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A RARE CASE OF CYSTIC ECHINOCOCCOSIS PRESENTING WITH MACROSCOPIC HYDATIDURIA

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ABSTRACT
Hydatid cyst in urinary tract is a rarity but passage of hydatid cysts in urine is something seldom reported before. A 55-year-old lady reported to us with complaints of burning micturition and passage of white cysts in urine. The blood showed a picture of hypersensitivity while the urine showed an infective picture. The radiological evaluations and histopathological study of the cysts confirmed diagnosis of Cystic Echinococcosis in the patient. The haematological spread of the disease is perhaps the only answer to presence of the multiple cysts in rare locations as observed in this patient.

KEYWORDS: Zoonotic, Hypersensitivity, Hydatiduria.

INTRODUCTION
Echinococcosis is a zoonotic disease with humans as dead-end hosts as shown in figure 1. Patients who were detected with symptomatic disease were ranging from age of 1 to 75 years with no predilection to particular sex.[1,2] Cystic echinococcosis is usually asymptomatic unless and until complications occur. Rupture with resultant infection or anaphylaxis, fistula development with adjacent structures or mass effect on neighbouring structures are the chief mechanisms of manifestation of illness. Most cases of cystic echinococcosis result in a single cystic lesion located in a single organ. It is observed that more than 90% of cysts involve liver, lungs or both followed by occasional involvement in the kidney, spleen, peritoneal cavity, muscles and skin.[2] Other organs urinary bladder are rarely known to be the locations for cyst development. Cystic echinococcosis has a highly variable clinical presentation.[3]

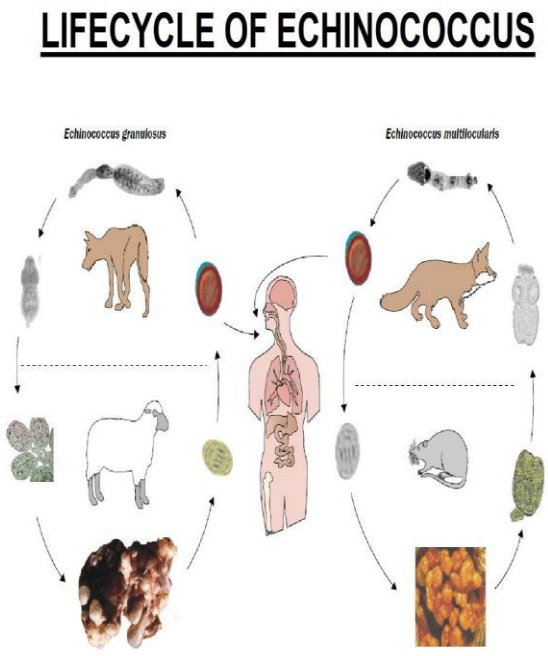

Figure. 1.

CASE REPORT

• 55-year-old lady reported with complaints of burning micturition for 1 week associated with suprapubic pain and passage of cystic grape like objects in urine for 3 days (figure 2,3). She also gave history of occasional spikes of mild grade fever. The initial evaluations revealed protein in urine (+++), numerous pus cells and WBC casts were also seen along with 2-4 epithelial cells per high powerfield.24hour urinary protein was 61.2 mg/dl (which paints a picture of urinary tract infection). Initial blood evaluation showed eosinophil 5%(which paints a picture of hypersensitivity reaction). Ultrasonography was suggestive of multiple cystic lesions of varying sizes with septa and low level echoes within it noted in abdominal cavity with impression likely hydatid cyst(figure 4). CECT abdomen revealed multiple (3) thick walled multi-loculated cystic lesion within peritoneal cavity with one in midline (91 mm x 70 mm), one near tail of pancreas (61 mm x 77 mm) and one in pelvis (80 mm x 60 mm) with impression of likely hydatid cysts(figure 5). No obvious communication with internal organs was seen. CECT chest and fundoscopy revealed normal study. Sections from cyst were acellular and showed many fragments of lamellated membranes only. No epithelial cells, scolex or hooklets seen or atypical cells were seen in the cyst. Hence the histopathological examination was also suggestive of hydatid cyst. The IgG for Echinococcus antibodies was 16.6 NTU (>11NTU is considered positive). The total IgE was detected to be 1908.8 IU/ml (>378 IU/ml is considered positive). The patient was managed conservatively with anti-infective therapy viz albendazole and the patient responded favorably. The patient was advised to follow-up for subsequent PAIR(Puncture,Aspiration,Injection,Reaspiration) but she didn't consent for the same. On her 2-monthly follow-up,she was found to be asymptomatic.

Figure. 2.

Figure. 3.

USG ABDOMEN

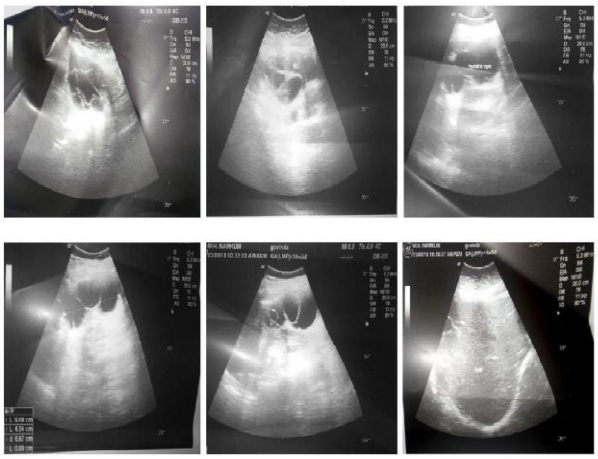

Figure. 4.

CECT ABDOMEN

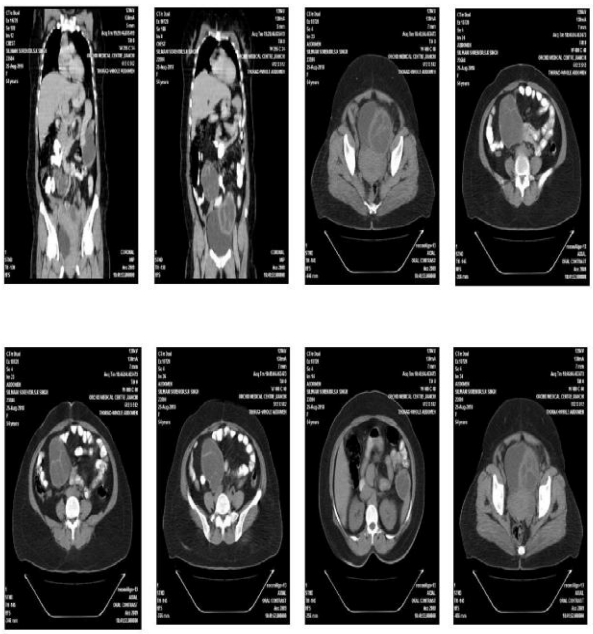

Figure. 5.

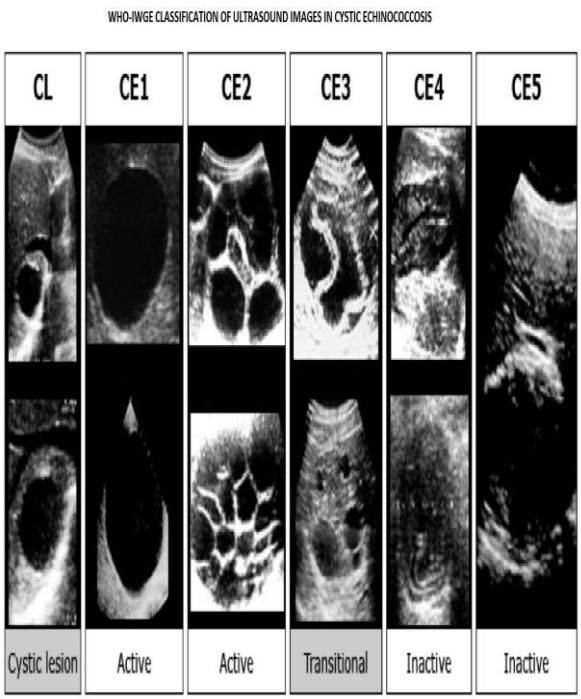

Figure. 6.

(Panel CL) Consists of unilocular, cystic lesion(s) (CL) with uniform anechoic content. The cyst wall is not clearly visible; lesions are usually round but may be oval. If these lesions are caused by cystic echinococcosis at an early stage of development, they are usually not fertile. Definitive diagnosis cannot be made by ultrasound findings alone.
(Panel CE1) Consists of unilocular, simple cyst with uniform anechoic content. The cyst wall is visible; lesions are round or oval. Cyst may exhibit fine echoes due to shifting of brood capsules called hydatid sand ("snow flake sign").
(Panel CE2) Consists of multivesicular, multiseptated cysts. The cyst wall is normally visible; lesions are round or oval. Septations produce "wheellike" structures. The presence of daughter cysts is indicated by rosette-like or honeycomb-like structures. Daughter cysts may partly or completely fill the unilocular mother cyst.
(Panel CE3) Consists of a unilocular cyst which may contain daughter cysts. Anechoic content with detachment of laminated membrane from the cyst wall may be visible as floating membrane or as "water-lily sign" which is indicative of wavy membranes floating on top of remaining cyst fluid. The cyst form may be less round because of decreased intracystic pressure. The cyst which may degenerate further or may give rise to daughter cysts.
(Panel CE4) Consists of heterogenous hypoechoic or hyperechoic degenerative contents; no daughter cysts are present. A "ball of wool" sign may be seen, which is indicative of degenerating membranes. Most cysts of this type are not fertile. Definitive diagnosis cannot be made by ultrasound findings alone.
(Panel CE5) Cysts characterized by a thick calcified wall that is arch shaped, producing a cone shaped shadow. The degree of calcification varies from partial to complete. These cysts are not fertile in most case. Definitive diagnosis cannot be made by ultrasound findings alone.

Figure. 7.

World Health Organization classification of cystic echinococcosis and treatment stratified by cyst stage

WHO stage	Description	Stage	Size	Preferred treatment	Alternate treatment
CE1	Unilocular unechoic cystic lesion with double line sign	Active	<5 cm	Albendazole alone	PAIR
			>5 cm	Albendazole + PAIR	PAIR
CE2	Multiseptated, "rosette-like" "honeycomb" cyst	Active	Any	Albendazole + either modified catheterization or surgery	Modified catheterization
CE3a	Cyst with detached membranes (water-lily sign)	Transitional	<5 cm	Albendazole alone	PAIR
			>5 cm	Albendazole + PAIR	PAIR
CE3b	Cyst with daughter cysts in solid matrix	Transitional	Any	Albendazole + either modified catheterization or surgery	Modified catheterization
CE4	Cyst with heterogenous hypoechoic/hyperechoic contents; no daughter cysts	Inactive	Any	Observation	-
CE5	Solid plus calcified wall	Inactive	Any	Observation	-

Albendazole is dosed 10 to 15 mg/kg per day in two divided doses; the usual dose for adults is 400 mg twice daily. Duration of therapy is discussed in the text.

WHO: World Health Organization; CE: cystic echinococcosis; PAIR: puncture, aspiration, injection, reaspiration.

Figure. 8.

• DISCUSSION

Echinococcal disease is caused by infection with the metacestode stage of the tapeworm *Echinococcus*, which belongs to the family Taeniidae Echinococcosis, as mentioned earlier, manifests with anaphylaxis or infection due to ruptured cysts but otherwise remains asymptomatic. Our patient reported with a mildly raised eosinophilic count with passage of cysts through urine which fits to the feature of the disease i.e. rupture of the cyst with subsequent hypersensitivity reaction. However, evidences from ultrasonography of abdomen, CE CT abdomen, histopathological study, Echinococcus specific IgG and total IgE reports confirmed our diagnosis of Cystic Echinococcosis in this patient. The ultrasonography finding was consistent with CE 3A of WHO-IWGE (World Health Organisation Informal Work Group on Echinococcosis) classification of ultrasound images of cystic Echinococcosis cysts (figure 6,7). [4]

Although papers have been published of rare incidences of cysts involving urinary tracts[5,6], the passage of cysts in urine makes this case a rarity among the rare cases seldom such cases have been reported before(hydatiduria).[7]

Another interesting feature of this case is that lungs and liver are not involved at all although they are the most commonly involved organs.

The classical pathogenesis of the disease says the organism has a haematological spread followed by cyst formation in an organ and rupture. That explains the 3 unrelated cysts that were detected on CECT abdomen and the multiple cysts passed from urine with no obvious communications with the organs was found.

The most probable explanation for the hypersensitivity picture of the blood and infective picture of the urine is the rupture of the cysts that had developed in the urinary bladder following which the cysts were excreted via urine.

Since the patient fit into the category CE3A as per WHO IWGE classification of ultrasound images of cystic echinococcosis, she was planned to be managed with albendazole therapy followed by PAIR(Puncture, Aspiration, Injection, Reaspiration) as per WHO guidelines (figure 8).[8]

CONCLUSION

Medical science never fails to surprise us as in this case where the disease was found in the place where its least occurring while its most common site of occurrence remaining absolutely free and untouched.

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