Primary Echinococcosis of the Sternocleidomastoid Muscle

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Muscular echinococcosis accounts for 0.5% to 5.4% of all hydatid disease cases, with very little data on the incidence of muscular echinococcosis of the head and neck. We report a unique case of primary echinococcosis of the right sternocleidomastoid muscle in a 56-year-old man. Preoperative assessment by ultrasound and fine needle aspiration did not point to echinococcosis. We suspected the right diagnosis intraoperatively and confirmed it postoperatively by pathohistology and serologic tests. Echinococcosis of the liver and the lungs was also excluded postoperatively. Combination of operative treatment and postoperative albendazole therapy in two 28-day cycles one month apart resulted in complete regression of the disease. Echinococcosis should be considered as differential diagnosis of a multicystic mass in neck, particularly if it is of longstanding duration. Serologic tests for echinococcosis should be included in differential diagnostic procedures for each multicystic formation on the neck, especially in endemic areas.

Key words: albendazole; cysts; diagnosis, differential; echinococcosis; endemic disease; neck muscles; serologic tests

Echinococcosis (hydatid disease) is caused by Echinococcus granulosus, a small tapeworm parasite in dogs and wolves, and occasionally in cats (1). It occurs endemically in many areas worldwide, including the Mediterranean countries (1).

The main host and source of infection for humans is the dog, which acquires the infection through ingestion of infected cystic organs of another affected animal. The feces of infected animal contains a large number of eggs, which the dog transfers to its muzzle, tongue, and fur by licking its anal region. The humans get the infection from unwashed hands after having touched the dog that had swallowed the tapeworm eggs. Indirectly, the infection in humans may result from drinking water contaminated with dog feces or by consuming fresh fruit and vegetables washed or irrigated with contaminated water. The embryo within the swallowed egg is released from its envelope by the action of duodenal and intestinal juice, wherefore it penetrates intestinal mucosa, enters the circulation, and via the portal vein reaches the liver (1,2). Around 75% of the parasites are retained in the capillary circulation, thus making the liver the most common localization of the disease (1,3-5). If an embryo passes the obstacle, it migrates via the vena cava and right heart to the pulmonary capillaries. Therefore, the lungs are the second most common localization of echinococcal cysts (1,3,4). However, the embryo may pass this mechanical obstacle as well and reach via greater circulation any other organ in the body such as kidney, spleen, brain, heart, bone, orbit, and muscle and develop there (1,2,4,6-9).

Since the clinical picture is determined by the site, size, and number of cysts, it may vary greatly (1,2,6-9). The symptoms of compression are usually the initial clinical manifestation of the disease, preceded by a relatively long asymptomatic period (1-3,9). The patient’s general condition may exacerbate dramatically due to the complications of cyst rupture or infection (1,2,5,11).

Making the diagnosis of muscular echinococcosis is usually a time-consuming procedure (2-4,6,7,9,12). The diagnostic procedure includes obtaining history data, with special reference to the patient’s occupation and residence, clinical examination, ultrasonography, computed tomography, magnetic resonance imaging, fine needle aspiration, and serology (1-4,6-9,12).

In 1970, the incidence of echinococcosis was 0.24% in Dalmatia (southeastern region of Croatia) and almost two-fold in the wider area of Split, the capital of Dalmatia, and on some islands (1,2). Since 1950, the incidence of echinococcosis in the region has decreased by more than 70%, which is attributed to improved hygienic conditions and health education of the population (1,2).
According to different authors, muscular echinococcosis accounts for 0.5% to 5.4% of all hydatid disease cases (2,4,6,7,9,12). There are very little data on the incidence of hydatid disease of the head and neck (excluding the brain), and especially of muscular echinococcosis of the region. Cysts of the mandible, maxillary sinus, submandibular and parotid glands, mastoid process, infratemporal and palatine fossa, and parapharyngeal space have been described (2,3,8,9). We report a unique case of primary echinococcosis of the sternocleidomastoid muscle. To the best of our knowledge there has been no similar case reported in the literature.

Case Report

In May 1999, a 56-year-old man was admitted to the Department of Otorhinolaryngology for a tumorous formation on the right side of the neck. Seven years before, fine needle aspiration of the nodule had been performed, whereupon it started growing, especially during the past year. The patient reported no symptoms apart from the cosmetic one. The nodule was present on the right side of the neck from his early childhood, with the nodule growing to the size of an almond when he was 11 years old.

Inspection and palpation of the right side of the patient’s neck revealed a tumorous formation that seemed to consist of two parts, ie, medial part was a spherical formation of some 5 cm in diameter, and lateral part was ovoid formation of solid consistency, 7x4 cm in dimension. Both parts were sessile and mobile against the skin. Laboratory findings revealed erythrocyte sedimentation rate of 5 mm/h and white blood cells count 5.75x10⁹/L, with differential count of neutrophils 64%, lymphocytes 25%, basophils 1%, monocytes 8%, and eosinophils 2%. Neck ultrasonography revealed a multicystic formation in the right antero-lateral region of the neck (Fig. 1). Fine needle aspiration revealed some lymphocytes and histiocytes, one multinuclear giant cell, fine granular needle aspiration revealed some lymphocytes and metastatic infiltration (Fig. 2). The adjacent tissue consisted of connective tissue, mature adipose tissue, some striated musculature, and a single lymph nodule showing a picture of sinus histiocytosis. Based on the clinical picture and the findings obtained, additional examinations were required in order to reach definitive diagnosis. The indirect hemagglutination test for Echinococcus was positive (1:3200). Because the abdominal ultrasonography showed no focal lesions of the liver and lung X-ray was normal, the diagnosis of primary muscular neck echinococcosis was established.

The patient was prescribed albendazole (Dalben, Krka, Novo Mesto, Slovenia) 2x1 tablet for 28 days. Upon the completion of the first 28-day course of therapy, indirect hemagglutination test for Echinococcus was still positive (1:32), so the same therapy was repeated after one month. The following serologic tests were negative. Local findings on the patient’s neck showed no signs of recurrence.

Discussion

In our case, preoperative echosonography and fine needle aspiration failed to provide elements indicative for the diagnosis of hydatid disease. Only the postoperative examination demonstrated the presence of primary echinococcosis of the right sternocleidomastoid muscle.

It is recommended to include echinococcosis among differential diagnoses of the head and neck tumors, although it is quite rare (2,8,9). The patients with echinococcosis typically have a long disease history without specific symptomatology, which makes the diagnosis even more complex (1,2,9). In 1977, Petričević et al (2) studied patients with rare echinococcosis localizations, emphasizing that in such cases the disease pattern mimics the picture of a tumor, thus making the preoperative diagnosis extremely difficult. Although the disease occurs quite frequently in the Split endemic area (area at the Croatian Adriatic coast), many diagnoses are made intraoperatively or even by postoperative pathohistological examination of the extirpated tumor, as in the patient described (2,9).

Therapy with albendazole can be the therapy of choice in inoperable echinococcal cysts of the liver and in case of spinal cord echinococcosis, whereas in all other cases, albendazole therapy is combined with operative treatment or puncture-aspiration-injection-respiration therapy (1,2, 5-11,13). Albendazole in combination with operative therapy can be used pre- or postoperatively (1,2,10,13). When administrated preoperatively, it reduces the risk of secondary echinococcosis (1,2) and should be initiated at least 4 days before the operative procedure and continued for at least a month or preferably several months postoperatively. Recently, conti-
ous albendazole therapy is given even more preference to cyclic therapy (1,9-11). In our patient, a combination of operative treatment and postoperative albendazole therapy in two 28-day cycles one month apart resulted in complete regression of the disease.

Echinococcosis should be considered in the differential diagnosis of a multicystic mass in neck, particularly if it is of longstanding duration. Therefore, serologic tests for echinococcosis should be included in differential diagnostic procedures for each multicystic formation on the neck, especially in endemic areas, to make a timely and accurate diagnosis and provide the best possible and less expensive treatment (1,2).

References


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