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Multi-sited hydatid cystic disease with muscular involvement in a young female: A case report from Syria



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ABSTRACT

INTRODUCTION: Hydatidosis is a unique disease caused by a tapeworm called *Echinococcus granulosus*. Musculoskeletal involvement with hydatid cystic disease accounts for less than 5% of all cases. The main purpose of this study is to present a case of multi-sited hydatid cystic disease and how to manage it.

PRESENTATION OF CASE: Here we present a case of 14-year-old girl complained of a gradually enlarging mass on her left shoulder with a final diagnosis of triple-sited hydatid cystic disease including the right lung, the left trapezius muscle and the liver.

DISCUSSION: The diagnosis was made by non invasive radiological procedures (CT scan and MRI). The management consisted of aspiration and reinjection of hypertonic solution to the lung cyst followed by complete surgical resection, enucleation of the muscular lesion and conservative management of the liver lesion. After Three months of follow up there was no recurrence at the sites of operation and the liver cyst reduced in size.

CONCLUSION: Multi-sited hydatid cysts could be treated through one stage surgery followed by chemotherapy with benzimidazoles.

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1. Introduction

The present work has been reported in line with the SCARE criteria [1]. Echinococcosis is an infectious disease caused by the cestode *Echinococcus* commonly affecting liver and lungs [2]. The liver is affected in 75% of cases, the lungs in 15%, and the remainder to the rest of the body [3]. Musculoskeletal involvement with hydatid cystic disease accounts for less than 5% of all cases [4]. This low incidence is related to the filtering function of the liver and lungs which serve to prevent the echinococcus from entering the systemic circulation [5], and the high lactic acid concentration and the contractile activities of skeletal muscles [6,7]. Although most cases are asymptomatic, symptoms occasionally result from compression of adjacent structures or other complications [8].

We present a case from Almwassat hospital, Damascus University which is about a young girl with triple-sited hydatid cystic disease in the right lung, left trapezius muscle and liver. Triple cysts at the same organ (the liver) was mentioned in the literature [9], however the localization of cysts mentioned in our case has not been reported before. In our experience this is a rare case of multi-sited hydatidosis which was managed surgically followed by conservative treatment with benzimidazoles.

2. Presentation of case

A previously healthy 14-year-old girl with no medical, social or surgical history complained of a gradually enlarging mass on her left shoulder over the period of two months, with no pain or constitutional symptoms. On examination, there was a 5 × 6 cm cystic mass at the upper border of her left scapula, with soft and intact overlying skin. The cyst was freely mobile with no signs of localized inflammation. Clinical examination also revealed markedly decreased breath sounds over the lower right lung field. The serological tests were within normal limits. chest x-ray showed a rounded regular density in the right lower lobe (Fig. 1). Computed tomography confirmed the presence of a 10 × 8 cm cystic mass within the lower lobe of the right lung (Fig. 2A), a 5.5 × 4.5 cm cystic mass within the muscular components over the left scapula, and a 2 × 1.5 cm cystic mass within the right lobe of the liver (Fig. 2B). T1-weighted MRI images of the left shoulder cyst revealed a low signal intensity cystic mass with a low intensity rim and high signal intensity on T2-weighted images within the thickness of left trapezius muscle (Fig. 3C and D).

Decision was made to manage both lung and shoulder cysts surgically with one operation following 2 weeks of medical treatment with albendazole because of the extrahepatic involvement of the former and the superficial location prone to trauma of the latter. The right lung lesion was approached firstly through a right thoracotomy in the 5th inter-costal space, and the 10 × 6 cm cystic mass in the right lower lobe (Fig. 4) was excised by means of aspira-

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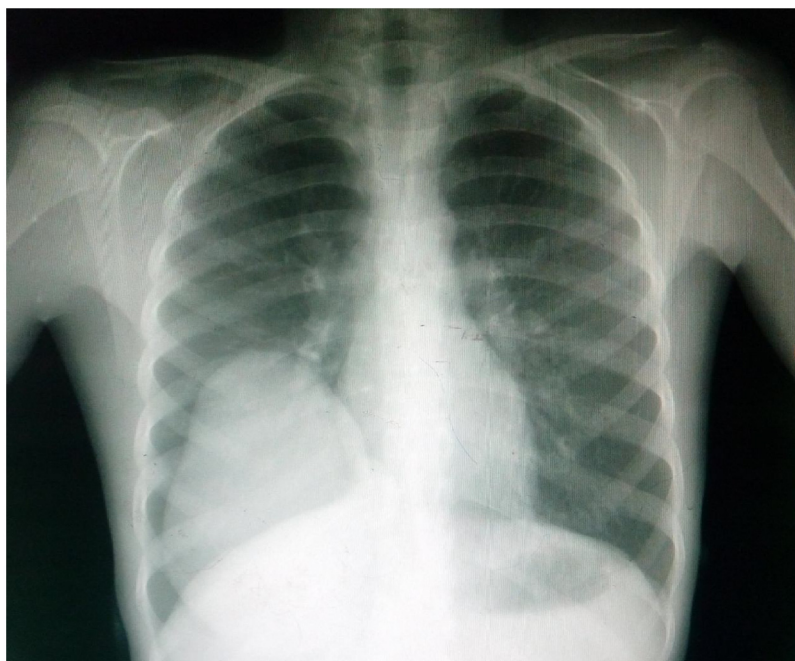


Fig. 1. On admission CXR:round regular density in the right lower lobe of the lung.

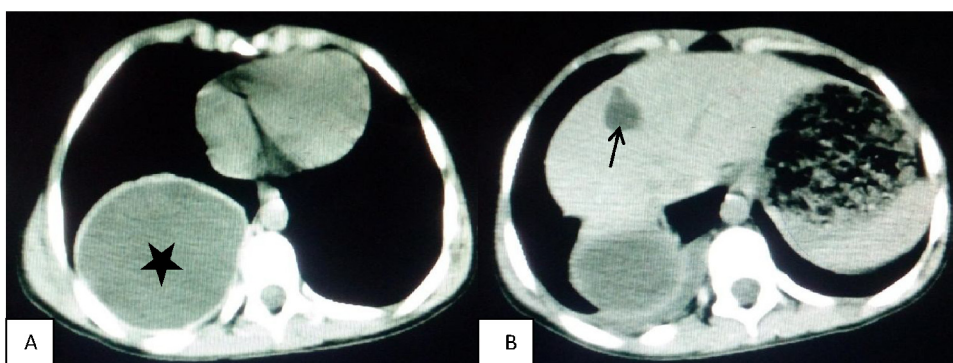


Fig. 2. Chest and abdominal CT scan with contrast: A: chest CT shows the 10*8 cm cyst in the right lower lobe of the lung(black star). B:abdominal CT reveals the small 1.5*2 cm cyst in the right lobe of the liver(black arrow).

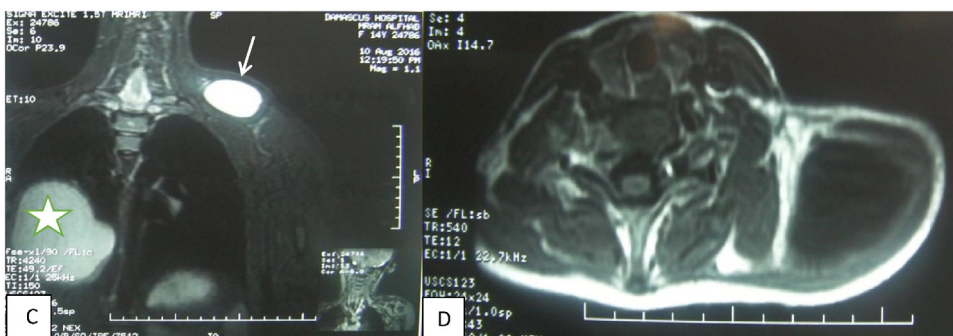


Fig. 3. Chest MRI: C: T2 weighted image(coronal section) shows two high sign intensity cystic lesions, one 5.5*4.5 cm within the thickness of left trapezius muscle(white arrow) and the other 10*8 cm in the right lower lobe of the lung(white star). D: T1 weighted image(axial section) reveals low sign intensity cystic lesion within left trapezius muscle.

tion, injection of 20% hypertonic saline and re-aspiration and then the germinal membrane then was enbloc excised. Bronchial fistulas and the remainder cavity were closed with 2.0 vicryl (absorbable) suture. A second incision was made over the left shoulder cyst. A 5 × 4 cystic mass was found within the thickness of the left trapez-

ius muscle, and was enucleated taking care not to induce any leak (Fig. 5) and the remainder cavity was closed with 2.0 vicryl suture. Complementary medical therapy with albendazole was continued for 3 months. The patient had been followed up for the next three



Fig. 4. The hydatid cyst in the right lower lobe of the lung during the operation.

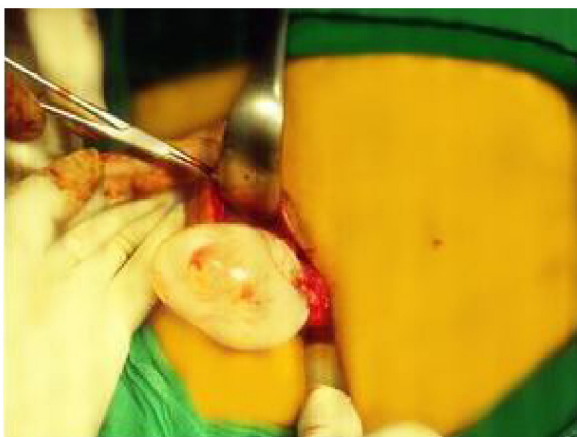


Fig. 5. The enucleation of left trapezius hydatid cyst.

months after surgery there were no signs of recurrence at the lung and trapezius cysts sites. The liver cyst reduced in size.

3. Discussion

Muscular hydatidosis usually occurs as isolated lesions without associated hepatic or pulmonary lesions [10]. The musculoskeletal involvement has been reported to occur in various anatomic regions including the thigh, quadriceps femoris, biceps femoris, psoas, diaphragm, triceps, pectoralis major, sartorius, gracilis, and supraspinatus muscles [11]. Muscular hydatidosis should be considered in the differential diagnosis of soft tissue lesions, especially in endemic areas, so detailed radiological studies should be performed to achieve a correct diagnosis. Generally most cases are asymptomatic and when apparent the symptoms are due to compression of adjacent structures or other complications. The Diagnosis of human echinococcosis remains highly dependent on imaging techniques to detect the cystic space occupying lesions [12]. Ultrasonography should be the first diagnostic tool used for detection of hydatid disease of soft tissue [3]. Computed tomography scan has an advantage over ultrasound for better documentation of site, size and structure of cyst. MRI may show an intense rim which has been proposed as a characteristic sign of hydatid disease [13]. Interventional diagnostic procedures should

be avoided to prevent anaphylaxis from spontaneous or iatrogenic rupture and to prevent recurrence.

The management of hydatid cysts can be done either surgically or medically. Surgery can be performed either by resection of the cyst preceded by aspiration and reinjection of scolicidal agents or by enucleation of the cyst. The surgical approach must be individualized depending on the cyst features. removal of the intact cyst is preferred, if feasible [14]. indications of surgical treatment include: complicated cysts, cysts with many daughter vesicles, diameter more than 10 cm, superficial cysts at risk of rupture due to trauma and extrahepatic disease [14], albendazole is generally administered one week prior to surgery and continued for at least four weeks postoperatively to prevent secondary echinococcosis in the event of cystic fluid spillage [14]. Prousalidis et al. concluded that conservative surgery can provide good results in multiple cystic echinococcosis. Radical surgery is ideal but only in properly selected cases [15]. Drug treatment alone is appropriate for management of small liver cysts with a single compartment and diameter less than 5 cm [16]. In our case the lung and the muscular lesions were managed surgically due to its size and site. The liver cyst was too small to be excised and it was difficult to be reached from the right thoractomy without complications.

4. Conclusion

Hydatidosis remains a serious health problem in endemic countries.

Any organ of human body could be affected by hydatid disease. The radiological procedures are critical in determining the site and relationships to the adjacent structures. Multiple hydatid disease is a complex surgical problem, and its treatment could be affected by size, anatomical sites, patient status, patient age and the experience of the centers. Multiple cysts could be managed surgically and The surgical treatment could be the best choice followed by chemotherapy especially with benzimidazoles. The optimal surgical approaches should be selected to reduce the recurrence rates and the other complications.

Conflicts of interest

None.

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None.

Ethical approval

None.

Consent

None.

Author contributions

Dr Hazem Aljaseem: study design, data analysis and writing the paper.

Dr Mohammad Almess: data collection and writing the paper.

Dr Monzer Bakgagi: data collection and writing the paper.

Guarantor

Hazem Aljaseem.

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