

Unusual Locations of the Hydatid Cyst: A Review from Iran

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Abstract

Hydatid disease is caused by *Echinococcus granulosus* and is endemic in many parts of the world, including Iran. This parasitic tapeworm can produce cysts in almost every organ of the body, with the liver and lung being the most frequently targeted organs. However, the cyst tends to appear in different and sometimes unusual body sites in various geographical areas of the world.

This review provides information on the reported cases of the unusual body sites of the hydatid cyst from Iran in the last 20 years. A literature search was performed through PubMed, Scopus, Google Scholar, IranMedex, Society Information Display (SID), Magiran, and Irandoc using the keywords of “hydatid cyst and Iran” and “*Echinococcus granulosus* and Iran”, and 463 published cases of the hydatid cyst in unusual body sites from Iran were reviewed, evaluated, and discussed. The most common locations were the central nervous system (brain, spinal cord, and orbit), musculoskeletal system, heart, and kidney, while some less common locations were the spleen, pancreas, appendix, thyroid, salivary gland, adrenal gland, breast, and ovary.

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Introduction

The hydatid cyst is a zoonosis caused by adult or larval stages of tapeworms belonging to the genus *Echinococcus granulosus*.¹ The tapeworm stage is harbored in the intestine of carnivores such as dogs, which constitute the definitive host,² and the eggs are passed in the feces of the infected carnivores and ingested by herbivores such as sheep, which comprise the intermediate host. Humans are the incidental intermediate host. Larvae emerge from the eggs in the intestine; and after invasion to the blood vessels, they can migrate into almost every part of the body.³ The usual destination is the liver via the portal tract, but sometimes the larvae pass through the liver barrier and reach the lungs and all the other viscera, where they transform into small cysts.⁴

Echinococcosis/hydatidosis is one of the most important zoonotic diseases inasmuch as it occurs in different parts of Iran.⁵ Adult worms have been recovered from dogs, jackals, and wolves, but human cases have been reported from hospital archives by pathological reports of surgically proven cases in different

geographical areas of the country.⁶

In nearly all the previous reports, the liver was the most common location of the hydatid cyst, followed by the lung, with the approximate occurrence rates of 70% and 12%, respectively.^{7,8} There is a small number of reports of higher incidence rates of lung involvement in Iran, but such cases are very unusual.^{9,10} The reported incidence in children has been a point of controversy in a few previous investigations, reporting incidence rates of 41-70% for the lung and 43-48% for the liver hydatid cyst.^{11,12}

Although most reported Iranians with Echinococcosis had cysts in their lungs and livers, more unusual cyst locations were also recorded.^{9,13} In a few previous reviews on hydatidosis from Iran, unusual body sites such as the heart, orbit, brain, muscle, salivary gland, bone, urinary tract, and pancreas were reported.⁹

The aim of this paper is to provide an overview of the published cases of the hydatid cyst in unusual body sites from Iran to delineate the most important demographic findings and locations of the disease in this hyperendemic country.

Methods

The published cases of the hydatid cyst in unusual body sites from Iran were reviewed via a search in PubMed, Scopus, Google Scholar, IranMedex, Scientific Information Database (SID), Magiran, and Irandoc (1990-2011), using the keywords of "hydatid cyst and Iran" and "Echinococcus granulosus and Iran".

The following inclusion criteria were employed:

- 1) Articles must be written in English and Farsi;
- 2) Articles must have been published between 1990 and 2011;
- 3) Studies must be from Iran and contain case report(s), diagnosing the hydatid cyst in unusual locations (i.e. other than the liver and lung); and
- 4) Cases must have been pathologically confirmed postoperatively.

Results

In the last 20 years, about 463 cases of the hydatid cyst located in different parts of the body, excluding the liver and lung, have been published from Iran. Table 1 depicts the details of the published cases.

The most common locations were the central nervous system (brain, spinal cord, and orbit), musculoskeletal system, heart, and kidney, whereas some less common locations were the spleen, pancreas, appendix, thyroid, salivary gland, adrenal gland, breast, and ovary. Other cases such as retroperitoneal and mediastinal hydatid cysts were also reported.

Most of the published cases were reported from Tehran (as a referral center for the whole country); nevertheless, other centers such as Khorasan, Azerbaijan, Fars, Isfahan, and Yazd also reported unusual locations of the hydatid cyst.

Central Nervous System

In the last 20 years, about 256 cases of the hydatid cyst in the brain, spinal cord, and orbit have been reported from different geographical areas

Table 1: The published cases of the hydatid cyst with unusual locations from Iran

Locations	Numbers	Most common clinical manifestations	References
Intracranial	256	Headache	14-37
Spinal		Low back pain	
Orbit		Visual impairment	
Musculoskeletal	57	Swelling Pathologic fracture	38-66
Cardiovascular	42	Angina, dyspnea and palpitation, pressure effect	67-82
Kidney and Urinary Tract	31	Flank pain	83-90
Spleen	20	Left upper quadrant pain	91-94
Ovary	11	Ovarian mass	95-103
Uterus		Lower abdominal pain	
Fallopian Tube			
Pancreas	6	Epigastric pain	104-109
Salivary Gland	9	Painless swelling	110-118
Breast	8	Breast mass	119-125
Thyroid	4	Thyroid enlargement	126-129
Adrenal	2	Flank pain	130-131
Appendix	1	Abdominal pain	132
Mediastinum	7	Pressure effect on adjacent organs	133-139
Omental, Mesenteric, Retroperitoneal	7	Mostly asymptomatic	140-146
Parapharyngeal	1	Nonspecific	147
Nasolabial	1	Nonspecific	148
Total	463	-	-

of Iran.¹⁴⁻³⁷ There are two reviews by Abassioun et al.^{14,15} who reported 69 cases of the brain hydatid cyst. These patients were 3 to 50 years of age, with a slight male preponderance.¹⁴ Among these 69 reported cases, 5 cysts were in the posterior fossa, 2 in the cerebellum, one in the CP angle, one in the fourth ventricle, one in the pons, and 59 cases in the brain parenchyma.¹⁴ The hydatid cyst of the orbit in the above-mentioned review was detected in 28 patients, with an age range of 5 to 54 years.¹⁵

Abassioun et al.¹⁵ also reported 36 cases of the spinal hydatid cyst, both intra and extradural, 20 of which were male and 16 cases were female patients.

Apart from the above reviews, 105 other intracranial hydatid cysts were reported in 73 males and 32 females, with an age range of 5 to 60 years.^{6,7,9,16-28} Most of the intracranial hydatid cysts were within the brain hemisphere,¹⁸ and the most common presenting symptoms were headache and vomiting.

As a rule, the hydatid cyst of the brain tends to be solitary and spherical.¹⁴ Serologic tests are not diagnostic, and imaging studies such as computed tomography (CT) scan and magnetic resonance imaging (MRI) are necessary for preoperative diagnosis.¹⁴

There were 11 cases of the spinal hydatid cyst; they were all adults above 20 years of age^{2,29-33} and presented with signs and symptoms related to cord compression such as low back pain, radicular pain, and paraparesis.²⁹ The majority of the spinal hydatid cysts were extradural, and primary intradural hydatid cysts were very rare.³⁰

Aside from the aforementioned review, the orbital hydatid cyst was rarely reported from Iran: there were only 8 cases, all presenting in childhood.³⁴⁻³⁷ The reported symptoms were visual impairment and proptosis,³⁵ and anatomically most of the orbital cysts were in the intraconal space because most branches of the ophthalmic artery supply the intraconal space.³⁶

Musculoskeletal System

In the last 20 years, the skeletal hydatid cyst has been reported in 44 patients,³⁸⁻⁵⁷ comprised of 28 males and 16 females with an age range of 5-71 years (mean age=41.5 years). The locations of the skeletal hydatid cysts were varied such as the maxillary sinus,³⁸ mandible,³⁹ knee,⁴⁰⁻⁴¹ long bones,⁴²⁻⁴⁶ and ilium.⁴⁷⁻⁵² Other less common locations were the chest wall and vertebra.⁵³⁻⁵⁴

The clinical manifestations of the osseous hydatid cyst may take a long time to become obvious, and that is when the cyst is detected by swelling, pathologic fracture, and secondary infection.⁵³⁻⁵⁷ The bone hydatid cyst is polycystic

in contrast to other non-osseous locations, which is because of the absence of adventitia around the cyst.⁵³ The diagnosis of the osseous hydatid cyst is based on imaging modalities such as CT scan.⁵⁵ Serologic studies are usually negative and unreliable for preoperative studies.⁵⁶

The hydatid cyst involvement of the skeletal muscle is even less common than that of the bone. In our review of Iranian cases, we found 11 reported patients, 8 males and 3 females with an age range of 22-80 years (mean age=29 years), with the hydatid cyst of the skeletal muscle.^{7,58-66} The reported locations were in the latissimus dorsi,⁵⁸ gluteal muscle,^{59,60} cervical muscles of the paraspinal area,^{60,61} and thigh.⁶³⁻⁶⁶ The most common presenting symptoms were painless swelling,⁵⁸ causing symptoms secondary to the compression effect on the adjacent organs.⁶⁰ Radiological studies, including MRI, are the mainstay of the preoperative diagnosis of the skeletal muscle hydatid cyst.⁶¹

Cardiovascular System

The third most common unusual location of the hydatid cyst reported from Iran is the cardiovascular system, with 42 cases having been reported in the last 20 years.⁶⁷⁻⁸² The cases comprised 25 males and 17 females with an age range of 8 to 73 years (mean age=29.5 years). Most of the cardiac hydatid cysts were located in the ventricular wall,⁶⁷⁻⁷⁶ and the most common presenting symptoms were angina, dyspnea, and palpitation, in consequence of the pressure effects of the cyst on the coronary and conducting system.⁷⁰ Some less frequent symptoms related to pericardial tamponade were also reported.⁷² The hydatid cyst of the interatrial and interventricular septum was rarely reported.⁷⁸⁻⁷⁹ Likewise, only 2 cases of the intrapericardial,⁸⁰ and endocardial,⁸¹ hydatid cysts were reported from Iran.

The vascular hydatid cyst in the aorta and superior vena cava with invasion to the myocardium was reported in a study from Iran.⁸¹

There were reports of very infrequent asymptomatic cases of the hydatid cyst of the heart detected during EKG evaluations for another surgery.⁷¹

Serologic tests are positive in about 50% of the patients,⁷¹ but transesophageal echocardiography (TEE) is known as the imaging procedure of choice for the diagnosis of the cardiovascular hydatid cyst.⁶⁹

Kidney and Urinary Tract

Our investigation yielded 31 published cases, 23 males and 8 females with an age range of 9 to 73 years (mean age=44 years), of the hydatid cyst of the kidney and urinary tract.^{6,83-90} Among

these cases, 29 patients had the renal hydatid cyst and 2 had the bladder wall hydatid cyst. The most common clinical symptom was flank pain.⁸⁴

There is no serologic and immunological test pathognomonic for the diagnosis of the renal hydatid cyst, but ultrasonography and, in particular, CT scan can be of great help.⁸⁴⁻⁸⁹

Spleen

There were 20 cases of the splenic hydatid cyst from Iran in 13 males and 7 females with the reported age ranging from childhood to 75 years.^{6-8,91-94} The splenic hydatid cyst exhibits a variety of clinical features, requiring a high index of suspicion for diagnosis.⁹² The most common reported symptoms at presentation were the left upper quadrant pain.⁹²

The best diagnostic methods are ultrasonography and CT scan.⁹²

Uterus, Ovary, and Fallopian Tube

There were 9 published cases, with a mean age of 50 years (mean age=34-84 years), of the ovarian hydatid cyst from Iran.^{7,95-102} Most of the reported cases of the ovarian hydatid cyst were bilateral. The isolated hydatid cyst of the fallopian tube was very rarely reported.¹⁰³

The uterine hydatid cyst is extremely rare, and only one case was reported from Iran with the accompanied involvement of the fallopian tube in a 25-year-old female, who presented with lower abdominal pain. The diagnosis was made after laparotomy for the evaluation of the cause of the symptoms.¹⁰³

The most popular methods of diagnosis are ultrasonography, CT scan, and MRI, all of which are much more sensitive than immunologic tests.¹⁰²

Pancreas

In the last 20 years, 6 patients, 4 males and 2 females with a mean age of 34.5 years, have been reported with the pancreatic hydatid cyst.^{6,104-109} This cyst usually manifests as an epigastric mass, recurrent acute pancreatitis, chronic pancreatitis, and obstructive jaundice.¹⁰⁶ Complications of the pancreatic hydatid cyst depend on the relationship between the cyst and the pancreatic duct.¹⁰⁶ The methods of choice for the diagnosis of the pancreatic hydatid cyst are CT scan and MRI.¹⁰⁶

Salivary Gland

There were 9 published cases, 4 males and 5 females with a mean age of 16.5 years, of the hydatid cyst of the salivary gland: 7 in the parotid gland and 2 in the submandibular gland.¹¹⁰⁻¹¹⁸ The most common presenting symptoms were

progressive and painless swelling.¹¹⁰ It has been stated that all hydatid cysts of the parotid gland are primary.¹¹¹

Breast

Eight cases of the breast hydatid cyst were published from Iran,^{6,119-125} all in the female breast with a median age of 40.7 years. The most common presenting symptom was a well-defined palpable breast mass, which can be confirmed by mammography and ultrasonography.¹¹⁹

Thyroid

In the last 20 years, only 4 cases of the thyroid hydatid cyst have been reported from Iran, all in females between 17 and 35 years of age (mean age=14.3 years).¹²⁶⁻¹²⁹ The patients with the thyroid hydatid cyst presented with pressure symptoms and signs of dyspnea, hoarseness, goiter, and dysphagia.¹²⁹ Clinically, the thyroid hydatid cyst presents with a solitary mass, mimicking a thyroid cystic nodule.¹²⁷ The diagnosis can be made by fine needle aspiration (FNA) and isotope scanning.¹²⁸

Adrenal

The adrenal hydatid cyst in Iran was reported in only 2 cases: a 49-year-old female and a 42-year-old male.^{130,131} The adrenal hydatid cyst is mostly asymptomatic and is incidentally found by imaging; on rare occasions, however, it can cause hypertension.¹³⁰ Another case was reported, presenting with vague flank pain with a primary diagnosis of a renal tumor, for which surgery was undertaken.¹³¹

Appendix

There was only one reported case of the appendiceal hydatid cyst from Iran, diagnosed after laparotomy in a 47-year-old male worker presenting with vague abdominal pain.¹³²

Mediastinum

Seven cases, 5 males and 2 females with a mean age of 28.7 years, of the mediastinal hydatid cyst were reported from Iran.¹³³⁻¹³⁹ The symptoms related to the site of the pressure effect.¹³⁹

Omentum and Retroperitoneum

Seven cases of the mesenteric, diaphragmatic, omental, pelvic, and retroperitoneal hydatid cyst have been reported from Iran in the last 20 years.^{6,140-146} These cases may remain asymptomatic until reaching a large size,¹⁴⁰ and the clinical signs vary according to the site.

The parapharyngeal hydatid cyst in a 41-year-old female,¹⁴⁷ and the nasolabial hydatid cyst in an 11-year-old adolescent,¹⁴⁸ were the last two

extremely rare case reports in this review from Iran.^{147,148}

Discussion

Hydatid disease is a unique parasitic disease that is endemic in many parts of the world.¹⁴⁹ This parasitic disease is a significant public health concern in Iran, as an endemic country,¹⁵⁰ rendering a review of the published cases of hydatid disease from this hyperendemic country vitally important.

In hydatid disease, the liver and lung are the most common involved organs, but the disease can be seen in any organ of the body.¹⁵¹ The rates of the localization of hydatid disease in different body organs vary in the literature.¹⁵²

All the published cases in Iran included in this review are based on hospital experiences proven postoperatively by pathological examination. Our results demonstrated that the most common locations of the hydatid cyst, after the lung and liver, were the central nervous system, orbit, musculoskeletal system, cardiovascular system, kidney, and urinary tract. There were also reports of the spleen, uterus, ovary, pancreas, salivary gland, breast, adrenal, appendix, mediastinum, omentum, and retroperitoneum hydatid cysts.

The clinical manifestations in the hydatid cyst of most parts of the body are too nonspecific to make a diagnosis based on the signs and symptoms before surgery.¹⁴⁹⁻¹⁵⁴ In all of the previous reports from Iran and all around the world, it has been shown that serologic tests have many false-negative results, but imaging modalities such as ultrasonography, CT scan, and MRI have been the methods of choice, especially the latter, which has been the diagnostic method of choice for the preoperative diagnosis of the hydatid cyst in most unusual locations.¹⁵³

The best treatment for the hydatid cyst is surgical excision, accompanied by postoperative medical therapy.¹⁵¹

The next part of this review presents the salient points of each unusual site of the hydatid cyst extracted from the most recently published literature.

Central Nervous System, Spinal Cord, and Orbit

The cerebral and spinal cord hydatid cysts are very rare. Indeed, the existing literature contains about 300 articles,¹⁵⁵ accounting for 2-3% of all the cases of hydatidosis.¹⁵⁶

The most common location is the intraparenchymal supratentorial, and the most common presenting symptoms are headache and weakness in the previous reports from other parts of the world,¹⁵⁵⁻¹⁵⁷ which is very similar to the cases published from Iran.

In the last 20 years, 256 cases of the central nervous system hydatid cyst have been published from Iran. This cyst site accounted for the third common site of the hydatid cyst after the lung and liver.

The hydatid cyst of the spinal cord is less common. According to the recent literature, this cyst accounts for about 1% of all the cases of the hydatid cyst.²⁹ In this location, the intravertebral discs are usually preserved because the disease tends to progress beneath the periosteum and ligaments.²⁹

The orbital hydatid cyst accounts for about 1-2% of the cases in the previous literature and is most commonly detected in childhood.¹⁵⁸ Our survey yielded 36 cases of the orbital hydatid cyst published from Iran.^{15,26,27}

Musculoskeletal System

Osseous hydatid disease and muscular hydatidosis are uncommon and account for 0.5-4% and 0.5-2.5% of all hydatidosis cases, respectively (in endemic areas).¹⁵⁹ The most common locations of the osseous hydatid cyst are the vertebra, pelvis, and long bones in the previous records from other parts of the world.⁴¹ However, in the published cases from Iran, there were 55 cases with variable locations such as the long bone, mandible, maxilla, and pelvis.³⁸⁻⁵⁴

Muscle involvement of the hydatid cyst is reported as an uncommon location, because of high lactic acid, which is not a suitable environment for the parasite.⁵⁸

Cardiovascular System

The heart and large blood vessels also have been reported as the common unusual body sites of the hydatid cyst in endemic areas of the world, accounting for 0.5-2% of all the reported cases.¹⁶⁰

The diagnostic method unique for this part of the body is echocardiography, which has been claimed as the method of choice for the diagnosis of the cardiac hydatid cyst. Nonetheless, CT scan and MRI are also helpful in other parts of the body.¹⁶¹

Kidney and Urinary Tract

The kidney is the most common location in the urinary tract and has been reported in about 2-3% of all cases of the hydatid cyst.⁸³ In many of the previous reports from other parts of the globe, the kidney is reported as the third common site of the hydatid cyst after the liver and lung.¹⁶²⁻¹⁶⁴ In our survey of the published cases from Iran, however, the renal hydatid cyst was the fourth most common location of the hydatid cyst.

The clinical symptoms are nonspecific, and the only interesting and diagnostic symptom reported is hydatiduria.¹⁶³

The hydatid cyst of the urinary bladder is even less common, and only 2 cases were published from Iran.^{6,86} This cyst can also present with hydatiduria and is, otherwise, extremely difficult to diagnose before surgery.¹⁶⁴

Spleen

Less than 2-5% of the cases of the hydatid cyst have been reported from the spleen.¹⁶⁵ There were 20 cases of the splenic hydatid cyst published from Iran.⁹¹⁻⁹⁴ Many of the reported splenic hydatid cysts worldwide are asymptomatic, and a very small number of patients show nonspecific left upper quadrant pain.⁹⁴

Uterus, Ovary, and Fallopian Tube

The ovary is the most common site of hydatidosis in the female genital tract, but overall it is extremely uncommon (less than 1%). This cyst usually presents like a malignant tumor.¹⁶⁶ The clinical presentations are very nonspecific, and making a correct preoperative diagnosis is very difficult.¹⁶⁷ There were only 9 cases of the ovarian hydatid cyst published from Iran.⁹⁵⁻¹⁰²

The uterine hydatid cyst is even less common than ovarian hydatidosis, and its occurrence is exceptional.¹⁶⁸ The clinical presentation of this cyst is also very nonspecific, and it is difficult to diagnose the cyst before surgery.¹⁶⁹ Only one case of the uterine hydatid cyst was reported from Iran.¹⁰³

The hydatid cyst of the fallopian tube is most often accompanied by the ovary hydatid cyst and can cause infertility and spontaneous rupture of the tube.¹⁷⁰

Pancreas

The pancreatic hydatid cyst is very uncommon and accounts for less than 1% of the cases. It occurs mostly in the head of the pancreas.¹⁷¹

Salivary Gland

The hydatid cyst of the parotid and submandibular gland is very rarely reported.^{172,173} There are reports from Iran and other parts of the world about the diagnosis of the salivary gland hydatid cyst via FNA cytology. This cyst is reported to be capable of causing anaphylaxis and dissemination, but there are increasing numbers of reports on the diagnosis of the salivary gland hydatid cyst via FNA, without any complications.¹¹⁸

Breast

The breast involvement of the hydatid cyst is rare, with a reported incidence of 0.27%.¹⁷⁴ Mammography, CT scan, and MRI can help to diagnose the breast hydatid cyst before surgery. However, there are rare case reports of

preoperative diagnosis by FNA cytology without complications.¹²⁰ Our review demonstrated 8 cases of the breast hydatid cyst reported from Iran.¹¹⁹⁻¹²⁵

Thyroid

The hydatid cyst of the thyroid is very rare and clinically presents like a simple colloidal cyst.¹⁷⁵ There were only 4 cases reported from Iran.¹²⁶⁻¹²⁹ For all the reports of the role of FNA in the diagnosis of the thyroid hydatid cyst, as is the case in the salivary gland and breast, its application is controversial.¹⁷⁵

Adrenal

0.06% of the cases of the hydatid cyst have been reported in the adrenal.^{176,177} The adrenal hydatid cyst is most often asymptomatic, but reports of hypertension are also available.¹⁷⁷ In our survey, only 2 cases of the adrenal hydatid cyst were reported from Iran; both patients underwent surgery with the impression of the adrenal cyst.^{130,131}

Appendix

The hydatid cyst of the appendix is exceptional, and fewer than 10 cases have been reported worldwide. Most of the reported cases presented with acute appendicitis.¹³²

Mediastinum

The mediastinal hydatid cyst is uncommon but it should be included in the differential diagnosis of the mediastinal cyst in endemic parts of the world.¹⁷⁸

Our findings yielded 7 cases from Iran, presenting with cardiac or respiratory problems.¹³³⁻¹³⁹

Omentum and Retroperitoneum

The omental and retroperitoneal hydatid cysts are very uncommon, but these cysts can become huge in size.¹⁷⁹

Conclusion

The hydatid cyst can present in any part of the body and no site is immune. These unusual locations often produce nonspecific symptoms; consequently, it is advisable that the hydatid cyst be considered in the differential diagnosis of all cysts of the body, especially in endemic countries such as Iran.

Conflict of Interest: None declared.

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