

Primary solitary hydatid cyst in paraspinal cervical muscles: a case report and review of the literature

Pojedyncza torbiel bąblowcowa w mięśniach przykręgosłupowych w odcinku szyjnym: opis przypadku i przegląd piśmiennictwa

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Abstract

Hydatid disease caused by *Echinococcus granulosus* and *Echinococcus multilocularis* commonly presents with pulmonary and hepatic cysts. Primary paraspinal muscle cysts are a rare presentation. We report a case of hydatid cyst within paraspinal muscles presenting with cervical mass and associated pain. The hydatid disease serological test was negative. Neither hepatic nor pulmonary cystic lesions were found. Radiographic findings were unspecific for hydatid cysts. Surgical resection was planned due to the provisional diagnosis of muscular cystic neoplasm. During surgery, a cyst containing a clear liquid was found. The cyst wall was excised and the surgical field was irrigated with hypertonic saline. The patient's symptoms resolved by discharge day. Postoperative pathological examinations revealed a muscular hydatid cyst.

Key words: paracervical muscles, hydatid cyst, hydatidosis.

Streszczenie

Bąblowica spowodowana przez *Echinococcus granulosus* lub *Echinococcus multilocularis* często przejawia się torbielami płuc lub wątroby. Pierwotne torbiele w mięśniach przykręgosłupowych są rzadką manifestacją choroby. W pracy opisano przypadek torbieli bąblowcowej w mięśniach przykręgosłupowych, która objawiała się guzem szyi i bólem. Badania serologiczne w kierunku bąblowicy dały wynik ujemny. Nie stwierdzono również zmian w wątrobie ani w płucach. Wyniki badań obrazowych nie były swoiste dla torbieli bąblowcowej. W związku z roboczym rozpoznaniem torbielowatego nowotworu mięśnia przeprowadzono operację, w trakcie której stwierdzono torbiel wypełnioną przezroczystym płynem. Torbiel wycięto wraz z jej ścianą, a pole operacyjne przepłukano hipertonicznym roztworem chlorku sodu. Dolegliwości zgłaszane przez chorego ustąpiły do dnia wypisu ze szpitala. W badaniu histopatologicznym stwierdzono torbiel bąblowcową w mięśniu.

Key words: mięśnie przykręgosłupowe, torbiel bąblowcowa, bąblowica.

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Introduction

Hydatid cystic disease is endemic in many parts of the world, specially in Mediterranean countries [1]. *Echinococcus granulosus* and less commonly *Echinococcus multilocularis* are primary species responsible for hydatid cysts in humans [2,3]. Its cycle consists of humans as an intermediate host and dogs as the final and definitive host [4]. The larval phase of *Echinococcus* penetrates the intestinal wall and is commonly transferred to the liver via the portal circulation [5]. As a result, hepatic hydatid cyst is the most common. Primary soft tissue hydatidosis without hepatic or pulmonary involvement is a rare entity in hydatid disease. Especially paraspinal muscular involvement occurs very rarely and it has been reported in a limited number of cases in the available literature [6].

Here, we report a case of cervical paraspinal muscular hydatid cyst without identifiable hepatic or pulmonary hydatid involvement.

Case report

A 33-year-old man presented with complaints of pain and a mass in the posterior part of his neck since two weeks earlier. His pain was aggravated with motion and changing position. In physical examination, a $4 \times 3 \times 3$ cm mass was revealed in the left posterior cervical region which was cystic in consistency without tenderness or erythema of overlying skin. All neurological examinations were normal. Chest X-ray and liver profile were normal. Serological test for hydatid disease was negative. Magnetic resonance imaging demonstrated a well-defined cystic lesion in the muscular compartment of the neck in its left posterior aspect. No relation to the thecal sac or posterior fossa was revealed (Fig. 1). With primary diagnosis of cervical neoplastic muscular cyst, the patient underwent a surgical intervention. During surgery, the cyst was entered; it contained clear liquid. The cyst wall was excised completely and sent to a pathological laboratory. Because of high prevalence of hydatid cysts in Iran, one of the rare differential diagnoses was paraspinal hydatid cyst. Therefore, the surgical field was irrigated with hypertonic solution to deal with spillage of cyst contents.

Microscopic pathological examination revealed laminated histology consistent with muscular hydatid cyst.

The patient was discharged in good general condition without complaining of previous symptoms.

Discussion

Primary hydatid cyst in soft tissue and muscles without hepatic or pulmonary cysts is extremely rare [7]. A proposed explanation for this observation is effective filtering effects of hepatic and pulmonary circulation which trap the echinococcal larvae [8]. In 10-15% of cases, however, larvae can escape from this filtering effect and form hydatid cysts in other organs [9].

Sener *et al.* [10] provided an alternative mechanism for bypassing the hepatic and pulmonary circulation in formation of primary widespread spinal and paraspinal hydatid cysts. Based on presence of porto-systemic anastomoses in various anatomical locations, they proposed that the larvae penetrate the intestinal muscle and may directly enter the inferior vena cava system through small venous connections at the intestinal walls instead of entering the portal circulation.

A palpable slow-growing mass is the most constant clinical finding in soft tissue hydatid disease. Symptoms related to their compressive effects on adjacent organs are another common finding [9]. Overall, paravertebral muscular hydatidosis presents with non-specific symptoms such as local oedema, pain, and tenderness [11-13]. Our patient's main complaint was consistent with findings described in the literature [14].

Rupture or intraoperative spillage of cyst contents may present as anaphylactic shock or cyst recurrence [8]. Muscular hydatid disease may mimic congenital cysts, pseudocysts, cystic tumours, abscess and haematomas. Therefore their preoperative diagnosis imposes a clinical challenge [7].

Radiographic tools are the mainstay of preoperative diagnosis of soft tissue cysts [15]. The MRI scan of the present case demonstrates a regular thin-walled cystic lesion without septation and enhancement resembling a simple muscle cyst. A multilocular lesion with several daughter cysts inside a mother cyst is considered characteristic although endovesicular daughter cysts are regarded as unusual in muscular hydatid cysts by some authors [15].

Some features are suggested to be helpful in diagnosis. The rim sign in the MRI appears as low signal intensity surrounding the cyst and can help to distinguish hydatid cysts from other pathologies [16]. A characteristic appearance resembling a bunch of grapes is described by Mirhoseini *et al.* In their case, the cyst wall was hypointense in both T1- and T2-weighted images, and they were thin-walled non-septated cysts without enhancement [17]. Detachment of the germinative lay-

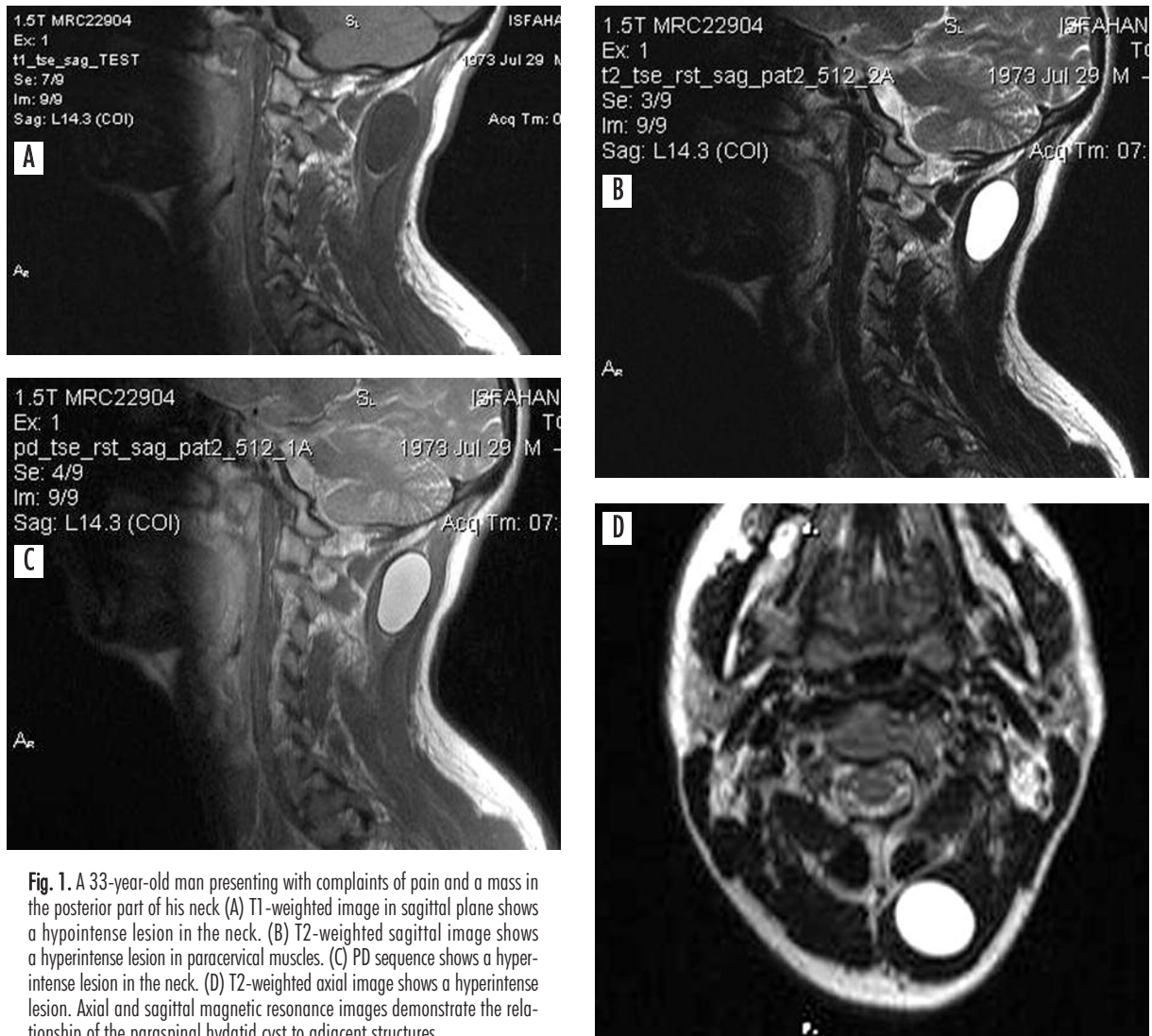


Fig. 1. A 33-year-old man presenting with complaints of pain and a mass in the posterior part of his neck (A) T1-weighted image in sagittal plane shows a hypointense lesion in the neck. (B) T2-weighted sagittal image shows a hyperintense lesion in paracervical muscles. (C) PD sequence shows a hyperintense lesion in the neck. (D) T2-weighted axial image shows a hyperintense lesion. Axial and sagittal magnetic resonance images demonstrate the relationship of the paraspinal hydatid cyst to adjacent structures

er from the pericyst (water lily sign) is considered to be pathognomonic but it is rare in musculoskeletal hydatid cysts [9].

In our case serological tests before surgery were negative. Serological tests such as haemagglutinin, complement fixation and ELISA may aid in diagnosis but are not positive in all cases of muscular hydatid cyst [7]. Although immunological tests are valuable in the diagnosis of hydatid cysts, only a positive test is helpful [18]. Therefore, complete reliance on serology for definitive diagnosis is not recommended [19]. The hydatid cyst capsule may play a role in false-negative results in serological tests because of isolation of the parasite from the host immune system by the cyst capsule [18]. Another proposed explanation is inadequate

Th-2 cell activation and cytokine production that is implicated in immunoglobulin expression in cystic echinococcosis [18].

Different treatment modalities have been proposed for treatment of hepatic and extrahepatic hydatid cysts. Therapy for extrahepatic echinococcal disease is dependent on size, location and clinical presentation of the cyst besides the general health status of the patient [6]. Small cysts can be managed with anti-helminthic drugs [6]. For muscular hydatid cysts, surgery with a broad safe margin is considered the best treatment [14]. García-Alvarez *et al.* reported that cysts that are not amenable to radical resection failed to heal and had to be operated again regardless of chemotherapy [16]. Adjuvant administration of benzimidazole derivatives pre- and

postoperatively is advocated by some authors [16]. Because of difficulties in definitive preoperative diagnosis of muscular paraspinous hydatid cyst and possible morbidities of radical en bloc resection of the cyst, percutaneous drainage techniques have been developed.

Conventional simple percutaneous drainage in hepatic cysts was not used routinely because of the dissemination risk. PAIR (puncture, aspiration, injection, re-aspiration) as a new version of percutaneous techniques with concurrent chemotherapy is used for hepatic cysts with promising results. It has been shown to be effective with low morbidity and recurrence [20,21].

Recently Biljic *et al.* reported the first case of paraspinous muscular hydatid cyst treated with the PAIR technique with subsequent albendazole. They performed percutaneous drainage, 95% ethanol injection and re-aspiration. Neither procedure-related complications nor recurrence were reported in 26 months of follow-up [19]. However, efficacy and potential complications of this technique are not confirmed in larger groups of patients and it seems that more experience is needed before advocating PAIR as an alternative therapy for surgery.

Although the excision of the intact cyst is considered ideal, it is not always possible. To deal with spillage of cyst contents and prevent formation of new cysts, methods of formalin or aqueous iodine, silver nitrate or hypertonic saline irrigation have been suggested in previous studies [17].

In the present case, based on radiographic and serological appearance, a simple muscle cyst was more likely. Therefore, the cyst was opened and the cyst wall was excised completely. Because hydatid cyst is known to be endemic in Iran [17], we considered the rare possibility of hydatid cyst. So the surgical field was irrigated with hypertonic solution in order to prevent dissemination of a possible hydatid cyst.

Based on the findings of the presented case, we emphasize considering hydatid disease in differential diagnosis of paraspinous muscular cysts, especially in endemic areas.

Disclosure

Authors report no conflict of interest.

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