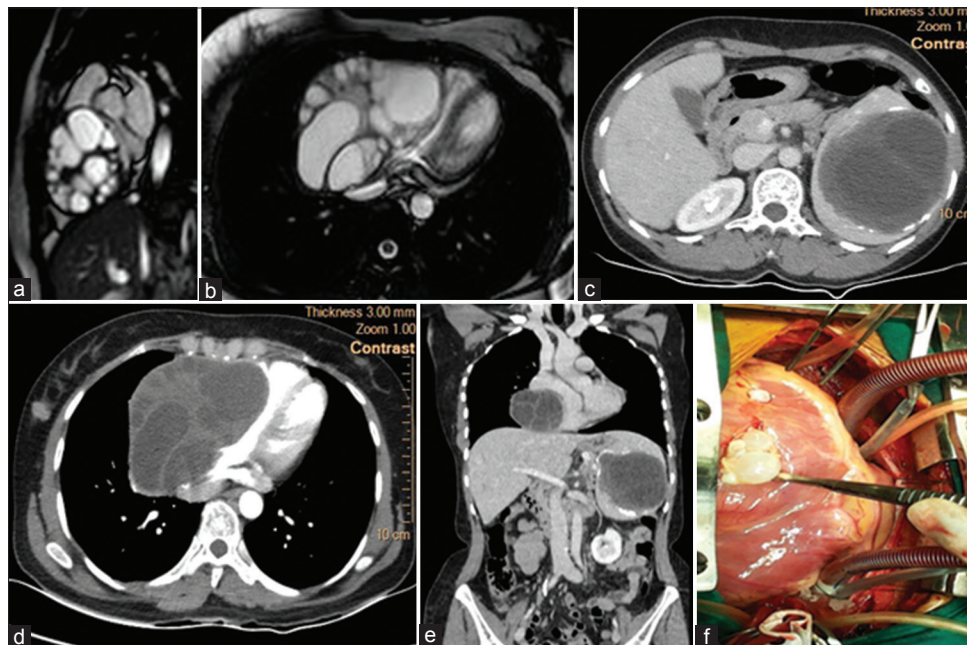
## DISCUSSION

HD is endemic in India. The annual incidence of HD per 100,000 persons varies from 1 to 200.<sup>5</sup> HC has been reported from every part of the human body such as liver, lungs, peritoneum, bones, ovaries, breast, and brain.<sup>6</sup> Although portal blood stream remains the main pathway of parasite spread, normally existing portocaval shunts, lymphatic invasion by the parasite, and retrograde migration from vena cava to subclavian vein have been documented and explain some of these rare sites.<sup>7</sup>

Spleen is the third most common site for *Echinococcus*.<sup>8</sup> Sonography is helpful, especially in the early stages, when the lesion is cystic, in detecting daughter cysts, hydatid



**Figure 6:** Well-defined lesion, in right parietal region, appears homogeneously hypointense on T1W (a) and hyperintense on T2W (b) sequences. Magnetic resonance imaging showing neither any enhancement (c - Sagittal T1 post-gadolinium contrast) nor blooming on susceptibility-weighted imaging and diffusion restriction (d - axial DWI and e - axial SWI). Intra-operative intracranial HC (f)



**Figure 7:** Cardiac computed tomography and magnetic resonance imaging showing well-defined multiloculated non-enhancing cystic lesion in right myocardium. Similar cystic lesion is present in spleen (a-d). Coronal image showing cardiac and splenic cystic lesions (e). Cardiac hydatid cyst intraoperatively (f)

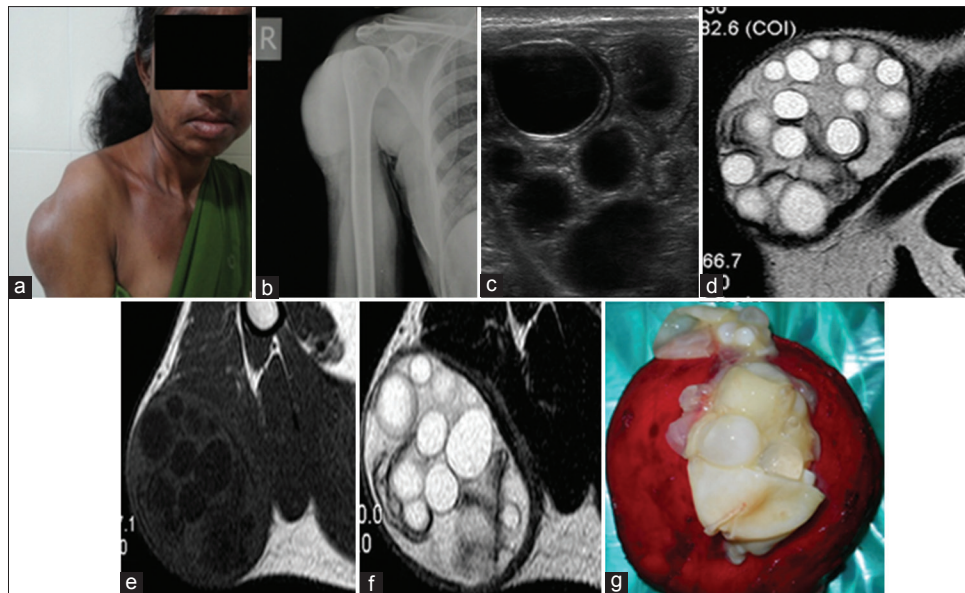
membranes, and hydatid sand.<sup>9,10</sup> When ultrasound reveals infoldings of the inner cyst wall, separation of the hydatid membrane from the wall of the cyst, or hydatid sand, a diagnosis of HD is probable.<sup>11</sup>

CT is the best mode for determining the number, size, and anatomic location of the cysts. CT may also be

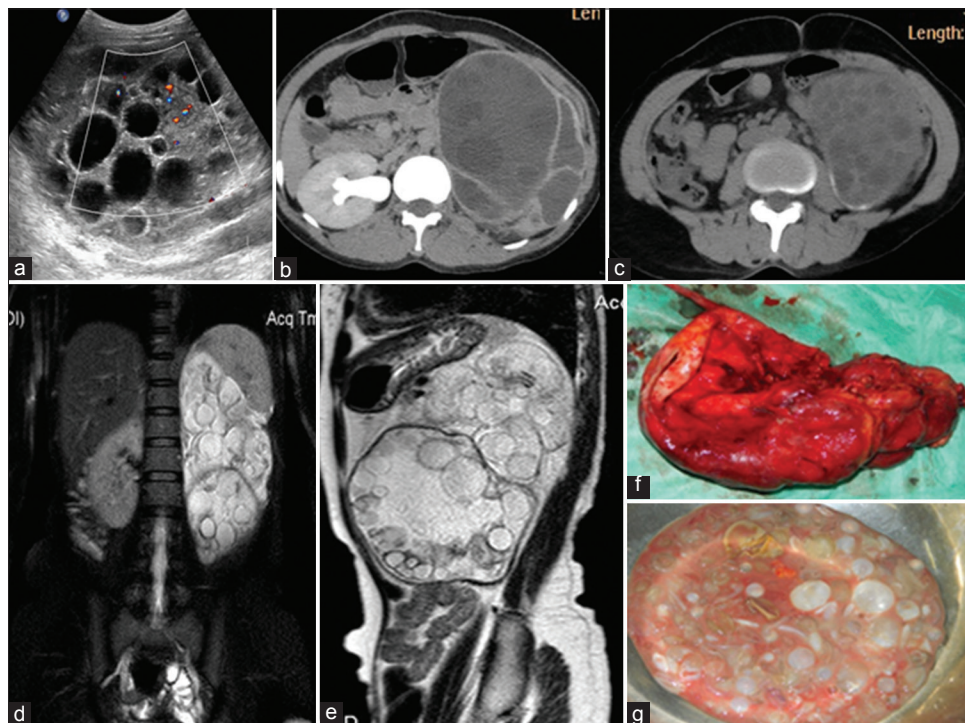
used for monitoring lesions during therapy and to detect recurrences.<sup>12</sup> Calcification is usually curvilinear or ring-like and involves the pericyst.<sup>13</sup>

Isolated retroperitoneal HC is very rare as mentioned by various literatures. Ghonge *et al.* reported a case with retroperitoneal cystic lesion in the posterior pararenal





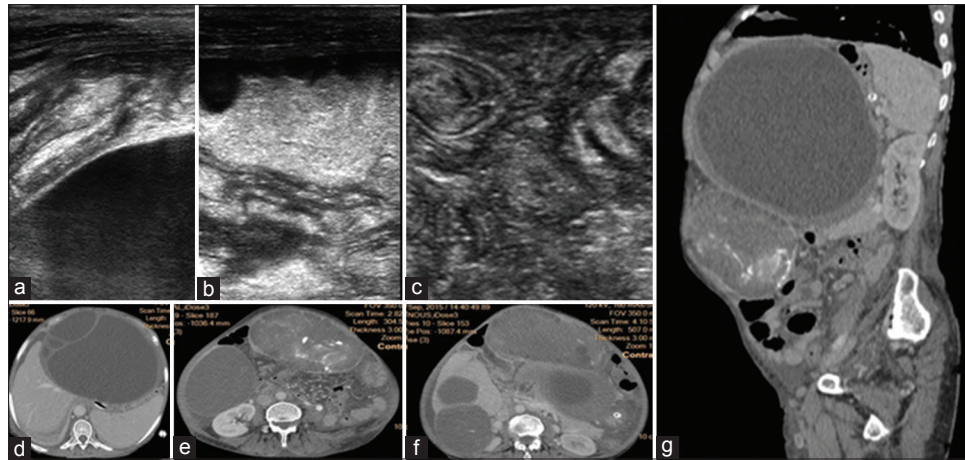
**Figure 8:** Female patient with swelling over upper third of right arm (a). X-ray of right shoulder shows soft tissue density lesion in upper 1/3<sup>rd</sup> of right upper limb (b). Ultrasonography shows heterogeneous lesion in subcutaneous plane with multiple rounded anechoic cysts within surrounded by double membrane (c). Magnetic resonance imaging shows mixed signal intensity lesion with multiple intralesional cysts in subcutaneous plane (d - T2W sagittal, e - T1W coronal, f - T2W coronal). Cut section of operative specimen showing hydatid membranes (g)



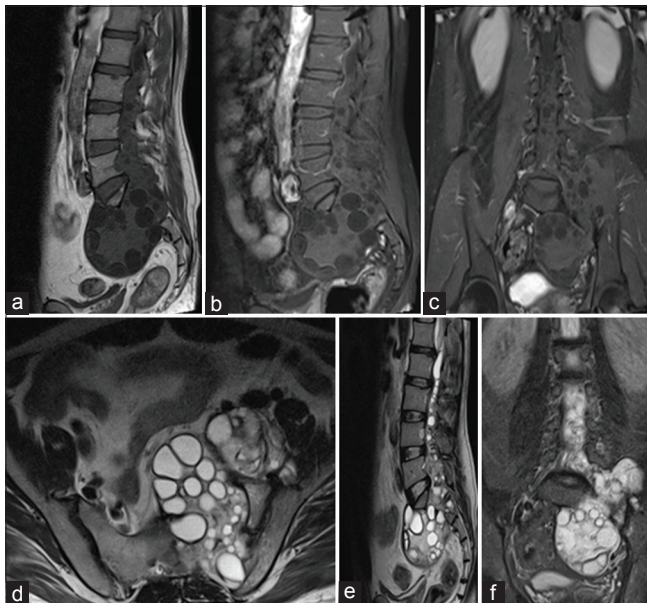
**Figure 9:** Ultrasonography reveals enlarged and multicystic left kidney. Cysts showed double wall with membranous structures within (a). Computed tomography shows large cystic lesion with multiple daughter cysts and peripheral wall calcifications and enhancement (b and c). Magnetic resonance imaging abdomen confirmed hydatid cysts (HCs) (d and e). Post-operative specimens showing multiple HCs (f and g)

space with splenomegaly. CT scan was done for further characterization of the lesion and showed a large cystic mass lesion with multiple septations posterior to the left kidney.<sup>14</sup>

Primary muscle involvement located in the skeletal muscle is very rare.<sup>15</sup> Sipahioglu *et al.* reported a case with primary HC located in the medial thigh. A multilocular cystic mass was detected in the MRI.<sup>15</sup>



**Figure 10: Ultrasonography shows multiple large well defined intraperitoneal anechoic cystic lesions with crumpled membranes and small daughter cysts within walls of lesions (a-c). Contrast enhanced computed tomography shows multiple large ill-defined cystic lesions involving the peritoneal cavity and right lobe of liver with peripherally arranged daughter cysts and calcifications in few lesions (d-g)**



**Figure 11: T1W plain and post contrast images show evidence of well-defined multiloculated hypointense lesion seen involving left hemipelvis with extension in to the spinal canal and compressing the cauda equina. The lesion shows minimal peripheral enhancement on post contrast images (a-c). T2W sagittal, axial, and coronal images show evidence of well-defined multiloculated hyperintense cystic lesions involving the left hemipelvis in presacral region. The lesion is seen extending in to spinal canal and compressing the cauda equine (d-f)**

HD of the breast is extremely rare even in endemic areas, its only accounts for 0.27% of all cases.<sup>16</sup> Recently, Kumar *et al.* reported a case of isolated HC of the breast with USG study of the breasts revealed a thick walled cystic lesion with floating membranes and internal echoes in the subareolar region of the breast.<sup>17</sup>

HD affects the central nervous system in <2% of cases.<sup>18</sup> Although in other organs they are multiple, they are usually

solitary in brain.<sup>19</sup> Although HD may be located anywhere in the brain, it is most frequently located in the middle cerebral artery territory.<sup>20</sup> MRI shows a well-defined oval or cystic mass with signal intensities similar to cerebrospinal fluid (CSF), sometimes with a low-intensity rim. MRI may also show pericyst as a halo of high signal intensity on T2-weighted images, and in some cases, perifocal edema may be detected.<sup>20</sup>

Cardiac echinococcosis is scarcely encountered with a frequency of 0.01-2%.<sup>21</sup> The most commonly affected cardiac chambers are the left ventricle (50-60% cases).<sup>22</sup> Cardiac HCs have low signal intensity on T1-weighted MRI and high signal intensity on T2-weighted images.<sup>9</sup>

Primary HD of the soft tissue is extremely rare, even in endemic areas, and accounts for 0.5-5.4% of all HD cases, with very little data on the incidence of subcutaneous echinococcosis.<sup>23</sup> Vecchio *et al.* reported a case with growing mass in the deltoid region with USG and MRI strongly suggested HC.<sup>24</sup>

Renal HC involvement is rare and constitutes only 2-3% of all patients.<sup>25</sup> Kidney involvement is usually single and located in cortex. USG shows multicystic mass and CT shows uni or multilocular cyst with well-defined walls.<sup>26</sup>

Primary peritoneal hydatidosis accounts for <2% of intra-abdominal hydatidosis.<sup>27</sup> A case of peritoneal hydatidosis was reported by Hegde and Hiremath and on the USG of the abdomen and pelvis, there was evidence of a large multicystic space-occupying lesion in the left hypochondrium and the left lumbar region. The cyst showed a central echogenic area and CECT showed a large lobulated cystic lesion in the left hypochondrium.<sup>27</sup>



Skeletal involvement with the primary HC is found in 0.5-4%. The vertebra, long bone epiphyses, ileum, skull, and ribs are affected in descending order.<sup>28</sup> HC usually begins in the vertebral body. Extension into the spinal canal results in spinal cord compression. The imaging appearance of the HC usually comprises a single spherical lesion with | clearly defined borders containing fluid with the intensity similar to that of CSF on both CT and MRI.<sup>29</sup>

## CONCLUSION

These 11 numbers of cases of HCs in unusual sites within a span of 1 year, automatically raises a finger that this North-East state is not lagging behind than other states of India in occurrence of HD even in rare sites. Moreover, radiologists should keep HCs in their mind as differential diagnosis while dealing patients with cystic lesions of this part of the country.

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