**Answer to the Letter to the Editor of F.-A. Dauchy et al.
concerning manuscript “Intradural extramedullary primary
hydatid cyst of the spine: a case report and review of literature”.
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S. Sayyahmelli, A. Mahdkhah, M. Shoja M (2012)**

Recurrences in spinal echinococcosis

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Hydatid is a Greek word meaning “watery cyst”. Hydatid disease is caused by the parasitic tapeworm *Echinococcus* (*E. granulosus*, and less commonly *E. multilocularis*) [1]. This disease is present in all continents and it has been described in 100 countries [2]. Although echinococcosis is a worldwide problem; however, it is mostly reported from endemic areas [3]. Hydatid cysts are mostly localized in the liver. The second most common site for hydatid cyst is the lungs. The cysts have also been detected in the spleen, kidney, heart, bones, the central nervous system, and other organs, but with less frequency [4]. Bone involvement is very rare and spine is the most common site for the hydatid disease of the bone [1].

The life cycle of this worm involves two hosts: the definitive hosts including dogs, wolves, jackals, and foxes, whereas the intermediate hosts which are humans, cattle, sheep, goats, and horses. The adult tapeworm lives in the intestines of the definitive host and discharges eggs into the feces, which are ingested by the intermediate host. Oncospheres released by the eggs are swallowed by intermediate hosts, invade the duodenum, and enter the portal circulation. From the portal circulation, the oncospheres can lodge in the liver, lungs, and other organs and develop into hydatid cysts [5]. The embryos reproduce asexually and form multiloculated cysts. Within 3 weeks, the embryo develops into a larva, and a cyst is formed [6]. The hydatid cysts contain numerous scolices, each of which is capable of developing into an adult tapeworm [5] if ingested by definitive host. Humans contract the disease by

contamination through direct contact with the definitive host or its feces, or by ingesting food infected with ova [7]. The second reported method of human infestation is by inhaling dust carrying dried ova, which hatch in the lungs and form a cyst [8]. The cysts consist of an outer fibrous layer and a cestode-derived inner germinal membrane containing scolices [9]. The innermost germinal layer produces hydatid fluid and may include numerous embryonal scolices termed “hydatid sand” [10]. Scolices, the infectious embryonic tapeworms, develop from an out-pouching of the germinal layer known as the brood capsule. Freed scolices together with brood capsules form hydatid sand, which settles in the dependent part of the cyst [11]. For the treatment of spinal hydatid disease, surgery is currently considered the treatment of choice [1, 9] and surgical excision without intraoperative rupture results in cure [9]. In ruptured cysts, the rate of recurrence will be higher and irrigation with normal saline as well as medical therapy should be performed. Totally, the rate of recurrence is dramatically high, and in different reports the intraoperative seeding is commonly leading to recurrence rates up to 18–100 % [7, 10, 12–15]. Recurrences occur 2–28 months (mean 25.2 months) after the initial operation [16, 17].

Spinal hydatid cysts account for 1 % of all cases of hydatid disease [18]. Braithwaite and Lees have classified the spinal involvement by hydatid disease in five groups: (1) primary intramedullary hydatid cyst; (2) intradural extramedullary hydatid cyst; (3) extradural intraspinal hydatid cyst; (4) hydatid disease of the vertebrae; and (5) paravertebral hydatid disease. The first three types in this group are considered rare [19, 20]. Recurrence is very rare with the intradural extramedullary form of hydatid disease [9, 11].

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Recurrence can be seen in all of these types, but we should notify that there is a difference between intradural and other types of hydatid cysts. In contrast to extraosseous involvement, larval penetration of the cancellous bone in the vertebrae results in a multivesicular and diffuse infiltration [8, 17]. The main cause may be related to defensive reaction from the bone results in a microvesicular infiltrative growth of the hydatid lesions, making it total surgical removal almost impossible in cases of vertebral involvement [21]. Rupture occurs most frequently in spinal hydatidosis with bone involvement, because the cysts are contained in narrow spaces within bone boundaries, making removal particularly difficult [7]. In these cases, the possibility of retain and rupture is high, but the cysts of soft tissues and extra osseous lesions are usually unilocular with thicker cystic walls and spherical shape due to the lack of resistance from the hard bone tissue during their growth [1] such as our case [22]. As a result, it is easier to remove in intact forms and the rate of recurrence will be lower. Recurrence was reported in 28 (17.8 %) cases from 45 intradural extramedullary hydatid cysts [22]. Because of short time of study and unmentioned recurrence in 17 cases, the real rate may be even more. In 56 % of reported intradural extramedullary hydatid cysts cases, the cysts were univesicular [22]. Another important point is that the cysts containing hydatid sand are fertile cysts. If no sand is present, the cyst is sterile and does not form secondary cysts in case of spillage. It takes 6–9 months for a cyst to become fertile [8]. The surgical intervention is usually performed in the primarily infertile stages of intradural lesions because of early neurological manifestations due to low accessible spaces. Surgery during sterile phases will be resulted in reduced recurrence rate even in ruptured cases. Patients who experience an intraoperative cyst rupture should be operated on again within that period to avoid the possibility of recurrence [8].

Despite all of these, “recurrence” is a common and significant dilemma and for this reason, spinal hydatidosis has a poor prognosis and has been compared with spinal malignancies [13]. Recurrence is actually never seen following complete resection of an intact cyst with radical surgical interventions when feasible [23]. The best treatment remains active nationwide prevention of the disease [24] and postoperative follow-up period should be at least 3 years and continued as long as possible [23].

Conflict of interest None.

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