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REVIEW PAPER

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Hydatidosis of central nervous system and its coverings in the pediatric and adolescent age groups in Turkey during the last century: a critical review of 137 cases

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Abstract *Introduction:* Hydatid disease is still a major problem in infested areas of the world, especially in the rural areas, including Turkey. *Objective:* The objective of this review was to analyze the literature on the management of central nervous system (CNS) hydatidosis with an emphasis on their specificities in childhood and adolescence, with the aim of determining the clinical and neuroradiological findings and treatment modalities, medical or surgical, in these age groups. *Materials and methods:* To establish some guidelines for the diagnosis and treatment of this controversial condition, publications reported from Turkey in national (n=33) and international (n=55) journals during the last century and databases containing medical literature were used. Strikingly, the numbers of articles produced by Turkish authors on CNS hydatidosis have risen tremendously during the study period. Although a total of 272 cases of intracranial and intraspinal hydatid cysts were reported from Turkey, only 137 cases for which detailed information was available were selected for further analysis, in keeping with our inclusion and exclusion criteria. *Results and discussion:* Despite the inherent limitations, this type of study indicates that the incidence of hydatidosis has not decreased in Turkey in recent years. The clinical findings were mostly atypical, and it was interesting that 4 patients were described as having cerebrovascular occlusive disease and 3 as having symptoms of movement disorders. Computed tomography and/or MRI techniques were extremely useful, both in reaching the correct diagnosis and for proper surgical management of hydatid disease, because of the absence of a pathognomonic clinical picture of this disease. The treatment of choice for hydatid disease of the CNS and its coverings was complete intact removal of the cyst. In contrast to that in intracranial hydatid cysts, however, surgical intervention was palliative, not curative, in almost all cases of intraspinal hydatidosis. According to this critical review of the literature, CNS hydatidosis is therefore still a life-threatening condition, in spite of all the advances in imaging techniques and therapeutic methods. The most important factors in prognosis are the localization of the focus of infection, rupture and of the cyst and dissemination of its content, and treatment modality. At present, surgical intervention preceded by careful neuroradiological evaluation remains the best surgical therapy, and this plus adjuvant chemotherapy is advocated in some cases as the gold standard for therapy.

Keywords Adolescent · Central nervous system · Chemotherapy · Child/children · Hydatid disease · Surgery

Introduction

Although hydatidosis in the central nervous system (CNS) and its coverings is very rare, it is still a serious problem in the infested countries. Echinococcosis is caused by the larval stage of the tapeworm *Echinococcus granulosus* (cystic hydatid disease), *Echinococcus multilocularis* (alveolar hydatid disease), *Echinococcus vogeli* (polycystic type), or *Echinococcus oligarthus* [64, 89]. However, the first two types are of clinical importance in humans and can occur in the viscera, particularly liver (60–75%) and lung (15–30%) [99]. In 50–75% of cases, CNS hydatidosis is found in the pediatric and adolescent age groups [30]. In these age groups, CNS hydatidosis can be associated with involvement of other organs or may be an isolated infestation of the CNS and its coverings. Only embryos that succeed in passing through the filtering barrier systems in the liver and lung reach the brain or spinal cord by way of the systemic circulation [89]. Once the hexacanth embryo has arrived in the target organ, it will form a hydatid cyst because of the ideal conditions for growth of such a cyst.

It has been mentioned that hydatid disease is located in the intracranial cavity in only 0.5–3% of all cases [11, 24, 25, 28, 30, 32, 40, 54, 61, 74]. In a similar fashion, intraspinal involvement is also uncommon, being found in 0.5–2% of all cases of hydatidosis [4, 58, 77, 89]. In clinical practice, children with hydatid cysts of the CNS and its coverings are frequently seen by neurosurgeons, pediatric neurologists and spine surgeons, especially in agricultural parts of the world, and in these especially where sheep rearing is widespread. At present, it is one of the most important helminthic diseases in Turkey, as it is a serious cause of public health and economic problems [11, 15, 94, 98, 99, 100, 102, 103].

In a recent comprehensive review of the reports on CNS hydatid disease from Turkey, Altınörs et al. [11] discovered 458 cases, including 219 cases that were the subjects of cooperative study. However, adult and pediatric patients were dealt with together. There is a need for a critical evaluation of the disease in the pediatric and adolescent age groups to provide information that will be helpful in diagnostic and therapeutic strategies. This paper focuses on the clinical, imaging, and therapeutic features of CNS hydatidosis, with an emphasis on their specificities in these age groups. This study is based on a critical review of Turkish cases (119 with intracranial hydatid cysts and 18 with intraspinal hydatid cysts) published during the last century.

Clinical materials and methods

For this study all reports so far published by Turkish authors based at different centers in Turkey were reviewed. I conducted a search in Index Medicus and its recognized database Medline, which indexes papers from peer-reviewed scientific publications, national and international library periodicals, and congress proceedings, as the basis of this review. To be included in this literature survey analysis, patients had to have been younger than 21 years when they were diagnosed with hydatid disease of the CNS and/or its coverings. Papers involving hydatid cysts of other organs, including the infratemporal fossa and the orbita, were excluded.

Regarding the type of paper, articles included in "Viewbox" and "Short report" sections were classed as "Case reports." On the other hand, articles describing imaging features of CNS hydatidosis and appearing under the rubric "Pictorial Essay" were classed as "Review" articles. As a general rule, the earlier reports were omitted if the data in them were also included in later publications. Duplicative publications and those giving no detailed data on the patients were also excluded.

Results

The information on this type of cyst was derived from 88 articles, 33 of which were published in national Turkish journals and 55 in international journals (Tables 1, 2) [1, 2, 3, 4, 5, 6, 7, 10, 11, 13, 14, 16, 17, 18, 19, 21, 22, 23, 25, 26, 27, 29, 31, 32, 33, 34, 35, 36, 37, 38, 39, 40, 41, 42, 43, 44, 45, 46, 47, 48, 49, 50, 52, 53, 54, 55, 56, 57, 58, 59, 60, 62, 63, 66, 67, 68, 69, 70, 71, 72, 73, 74, 75, 76, 77, 78, 79, 80, 81, 82, 83, 85, 86, 88, 90, 91, 93, 94, 99, 100, 103, 104, 105, 106, 107, 108, 109, 110, 111]. In most of these papers, the hospitals concerned are the major regional referral centers for hydatid disease in Turkey. In this retrospective study, the numbers of articles produced by Turkish authors on CNS hydatidosis were found to have risen tremendously during the study period. Of all publications on CNS hydatidosis, 60 (68%) were published between 1991 and 2002, and 63% of the papers were case reports, as expected. When classified by localization of the hydatid disease, these articles fell into three main groups: about intracranial disease (n=62), about intraspinal disease (n=19), and about both (n=7). They are shown according to year of publication, number of authors, institutions, type of cystic lesion and type of study (letter, abstract, case report, clinical series, review) in Table 3. The rate of articles per author has increased from 3.4% to 4.1% since 1991. Despite the inherent limitations, this type of study indicates that the incidence of hydatid disease has not decreased in Turkey over recent years. Even though it is an intuitive conclusion, in addition to which there are reports of cases with insufficient information in the literature, I believe that many patients have been treated for CNS hydatidosis without their cases being reported in any scientific publication or at any scientific meeting in Turkey. At the present time, most neurosurgeons and neuroradiologists in Turkey have clinical experience in the diagnostic and therapeutic aspects of hydatid disease. In the cases reviewed in this study, hydatid disease was caused by infestation by larvae of the tapeworm *Echinococcus granulosus* in all cases with intracranial or intraspinal involvement. We present the results of our literature re-

Table 1 Two hundred fifty-two pediatric and adolescent patients with intracranial hydatid cyst reported by Turkish authors in national and international journals during the last century. *NS* not stated

Reference	No. of authors	Institutions/city	Year of publication	Type of cyst		Total
				Solitary cyst	Multiple cyst	
[18]	1	Ankara Numune State, Ankara	1944	1	–	1
[90]	1	İstanbul University, Çapa, İstanbul	1961	4	–	4
[42]	2	Bakırköy State, İstanbul	1962	1	–	1
[81]	3	Haydarpaşa Numune State, İstanbul	1968	1	–	1
[91]	2	Hacettepe University, Ankara	1968	3	–	3
[54]	4	İstanbul University, Çapa, İstanbul	1975	NS	NS	NS
[38]	3	Ankara University, Ankara	1979	1	–	1
[74]	6	Hacettepe University, Ankara	1979	NS	NS	NS
[93]	5	Atatürk University, Erzurum	1983	1	3	4
[75]	4	Hacettepe University, Ankara	1984	1	–	1
[43]	4	Ankara Numune State, Ankara	1986	1	–	1
[6]	5	Sami Ulus State, Ankara	1986	1	–	
[39]	4	Ankara University, Ankara	1986	1	–	1
[21]	5	İstanbul University, Çapa, İstanbul	1987	1	–	1
[37]	2	Ankara University, Ankara	1988	1	–	1
[46]	5	Hacettepe University, Ankara	1989	–	2	2
[78]	3	Erciyes University, Kayseri	1989	–	1	1
[36]	3	Hacettepe University, Ankara	1989	1	–	1
[40][a]	4	Ankara University, Ankara	1989	NS	NS	16 (+4)
[26]	4	Hacettepe University, Ankara	1991	–	1	1
[29]	5	Haydarpaşa Military, İstanbul	1991	NS	NS	NS
[79]	4	Hacettepe University, Ankara	1991	–	1	1
[27]	5	Çukurova University, Adana	1991	NS	NS	17
[25][b]	5	Hacettepe University, Ankara	1992	NS	NS	89 (+9)
[47][c]	4	Hacettepe University, Ankara	1992	(1)	–	(1)
[55][d]	6	Çukurova University, Adana	1992	1 (+3)	–	1 (+3)
[14]	3	Taksim State, İstanbul	1992	–	1	1
[67]	5	İstanbul University, Çapa, İstanbul	1992	1	–	1
[109]	3	Erzurum Numune State, Erzurum	1993	1	–	1
[31]	4	Haydarpaşa Military, İstanbul	1993	–	1	1
[34]	3	Ege University, İzmir	1993	17	2	19
[5]	4	Çukurova University, Adana	1994	–	1	1
[33]	4	Selçuk University, Konya	1994	–	1	1
[22][e]	4	İstanbul University, Çapa, İstanbul	1994	(2)	–	(2)
[73]	1	Marmara University, İstanbul	1994	3	1	4
[88]	5	Erciyes University, Kayseri	1994	1	–	1
[23]	2	Dokuz Eylül University, İzmir	1995	1	–	1
[10]	6	Ankara Social Security, Ankara	1995	1	2	3
[52][f]	5	Atatürk University, Erzurum	1995	9 (+1)	1 (+3)	10 (+4)
[35]	4	Ege University, İzmir	1995	1	–	1
[106]	4	İnönü University, Malatya	1996	1	–	1
[19]	3	Ankara Numune State, Ankara	1996	1	–	1
[62]	3	Bakırköy State, İstanbul	1996	1	–	1
[85][g]	1	Ege University, İzmir	1996	(1)	–	(1)
[103]	3	Hacettepe University, Ankara	1997	–	1	1
[32]	4	İstanbul University, Cerrahpaşa, İstanbul	1997	3	–	3
[44]	5	Hacettepe University, Ankara	1997	1	1	2
[69]	6	İstanbul University, Çapa, İstanbul	1997	2	1	3
[105]	5	Ankara University, Ankara	1998	–	1	1
[104][h]	2	Ankara Social Security, Ankara	1998	2	(1)	2 (+1)
[110]	3	Yüzüncü Yıl University, Van	1998	–	1	1
[86][i]	4	Ege University, İzmir	1998	NS	NS	5(+1)
[100][j]	2	Adnan Menderes University, Aydın	1998	(1)	(1)	(2)
[111]	2	Ankara University, Ankara	1998	1	–	1
[60][k]	3	Ege University, İzmir	1999	2 (+18)	1 (+2)	3 (+20)
[66]	4	Dicle University, Diyarbakır	1999	–	1	1
[63]	6	Erciyes University, Kayseri	1999	1	–	1
[108]	2	Uludağ University, Bursa	1999	1	–	1
[11]	4	Başkent University, Ankara	2000	NS	NS	NS
[2]	5	Numune State, Ankara	2000	–	1	1
[99]	1	Adnan Menderes University, Aydın	2001	NS	NS	NS

Table 1 (continued)

Reference	No. of authors	Institutions/city	Year of publication	Type of cyst		Total
				Solitary cyst	Multiple cyst	
[50]	5	Ondokuz Mayıs University, Samsun	2001	1	–	1
[56]	6	Dicle University, Diyarbakır	2001	1	–	1
[13]	1	Erzurum Social Security, Erzurum	2001	–	1	1
[76]	3	Dicle University, Diyarbakır	2001	–	1	1
[71]	7	İnönü University, Malatya	2001	1	–	1
[80]	4	Bakırköy State, İstanbul	2001	1	–	1
[70][l]	11	İstanbul University, Çapa, İstanbul	2001	21 (+7)	1 (+1)	22 (+8)
[57][m]	6	Dicle University, Diyarbakır	2001	(1)	–	(1)
Total				96	29	252

[a] Because 4 of 20 cases reported by the authors had already been described by Gökalp et al. [38], Aksüyek et al. [6], Gökalp et al. [39], and Gökalp and Erdoğan [37] earlier, only 16 cases were considered in order to avoid an overlap
[b] Because 9 of 98 cases reported by the authors had been described by Tevrüz and Bertan [91], Özgen et al. [74], İplikçioğlu et al. [46], Gedikoğlu et al. [36], Çataltepe et al. [26], and Peker et al. [79] earlier, only the remaining 89 cases were considered
[c] The same patient as reported by the authors had been published by Gürçay et al. [43] previously
[d] Of the cases reported by the authors, 3 had already been described by Çetinalp et al. [27]
[e] Of the cases reported by the authors, 1 had been described by Tarcan [90] and the other 1 had been described by Canbolat et al. [21] previously
[f] Out of cases reported by the authors, 4 had already been described by Tümer et al. [93]
[g] Same case reported by the authors had been published by Erşahin et al. [35] previously
[h] Because 1 of 3 cases reported by the authors had been described by Altınörs et al. [10] earlier, 2 cases were considered
[i] Out of cases reported by the authors, 1 had already been described by Erşahin et al. [35] and Şener [85]
[j] Out of cases reported by the authors, 1 had been described by Talaslıoğlu et al. [88] and the other 1 had been described by Turgut et al. [103] previously
[k] Because 20 of 23 cases reported by the authors had already been described by Erşahin et al. [34] earlier, 3 cases were considered
[l] Because 8 of 30 cases reported by the authors had been described by Tarcan [90], Canbolat et al. [22], and Önal et al. [69] earlier only the remaining 22 cases were considered
[m] Same patient reported by the authors had been published by same authors [56] previously

Table 2 Twenty pediatric and adolescent patients with intraspinal hydatid cyst reported by Turkish authors in national and international journals during the last century

Reference	No. of authors	Institutions/city	Year of publication	Type of cyst		Total
				Solitary cyst	Multiple cyst	
[82]	1	Ankara University, Ankara	1947	–	1	1
[83][a]	1	Ankara University, Ankara	1948	–	(1)	(1)
[59]	1	NS, İstanbul	1958	–	1	1
[41]	1	Bakırköy State, İstanbul	1968	–	1	1
[77]	4	Hacettepe University, Ankara	1984	–	3	3
[7]	5	İzmir State, İzmir	1989	NS	NS	NS
[58]	5	Ankara Numune State, Ankara	1989	NS	NS	NS
[107]	6	Bezm-i Alem Valide Sultan, İstanbul	1989	1	–	1
[72]	4	Atatürk University, Erzurum	1990	–	1	1
[4]	5	Hacettepe University, Ankara	1991	1	–	1
[68]	9	İstanbul University, Çapa, İstanbul	1992	NS	NS	NS
[73]	1	Marmara University, İstanbul	1994	–	1	1
[1]	5	Bayındır Medical Center, Ankara	1996	–	1	1
[32]	4	İstanbul University, Cerrahpaşa, İstanbul	1997	1	–	1
[94]	1	Adnan Menderes University, Aydın	1997	NS	NS	NS
[49]	3	Ege University, İzmir	1998	–	1	1
[17]	3	Ankara University, Ankara	1998	–	1	1
[48][b]	8	Ege University, İzmir	1998	2(+1)	–	2(+1)
[16]	5	Gazi University, Ankara	2000	–	1	1
[3]	4	Taksim State, İstanbul	2001	–	1	1
[45]	4	İstanbul University, Cerrahpaşa, İstanbul	2002	1	–	1
[53]	4	Haydarpaşa Numune State, İstanbul	2002	1	–	1
Total				7	13	20

[a] Because the case described by the authors had already been reported by the same author [82] in another journal, this case was excluded
[b] Of the cases reported by the authors, one had already been described by İşlekel et al. [49]

Table 3 Number of journals and types of papers reporting on CNS hydatid cysts in the pediatric and adolescent age groups in Turkey

	Type of journal		Type of paper				
	National	International	Letter	Abstract	Case report	Clinical series	Review
Intracranial hydatid cyst	19	43	1	4	43	12	2
Intraspinal hydatid cyst	11	8	–	5	11	2	1
Both	3	4	–	–	1	3	3
Total	33 (37.5%)	55 (62.5%)	1 (1%)	9 (10%)	55 (63%)	17 (19%)	6 (7%)

view on CNS hydatidosis in Turkey; we differentiate between intracranial hydatid cysts and intraspinal hydatid cysts.

Intracranial hydatid cysts

So far, there have been 252 reported pediatric and adolescent cases of intracranial hydatid cyst in total. However, in keeping with our inclusion/exclusion criteria, only 119 of these patients were considered for further analysis, and the remaining 133 cases, for which the data were incomplete, were excluded [11, 23, 25, 27, 29, 40, 44, 54, 73, 74, 86, 99, 104]. Thus, there were 67 male (56%) and 52 female (44%) patients. Solitary cysts were diagnosed in 92 patients (77%) and multiple cysts in the remaining 27 (23%). Of those with multiple involvement, 15 patients were reported to have numerous cystic lesions [14, 33, 35, 44, 46, 60, 66, 70, 73, 76, 79, 91, 111]. The youngest patient was a 2-year-old boy, and the mean age at the time of diagnosis was 10.8 years (Table 4). The locations of the intracranial cysts are noted in Table 5. Of the total of 259 intracranial cysts in pediatric and adolescent age groups, only 13 (5%) were localized in the extradural space. Characteristically, there were no specific signs and the symptoms varied widely from patient to patient, depending on the location and type of the cystic lesion. Interestingly, 4 patients had been reported as "stroke victims" owing to embolization of echinococcal material [56, 57, 88, 100, 101, 105] and 3 with the symptoms affecting the extrapyramidal system, as having a "movement disorder" because of the anatomical localizations of the cysts [35, 43, 47, 81, 85]. Other diagnostic studies, such as X-ray films, pneumoencephalography, ventriculography, angiography, scintigraphy, and electroencephalography, were used in patients with intracranial hydatid cysts in earlier years. In the last two decades, however, CT scanning and MRI have proved to be the best imaging methods for diagnosis. A search for the presence of hydatid cysts in other body regions disclosed extraneural involvement in 30 patients (25%), and the sites of extraneural involvement reported are given in Table 6 [2, 5, 10, 18, 32, 34, 50, 57, 60, 62, 63, 66, 69, 70, 88, 93, 103, 105, 106, 108]. One patient with systemic organ involvement died before surgical intervention

Table 4 Distribution of 137 cases of (CNS) hydatidosis in Turkey by patient age

Group	Childhood (0–16 years)	Adolescent (17–20 years)	Total
Intracranial hydatid cyst	105	14	119
Intraspinal hydatid cyst	12	6	18
Total	117 (85%)	20 (15%)	137

Table 5 Anatomical locations of intracranial hydatid cysts in 119 pediatric and adolescent cases found in the literature search[a]

Location of the cyst	No. of all cysts	No. of extradural cysts	Percentage of total
Cerebral hemispheres (*n*=208)			80.0
Frontal	32	–	
Parietal	38	–	
Temporal	6	–	
Occipital	25	–	
Frontoparietal	18	1	
Frontotemporal	14	–	
Parietotemporal	10	1	
Parieto-occipital	31	–	
Frontotemporoparietal	6	–	
Parietotemporo-occipital	26	–	
Thalamic	2	–	
Ventricular system (*n*=33)			13.0
Lateral ventricle	21	–	
Fourth ventricle	1	–	
Aqueduct of Sylvius	8	–	
Periventricular area	2	–	
Not specified	1	–	
Posterior cranial fossa (*n*=9)			3.5
Cerebellum	5	1	
Pons	2	–	
Cerebellopontine angle	2	2	
Tentorium cerebelli (*n*=1)	1	–	0.5
Cranial base (*n*=4)			1.5
Sella turcica	1	1	
Parasellar area	1	1	
Retroclival	1	1	
Basal cisterns	1	–	
Skull (*n*=4)	4	4	1.5
Total	259	13 (5%)	100.0

[a] Some patients had two or more than two hydatid cysts in the intracranial cavity

Table 6 Sites of extraneural involvement in pediatric and adolescent cases of CNS hydatidosis with systemic involvement (n=30)[a, b]

Affected organ or system	No. of cases with		Total no. of cases
	Intracranial hydatid cyst	Intraspinal hydatid cyst	
Liver	24 (2)	2	26 (2)
Lung	14 (2)	2	16 (2)
Heart[c]	5	–	5
Vessel	3	–	3
Kidney[d]	7 (1)	1	8 (1)
Pancreas	1	–	1
Spleen	–	1	1
Not specified	–	1 (1)	1 (1)
Total	54 (5)	7 (1)	61 (6)

[a] Some patients had two or more than two hydatid cysts in the intracranial cavity
[b] The values within the parentheses indicate the number of cases with multiple hydatid cysts
[c] One out of five cysts was localized in the paracardiac region
[d] One out of seven cysts was localized in the suprarenal region

Table 7 Recurrence and mortality rates after different forms of treatment in pediatric and adolescent patients with intracranial hydatid cyst (n=119)[a]

Treatment	No. of cysts	Recurrence	Mortality
Surgical intervention alone (n=80)			
Hydatid birth/total intact removal	119	4	4
Removal with rupture	31	7	3
Puncture with/without resection	12	4	3
Not specified	11	–	–
Surgery plus chemotherapy (n=84)			
Hydatid birth/total intact removal + mebendazole	17	–	–
Hydatid birth/total intact removal + albendazole	37	–	–
Removal with rupture + mebendazole	10	2	–
Removal with rupture + albendazole	7	–	–
Puncture with/without resection + mebendazole	4	–	–
Puncture with/without resection + albendazole	1	–	–
Not specified	8	–	–
Surgery plus radiotherapy (n=1)			
Puncture with/without resection + radiotherapy	1	–	–
No treatment (n=1)	1	–	1
Total	259	17 (14%)	12 (10%)

[a] Some patients had two or more than two hydatid cysts in the intracranial cavity

was undertaken, and the diagnosis was made at autopsy. A total of 259 cystic lesions in the intracranial cavity were treated with some kind of surgical technique – hydatid birth or total intact removal, removal with rupture, and puncture with or without resection. "Hydatid birth," also called Dowling's technique, means removal of the cyst by forcing saline solution around it (hydrostatic expulsion) [99]. This surgical technique was used for 119 (46%) of the reported cystic lesions (Table 7). In 31 patients with cystic lesions (12%), the cyst was ruptured inadvertently during surgery. In the remaining 12 (5%) cases the cyst was punctured, its content was aspirated, the cystic cavity was irrigated with different topical disinfectants, and the cyst wall was excised (PAIR technique). Postoperatively, a shunt device was inserted for treatment of hydrocephalus in 4 cases, of subdural effusions in 1, and of porencephalic cyst in 1 [32, 34, 60, 70, 91]. Adjunctive drug therapy (mebendazole or albendazole) was used in 84 cystic lesions for a variety of indications: rupture or puncture of the cyst during surgery, systemic hydatid disease, inoperable cases with multiple cysts. Some patients treated with the hydatid birth technique for expulsion of intact cysts had received antihelminthic agents for these indications. However, in the case of 12 cysts in 4 of the patients, it is difficult to understand why chemotherapy was administered. In this series, secondary infection of hydatid cysts, i.e. "infected" hydatid cysts, was observed in 3 cases, making it an unusual finding [33, 73, 103]. Recurrence was reported for 17 cystic lesions (7%) at different times after surgery. In only 1 patient was a combination of surgery and radiotherapy used. Overall, 14% of the patients suffered recurrences of the infection. There were 12 deaths in patients with intracranial hydatid cysts, giving an overall

Table 8 Anatomical location of intraspinal hydatid cysts in 18 pediatric and adolescent cases found from the literature search[a, b]

Location of the cyst	No. of extradural cysts	No. of subdural cysts	Total of cysts	Percentage of total
Cervical	1	–	1	3.5
Thoracic	14 (4)	1	15 (4)	50.0
Thoracolumbar	1	–	1	3.5
Lumbar	9 (2)	1 (1)	10 (3)	33.0
Sacral	2 (1)	1	3 (1)	10.0
Total	27 (7)	3 (1)	30 (8)	100.0

[a] Some patients had two or more than two hydatid cysts in the intracranial cavity
[b] Values in parentheses indicate the number of cases with multiple hydatid cysts

Table 9 Recurrence and mortality rates after different forms of treatment in pediatric and adolescent patients with intraspinal hydatid cyst (n=18)[a]

Treatment	No. of cysts	Recurrence	Mortality
Surgical intervention alone (n=6)			
Hydatid birth/total intact removal	3	–	–
Removal with rupture	3	1	–
Not specified	3	–	–
Chemotherapy alone (n=1)			
Albendazole	1	–	–
Surgery plus chemotherapy (n=10)			
Hydatid birth/total intact removal + mebendazole	5	–	–
Hydatid birth/total intact removal + albendazole	3	–	–
Removal with rupture + mebendazole	1	–	–
Removal with rupture + albendazole	5	2	–
Not specified	5	1	–
No treatment (n=1)	1	–	1
Total	30	4 (22%)	1 (6%)

[a] Some patients had two or more than two intraspinal hydatid cysts

mortality rate of 10% (12 of 119). The average length of follow-up was 82.6 months.

Intraspinal hydatid cysts

To date, the total number of reported cases of intraspinal hydatid cyst is 20, but only 18 of the papers were found to be eligible for the study when the inclusion and exclusion criteria were applied. The data in the remaining papers were incomplete and the cases in these were excluded [7, 45, 58, 68, 83, 94]. There were 11 male (61%) and 7 female (39%) patients. Table 4 lists patients with this disease by age group. In the present study, the youngest case was a 6-year-old boy and the mean age at onset was 13.9 years. For diagnostic purposes, X-ray films were taken in all patients reported, CT scanning with/without myelography in 5 patients, and MRI in 7 patients. Extraneural involvement was present in only 3 patients, and a total of seven organs were affected (Table 6) [1, 4, 48]. Detailed information on lesion location was available for 18 of the patients reported (Table 8). At presentation, almost all extradural cysts were associated with vertebral and paraspinal involvement. Solitary cysts were diagnosed in 6 patients (33%), and multiple cysts in the remaining 12 (67%). Out of 12 cases with multiple involvement, 8 were reported to have numerous cystic lesions [1, 16, 41, 49, 72, 73, 77]. Chemotherapy was administered for 63% of the cystic lesions. As seen in Table 9, intraspinal cysts recurred in 22% of patients and the overall mortality rate was 5.5%. In the current study, one of the things that I found distressing was that most of the reported patients with intraspinal cysts had had previous operations for hydatid disease. The average length of follow-up was 17.1 months.

Discussion

Hydatidosis is most widespread in the Middle East (Lebanon, Syria, Palestine, Kuwait, Egypt, and Libya), Australia, New Zealand, Latin America (Argentina, Uruguay, and Paraguay), Central Europe, South Africa, and the Mediterranean countries (Spain, France, Italy, Greece, and Turkey) [99]. In Turkey, there are several factors known to exacerbate the problem of hydatidosis: the presence of numerous stray dogs, the high proportion of the population that is involved in farming and breed-

ing livestock, the infringements of sanitary regulations during the Muslim Festival of Sacrifice, the fact that livestock is slaughtered without veterinary supervision, and the absence of detailed prevalence studies in spite of the obligation to report new cases to the Turkish Ministry of Health and Social Assistance [12]. As a result, this condition causes severe economic losses through:

1. The high financial costs of medical and surgical treatment for the infected persons
2. The total costs of the human disability caused by the disease
3. Occupational losses during infected persons' hospitalization and recovery
4. Loss of the infected organs of livestock [84].

More recently, Altınörs et al. [11] mentioned that: "The clinical behavior of the disease is aggressive and is regarded as a potential malignancy." Actually, CNS hydatid cysts are still diagnostically and therapeutically challenging lesions despite our encouraging experience in most cases. According to many earlier reports, it is more common in children and adolescents than in adults, and the cyst takes several years to grow large enough to start compressing the surrounding structures [11, 30, 89, 99]. Characteristically, there is a preponderance of males and of children, because these groups associate more closely with animals than do females and adults, as seen in this meta-analysis. Basically, the cystic lesions can be single or multiple, and the wall of the hydatid cyst has three layers that can be distinguished on pathological examination: pericyst, ectocyst, and endocyst [89]. Daughter cysts, which may contain "hydatid sand" consisting of free scolices and fluid, develop from scolices [89]. The clinical course of hydatid disease of the CNS in children and adolescents simulates that of a slow-growing space-occupying tumor mass. According to the localization of hydatid cysts, it is possible to discuss CNS hydatidosis under two main headings: intracranial hydatid cysts and intraspinal hydatid cysts.

Intracranial hydatid cysts

Intracranial hydatid cyst presents with different clinical pictures depending on the involvement of intracranial structures. When a hydatid cyst reaches a considerable size, the clinical symptoms it causes depend on the age of the patient and the location of the cysts. Despite the large size of these cysts and their considerable mass effect, patients remain in remarkably good condition, with relatively little neurological deficit [61]. In contrast to focal neurological signs, such as hemiparesis and epileptic seizure, in adults, in children and adolescents the clinical picture usually includes the cardinal symptoms of increased intracranial pressure (IICP): headache and vomiting are the most common. On the other hand, clinical manifestations related to movement disorders and occlusive cerebrovascular diseases are extremely uncommon [47, 100, 103].

Hydatid disease of the CNS is most commonly located in the supratentorial region involving the territory of the middle cerebral artery, owing to the embolic nature of the infection, as seen in the present study. In pediatric or adolescent patients with otherwise unexplained multiple intracranial hydatidosis, the possibility of arterial embolism should be considered [96, 100, 103]. Infratentorial intraventricular and brain stem hydatid cysts have occasionally been reported from Turkey, including 1 case in the fourth ventricle, 2 in the pons and 3 in the aqueduct of Sylvius [2, 10, 37, 78, 79, 108]. In some patients, hydatid cysts have occupied two or even three cerebral lobes, but this fortunately had no negative effect on complete removal of the cysts. Moreover, extradural infestation caused by intracerebral cysts or by direct extension from calvarium is infrequently encountered [95].

Skull radiographs show the effects of IICP, such as thinning of the vault and diastases of sutures, in children and adolescents. In the pre-CT era, ventriculography and angiography were used in CNS hydatidosis, with low diagnostic specificity. At that time, angiography replaced air studies in the investigation of intracranial lesions. Today, in most patients the cystic lesions are diagnosed by CT scanning and/or MRI. These generally reveal a characteristic round, well-defined unilocular cystic lesion containing fluid with absorption values similar to that of CSF [5, 8, 9, 11, 31, 51, 85, 86, 89, 103, 104, 111]. Single cysts are found in the vast majority of patients, but multiple cysts can result from surgical, spontaneous, or traumatic rupture of a viable primary lesion [96, 100, 103]. Generally, they cause significant ventricular distortion and shifting of the midline structures. CT and MRI imaging have improved the diagnosis and also allowed better surgical planning of the cortical incision for evacuation of these lesions. In this sense, it is very important to determine the most superficial part of the cyst to the bone so as to reduce the risk of cyst rupture and contamination of the surrounding tissues by its content during operation and follow-up of patients in whom a cyst may have been ruptured during the surgical intervention [10, 54].

Differential diagnosis of a hydatid cyst against other cystic lesions, such as arachnoid cyst, porencephalic cyst and cystic tumors, is not difficult when the imaging features of these lesions are well known to both radiologists and neurosurgeons. There is no doubt that the diagnostic procedure of choice is MRI in these cases. Recent reports in the literature have described the MRI findings of CNS hydatidosis [2, 5, 11, 9, 35, 51, 85, 86, 89, 103]. Magnetic resonance imaging scans provide additional information about the layers of the cyst and the cyst content, which cannot be seen on CT [89]. Typically, the en-

docyst is also visible as a hyperintense rim on T1-weighted MRI [86, 89]. The pericyst seen on T2-weighted MRI as a hypointense rim shows contrast enhancement following i.v. administration of contrast medium, owing to its rich vascularization [85, 86, 89].

The treatment of hydatidosis is necessarily surgical, aimed at removal of the cysts intact without spillage of their content, and the resection should be as radical as possible because it may result in an anaphylactic reaction with circulatory collapse and cardiac arrest. In the current survey study, all but 1 of these children and adolescents had been operated on for extraneural hydatidosis in other clinics either before or after the neurosurgical intervention. In principle, preoperative diagnosis of hydatid disease is essential, because rupture and dissemination of the cyst may result in recurrence and even death. The Dowling-Orlando technique classically requires (1) a wide osteoplastic flap; (2) exposure of the cyst by a wide incision; (3) an inclined position of the head; and (4) saline irrigation by means of a rubber catheter [24, 73].

Another technique used in problematic cases is the tapping procedure, i.e. puncture with a needle followed by aspiration during surgery (PIAR technique). For example, in the case of deep-seated and tightly surrounded cysts that cannot be expelled by irrigation, the entire content of the cyst is aspirated and the collapsed cyst is lifted away. Although the area of spillage is irrigated with a scolicidal solution to prevent anaphylactic complications and recurrences, each puncture invariably leads to some spillage of the contents into the operative field, and cyst recurrences are very dangerous. The results of surgical treatment depend on several factors, including the location, size, and multiplicity of the cysts [34, 73, 93]. Unlike Ciurea et al. [28], I strongly believe that giant cysts with a thick wall are no more difficult to remove than smaller ones, which are prone to rupture. Furthermore, it is considered that patients with multiple cysts have high morbidity and mortality rates owing to the number of surgical interventions needed to remove all their cysts, but these patients frequently tolerate multiple surgical procedures [103]. The ratio of cysts to patients was 2.2, and the recurrence and mortality rates for intracranial hydatid cysts were 14% and 10%, respectively. Although it is rarely possible to expel a cyst intact from an extradural localization in the CNS without rupturing it, its rupture into this space fortunately does not inevitably lead to a severe anaphylactic response, in contrast to widespread subarachnoid dissemination.

In the postoperative period, subdural effusions and porencephalic cysts can occur as a complication in children and adolescents [32, 34, 60, 70, 91]. The shunting procedure is a technique used when postoperative CT/MRI shows ventricular dilatation, subdural effusion, and/or porencephalic cyst. Similarly, long-term antiepileptic therapy is required if the patient has a history of preoperative convulsions [70, 99]. Another interesting point in this survey is the presence of "infected" or "complicated" hydatid cysts leading to a misdiagnosis of cystic tumor [33, 73, 101].These solid lesions can be called "chitinomas," for a pathological description of acellular eosinophilic material with focal necrosis. Peripheral irregular, large homogeneous, multiple punctiform, or scattered calcifications are found in the majority of patients [101]. Although it is not easy to disclose the source of infection in these cases, a broad-spectrum antibacterial treatment is advocated for secondary infection.

The chemotherapy of human hydatid disease could be of great importance in patients with inoperable cysts or multiple cysts and in those who are unfit for surgery or may be suffering from recurrent disease [10, 11, 32, 34, 60, 70, 73, 87, 92, 96, 97, 102, 103]. Antihelminthic agents for the medical treatment of hydatid disease in both types of echinococcosis in humans as an adjunct to surgical intervention are mebendazole and albendazole [11, 32, 34, 60, 65, 87, 92, 97]. In 1988, Todorov et al. [92] reported a patient with multiple hydatid cysts who was successfully treated with albendazole. Consideration of the effects of both these drugs in the management of CNS hydatidosis reveals that so far their use has been successful in recurrent cases insofar as they have led to regression or arrest of growth of echinococcal lesions, probably by interfering with uptake of glycogen by the cestodes [33, 69, 80, 87]. I think that further randomized studies are now needed so that information allowing a correct decision on the role of these chemotherapeutic agents in CNS hydatidosis can be won.

Intraspinal hydatid cysts

Intraspinal hydatidosis differs from the condition with intracranial cystic lesions in that it is microvesicular and invasive in nature, which suggests the presence of a malignant disease [89]. It is possible that the mechanism of dissemination of the larvae resulting in spinal involvement involves the portal system in the majority of cases. In the bone, hydatid disease expands relatively slowly and asymptomatically, destroying the bone as a tumor does, as a result of "endogenous" vesiculation (formation of daughter cysts within the parent cyst) [102]. Nevertheless, it differs from the disease in soft tissue in that growth occurs along the lines of least resistance, i.e., along the intratrabecular spaces, with the formation of diverticulated cysts [20]. This results in dilatation of the bony spaces of the spongiosa and resorption of cancellous bone [20]. Then, in the extraosseous stage of hydatid infection, known as "exogenous" vesiculation, numerous cysts are formed in the extradural and paraspinal regions, causing impingement on nerve roots and the spinal cord [102]. In clinical practice, these cysts become manifest as radicular pain associated with objective sen-

sory and motor disturbances and local tenderness at the level of the involved vertebrae in childhood and adolescence. In brief, the disease begins in the vertebral body and is usually diagnosed when complications caused by nerve root and spinal cord compression appear, as they do in most cases [77, 94].

In hydatidosis involving the spine, extension into the extradural space or paraosseous soft tissues may not be detected by spine radiograph and its radiological features are not pathognomonic, being indistinguishable from those of a tumor or infection of the bone. Therefore, it was confused with spinal tuberculosis and brucellosis and also with spinal neoplasms in Turkey in earlier years [77, 94]. Computed tomography scanning and MRI have been the most reliable diagnostic methods for the past 10–20 years. The imaging features of intraspinal hydatid cyst usually comprise multiple spherical lesions with clearly defined borders, containing fluid that is similar to CSF in intensity CSF on both CT and MRI [1, 11, 16, 17, 44, 51, 94, 102]. At present, the imaging technique of choice is MRI, since it allows early diagnosis of spinal cord compression and more accurate localization of intraspinal and paraspinal infestation if multiplanar images are taken [102]. With the use of MRI, it is possible both to detect the exact anatomical location of the lesion and to show the viability of the hydatid cysts [51, 103]. On T2-weighted images, a cystic lesion has a high intensity, and a decrease in hyperintensity indicates a dead cyst [51]. Thus, a decreased signal in this sequence is a characteristic indicator of a dead cyst. Furthermore, the layers of the cyst can only be demonstrated by MRI, and the pericyst may show enhancement after contrast material administration owing to its rich vascularization [89].

Surgical decompression and chemotherapy are the principal forms of treatment in pediatric and adolescent cases of intraspinal hydatidosis. Interestingly, endoscopic spinal surgery was also prescribed as an option for the treatment of intraspinal hydatid cysts in 1996 [1]. Unfortunately, the recurrence rate of these lesions is high, and antihelminthic agents are recommended for at least 1 year after a neural decompression procedure [16]. The recurrence and mortality rates for intraspinal hydatid cysts were 22% and 6%, respectively, in the patients reported in the papers reviewed for this study. In a previous extensive review from Turkey, I found that recurrence rates were 5% and 34% in patients treated by surgery plus chemotherapy and by surgery alone, respectively [94]. Antihelminthic drugs are currently being used more often for recurrence, systemic involvement, intraoperative rupture or puncture, poor suitability for surgery, and prophylaxis [10, 11, 32, 94, 97]. Recently, Baykaner et al. [16] reported on a child with a viable residual hydatid cyst that was cured with albendazole on control MRI. In fact, it is worthy of note that patients at high risk for intraspinal hydatidosis should be subjected to long follow-up to allow diagnosis and treatment of disease recurrence at an early stage.

Another important fact drawn to my attention by the current meta-analysis is that the number of authors per article has increased markedly in Turkey in recent years owing to the proliferation of senior authors in the major regional referral centers. Certainly, senior authors' intellectual contributions have an important role in coauthored papers on this subject. For this reason, multiauthor publications have become more common in the recent medical literature. A further reason for this is thought to be the development of the interdisciplinary team approach in the past decade.

Conclusions

According to the results of this survey study, the optimal management strategy (Fig. 1) for a child or adolescent with CNS hydatidosis will depend on whether any of the following is/are present

1. Multiple cysts
2. Cysts ruptured during surgery
3. Punctured and aspirated cysts
4. Inoperability
5. Bone involvement
6. Systemic involvement
7. Recurrence

Although the current surgical management of hydatid disease essentially has not greatly changed, some en-

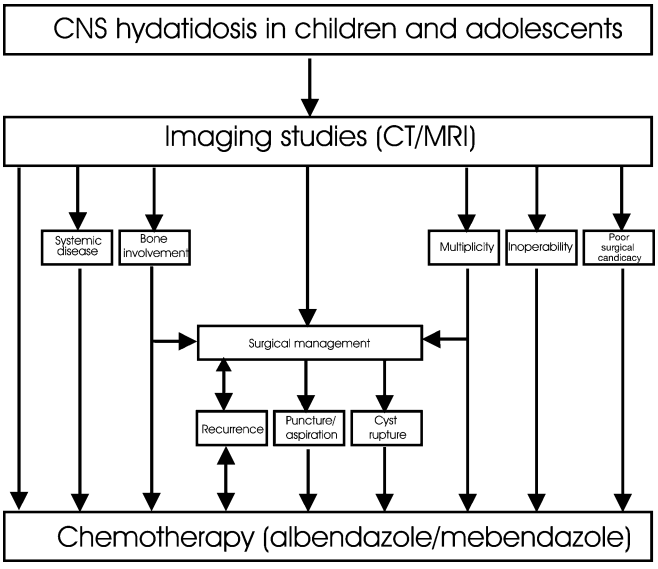

Fig. 1 Algorithm depicting the imaging and treatment management pathways for central nervous system (CNS) hydatidosis in pediatric and adolescent patient groups

couraging results obtained with chemotherapy have started to appear in the recent literature. Conclusions based on scientific publications by Turkish authors on clinical, imaging, and therapeutic features of CNS hydatidosis include the following notions.

1. Turkey is one of the countries where hydatidosis caused by the *Echinococcus granulosus* strain is a common parasitic disease in the pediatric and adolescent age groups.
2. The numbers of biomedical publications produced by Turkish authors on hydatid disease of the CNS and its coverings have risen tremendously over recent years even allowing for overlapping publications with the same data reported by the same or different authors.
3. Specific symptoms are decidedly lacking in pediatric or adolescent hydatidosis cases, and a diagnosis of hydatid disease was never suspected at presentation in any of the patients reported in the papers reviewed for this study. Therefore, a high index of suspicion is required to diagnose this condition.
4. Since the advent of the CT scan and later of MRI, therapeutic strategies have become well documented in the literature. In the vast majority of cases, both preoperative diagnosis and response to chemotherapy are best monitored by serial imaging studies.
5. In fact, the evaluation of any patient with intracranial cystic lesions should include the diagnostic probability of parasitic disease. For this reason, it should be kept in mind when a child or adolescent presents with the symptoms of IICP, spinal cord compression, or progressive myelopathy, especially in areas where parasitic disease is endemic, as it is in Turkey.
6. The intracranial form of the disease is usually seen as one single cyst, and multiplicity is more common feature of vertebral echinococcal cysts than of intracranial ones. The possibility of recurrence must always be kept in mind, particularly in patients with intraspinal involvement by hydatid disease.
7. Whole-body screening for systemic hydatidosis with CT scanning and ultrasonography is a useful combination both for achieving a correct diagnosis and for the planning of appropriate treatment.
8. Complete removal of the cysts intact without rupture remains the best surgical therapy, and surgery plus adjuvant chemotherapy is advocated as the gold standard of therapy. In cases with multiple cysts caused by parasitic embolism, however, surgical removal of the primary source is also very important as it is the only way of preventing recurrences.
9. During the surgical intervention, everything should be done to prevent spillage of cyst fluid because of the danger of anaphylactic shock and the high recurrence rate following cyst rupture.
10. Chemotherapy should be also considered in complicated intracranial and intraspinal hydatid disease, because of the increased risk of surgery. In general, proper medical and surgical treatment are the keystones in the achievement of declining morbidity and mortality in patients with CNS hydatidosis. With regard to chemotherapy, however, further randomized studies are necessary, with the aim of decreasing morbidity and mortality in CNS hydatidosis.
11. As a rule, long-term follow-up is mandatory in each case of CNS hydatidosis before any conclusion can be drawn about the value of any diagnostic or therapeutic procedure.
12. The aforementioned data indicate the complexity of the national and international problem posed by hydatidosis in Turkey, located as it is between Europe and the Middle East, because its incidence is so high. More importantly, they indicate the necessity for close international collaboration among the countries in the region, even though it is not (yet) so frequently seen in some developed Western countries, because of the increasing migration of large human groups from parts of the world where it is endemic to areas where it is not.

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References

1. Açıkgöz B, Sungur C, Özgen T, Çamurdanoğlu M, Berker M (1996) Endoscopic evacuation of sacral hydatid cyst: case report. Spinal Cord 34:361–364
2. Akdemir G, Ergün R, Gezici AR, Ökten AI, Ergüngör F (2000) Multiple hydatid cysts of the aqueduct of Sylvius: a case report with MRI study. Turk Neurosurg 10:142–144
3. Akduygu TL, Aslan Ö, Akdemir AO, Çolak A (2001) Spinal hydatid cyst: an evaluation of growth rates of cysts with paraspinal soft tissue localization (in Turkish). TND XV. Bilimsel Kongresi Özet Kitabı, p 240
4. Akhan O, Dinçer A, Saatçi I, Gülekon N, Besim A (1991) Spinal intradural hydatid cyst in a child. Br J Radiol 64:465–466
5. Aksungur EH, Oğuz M, Bıçakçı K, Altay M (1994) Cerebral hydatid lesion with daughter cysts: MR and CT findings (letter). AJR Am J Roentgenol 163:1269–1270
6. Aksüyek Ç, Karagöl U, Sayar N, Balkancı F, Arslan Z (1986) Intracerebral hydatid cyst. Case report (in Turkish). Nöroloji 14–15:113–119

7. Altınok A, Tancar F, Uçar B, Dikilitaş A, Keykubat N (1989) Spinal hydatid cysts (in Turkish). Türk Nörşir Derg Ek 1:58–61
8. Altınörs N, Kars Z, Çepoğlu C, Gürses L, Sağbil S, Arıyürek M (1991) CT findings and surgical treatment of double intracranial echinococcal cysts. Infection 19:110–114
9. Altınörs N, Şanlı M, Arıyürek M (1994) MRI imaging of intracranial alveolar echinococcal cysts (in Turkish). Bilgisayarlı Tomografi Bülteni 3:157–158
10. Altınörs N, Şenvelı E, Dönmez T, Bavbek M, Kars Z, Şanlı M (1995) Management of problematic intracranial hydatid cysts. Infection 23:283–287
11. Altınörs N, Bavbek M, Caner HH, Erdoğan B (2000) Central nervous system hydatidosis in Turkey: a cooperative study and literature survey analysis of 458 cases. J Neurosurg 93:1–8
12. Altıntaş N (1998) Cystic and alveolar echinococcosis in Turkey. Ann Trop Med Parasitol 92:637–642
13. Aydın MD (2001) Quadruplet hydatid cyst floating within the right lateral ventricle of the brain: case report (in Turkish). TND XV. Bilimsel Kongresi Özet Kitabı, pp 175–176
14. Aydın Y, Aydın F, Türe U (1992) Intradiploeic and cerebral hydatidosis: a case report and review of literature. Clin Neurol Neurosurg 94:229–233
15. Başak O, Turgut M, Aydın N, Gürel M (1998) [Unilocular cystic echinococcosis in Aydin] (in Turkish). T Parazitol Derg 22:262–267
16. Baykaner MK, Doğulu F, Öztürk G, Edalı N, Talı T (2000) A viable residual spinal hydatid cyst cured with albendazole. Case report. J Neurosurg [Spine 1] 93:142–144
17. Berk C, Çiftçi E, Erdoğan A (1998) MRI in primary intraspinal extradural hydatid disease: case report. Neuroradiology 40:390–392
18. Beşe S (1944) Two cases of hydatid cyst (kyste hydatique) presenting as a tumor in the spinal cord and brain (in Turkish). Anadolu Kl 11:106–109
19. Beşkonaklı E, Çaylı S, Yalçınlar Y (1996) Primary intracranial hydatid cyst extending above and below the tentorium. Br J Neurosurg 10:315–316
20. Braithwaite PA, Lees RF (1981) Vertebral hydatid disease: radiological assessment. Radiology 140:763–766
21. Canbolat A, Çoban T, Barlas O, Kaya U, Tolun R (1987) Extradural retroclival hydatid cyst. Med Bull İstanbul 20:129–134
22. Canbolat A, Önal C, Kaya U, Çoban TE (1994) Intracranial extradural hydatid cysts: report of three cases. Surg Neurol 41:230–234
23. Canda MS, Canda T (1995) Echinococcosis: report of 47 cases with hydatidosis in Turkey (in Turkish). T Parazitol Derg 19:64–82
24. Carrea R, Dowling E Jr, Guevara JA (1975) Surgical treatment of hydatid cysts of the central nervous system in the pediatric age (Dowling's technique). Childs Brain 1:4–21
25. Çataltepe O, Çolak A, Özcan OE, Özgen T, Erbengi A (1992) Intracranial hydatid cysts: experience with surgical treatment in 120 patients. Neurochirurgia (Stuttg) 35:108–111
26. Çataltepe O, Tahta K, Çolak A, Erbengi A (1991) Primary multiple cerebral hydatid cysts. Neurosurg Rev 14:231–234
27. Çetinalp E, Göçer AI, Bağdatoğlu H, Doğanay M, Kayacan M (1991) Craniospinal hydatidosis. Ç Ü Tıp Fak Derg 16:189–193
28. Ciurea AV, Vasilescu G, Nuteanu L, Carp N (1995) Cerebral hydatid cyst in children. Experience of 27 cases. Childs Nerv Syst 11:679–685
29. Demir K, Karslı AF, Kaya T, Devrimci E, Alkan K (1991) Cerebral hydatid cysts: CT findings. Neuroradiology 33:22–24
30. Dew HR (1995) Primary cerebral hydatid disease. Aust N Z J Surg 24:261–271
31. Diren HB, Özcanlı H, Bölük M, Kılıç C (1993) Unilocular orbital, cerebral and intraventricular hydatid cysts: CT diagnosis. Neuroradiology 35:149–150
32. Erdinçler P, Kaynar MY, Babuna O, Canbaz B (1997) The role of mebendazole in the surgical treatment of central nervous system hydatid disease. Br J Neurosurg 11:116–120
33. Erongun U, Özkal E, Acar O, Kocaoğullar Y (1994) Multiple and infected cerebral hydatid cysts. Case report. Neurosurg Rev 17:77–81
34. Erşahin Y, Mutluer S, Güzelbağ E (1993) Intracranial hydatid cysts in children. Neurosurgery 33:219–224
35. Erşahin Y, Mutluer S, Demirtaş E, Yurtseven T (1995) A case of thalamic hydatid cyst. Clin Neurol Neurosurg 97:321–323
36. Gedikoğlu Y, Çolak A, Bertan V (1989) Parasellar hydatid cyst. Case report. J Pediatr Neurosci 5:173–175
37. Gökalp HZ, Erdoğan A (1988) Hydatid cyst of the aqueduct of Sylvius. Case report. Clin Neurol Neurosurg 90:83–85
38. Gökalp HZ, Kanpolat Y, Gökben B (1979) Hydatid cyst of the skull. Neurosurgery 4:431–433
39. Gökalp HZ, Çağlar I, Karan O, Gürsoy F (1986) Cerebral hydatid cyst (in Turkish). Nöroloji 14–15:121–125
40. Gökalp HZ, Dinçer C, Şekerci Z, Birler K (1989) Cranial hydatid cysts (in Turkish). Türk Nöroşir Derg 1:65–71
41. Gökay H (1968) Vertebral hydatid cysts (à propos of 10 cases) (in Turkish). Türk Tıb Cem Mec 34:165–178
42. Gökay H, Adige F (1962) A case of cerebral hydatid cyst treated with surgical approach (in Turkish). Tıb Dünyası 35:236–239
43. Gürçay S, Özer F, Özbakır S, Emmezoğlu A (1986) A case of cerebral hydatid cyst presenting with chorea (in Turkish). Nöroloji 14–15: 127–133
44. Haliloğlu M, Saatçi I, Akhan O, Özmen MN, Besim A (1997) Spectrum of imaging findings in pediatric hydatid disease. AJR Am J Roentgenol 169:1627–1631
45. Hancı M, Aydıngöz Ö, Tanrıverdi T, Koçer N (2002) Primary hydatid cyst of lumbar spinal canal (in Turkish). Annual Meeting of the Turkish Neurosurgical Society, 03–08 June, 2002, İstanbul
46. İplikçioğlu AC, Özer AF, Benli K, Işık N, Erbengi A (1989) Multiple cerebral hydatid cysts: report of two cases. Br J Neurosurg 3:217–219
47. İplikçioğlu AC, Özek MM, Özer AF, Özgen T (1992) Periventricular hydatid cyst presenting with hemichorea. Childs Nerv Syst 8:292–293
48. İşlekel S, Erşahin Y, Zileli M, Oktar N, Öner K, Övül I, Özdamar N, Tunçbay E (1998) Spinal hydatid disease. Spinal Cord 36:166–170
49. İşlekel S, Zileli M, Erşahin Y (1998) Intradural spinal hydatid cysts. Eur Spine J 7:162–164
50. İyigün Ö, Uysal S, Sancak R, Hökelek M, Uyar Y (2001) A case of intracranial hydatid cyst in a two year old boy. XXth International Congress of Hydatidology, 4–8 June 2001, Kuşadası, Turkey; Congress Book p 358
51. Jena A, Tripahti RP, Jain AK (1991) Primary spinal echinococcosis causing paraplegia. Case report with MR and pathological correlation. AJNR Am J Neuroradiol 12:560
52. Kadıoğlu HH, Tüzün Y, Çiftçioğlu MA, Aydın MD, Aydın IH (1995) Intracranial hydatid cyst. Clinical analysis (in Turkish). AÜTD 27:110–113
53. Karadereler S, Orakdöğen M, Kılıç K, Özdoğan C (2002) Primary spinal extradural hydatid cyst in a child: case report and review of the literature. Eur Spine J (in press)
54. Kaya U, Özden B, Türker K, Tarcan B (1975) Intracranial hydatid cysts. Study of 17 cases. J Neurosurg 42:580–584
55. Kayacan M, Bağdatoğlu H, Uzuneyüpoğlu S, Boyar B, Hacıyakupoğlu S, Göçer I (1992) Posterior fossa hydatid cysts. Ann Med Sci 1:39–44

56. Kemaloğlu S, Özkan Ü, Aksu Y, Gönenç B, Aktaş G, Ceviz A (2001) Growth rate of primary cerebral hydatid cyst: case report and review of the literature (in Turkish). TND XV. Bilimsel Kongresi Özet Kitabı. p 257
57. Kemaloğlu S, Özkan Ü, Bükte Y, Acar M, Ceviz A (2001) Growth rate of cerebral hydatid cyst, with a review of the literature. Childs Nerv Syst 17:743–745
58. Kılıç C, Demirkazık M, Boyar B, Akalan N, Erdem H (1989) Spinal hydatid cysts (in Turkish). Türk Nöroşir Derg Ek 1:62–64
59. Kılıçhan E (1958) Vertebral hydatid cyst (in Turkish). Türk Tıp Cem Mec 6:311–317
60. Kocaman S, Erşahin Y, Mutluer S (1999) Cerebral hydatid cysts in children. J Neurosci Nurs 31:270–277
61. Krajewski R, Stelmasiak Z (1991) Cerebral hydatid cysts in children. Childs Nerv Syst 7:154–155
62. Küçükali I, Sofuoğlu E, Toplamoğlu H (1996) Cerebral hydatid cyst: a case report with definitive diagnosis (in Turkish). Düşünen Adam 9:58–61
63. Kurtsoy A, Öktem IS, Koç RK, Akdemir H, Menkü A, Tucer B (1999) Successful surgical treatment of a thalamic hydatid cyst with contralateral transcallosal approach. Case report and review of the literature. Pediatr Neurosurg 31:96–99
64. Merkle EM, Schulte M, Vogel J, Tomczak R, Rieber A, Kern P, Goerich J, Brambs HJ, Sokiranski R (1997) Musculoskletal involvement in cystic echinococcosis: report of eight cases and review of the literature. AJR Am J Roentgenol 168:1531–1534
65. Morris DL, Richards KS (1992) Hydatid disease, current medical and surgical management. Butterworth-Heinemann, Oxford
66. Nazaroğlu H, Özateş M, Bilici A, Şimşek M(1999) Multilocular cerebral hydatid disease withe extracalvarial extension. AJR Am J Roentgenol 172:1455–1456
67. Oğuz F, Sıdal M, Atabek A, Hamamıoğlu MK (1992) Hydatid cyst of the brain – case report (in Turkish). İst Tıp Fak Mecm 55:475–482
68. Önal Ç, Canbolat A, Gökay H, Kaya U, Türker K, Turantan MI, Barlas O, İzgi N, Ünal F (1992) Spinal hydatid cysts (in Turkish). Türk Nöroşir Derg Ek 5:34
69. Önal Ç, Barlas O, Orakdöğen M, Hepgül K, İzgi N, Ünal F (1997) Three unusual cases of intracranial hydatid cyst in the pediatric age group. Pediatr Neurosurg 26:208–213
70. Önal Ç, Ünal F, Barlas O, İzgi N, Hepgül K, Turantan MI, Canbolat A, Türker K, Bayındır C, Gökay HK, Kaya U (2001) Long-term follow-up and results of thirty pediatric intracranial hydatid cysts: half a century of experience in the Department of Neurosurgery of the School of Medicine at the University of İstanbul (1952–2001). Pediatr Neurosurg 35:72–81
71. Önal Ç, Yakıncı C, Erten F, Erguvan R, Çaylı S, Gül A, Aydın E (2001) Supratentorial hydatid cyst with cerebellar signs: a rare case of diaschisis. Childs Nerv Syst 17:746–749
72. Önder A, Aydın IH, Ak HE, Takçı E (1990) Vertebral hydatid cysts (in Turkish). Atatürk Üniv Tıp Fak Bült 22:253–258
73. Özek MM (1994) Complications of central nervous system hydatid disease. Pediatr Neurosurg 20:84–91
74. Özgen T, Erbengi A, Bertan V, Sağlam S, Gürçay Ö, Pırnar T (1979) The use of computerized tomography in the diagnosis of cerebral hydatid cysts. J Neurosurg 50:339–342
75. Özgen T, Bertan V, Kansu T, Akalın S (1984) Intrasellar hydatid cyst. J Neurosurg 60:647–648
76. Özkan Ü, Kemaloğlu MS, Selçuki M (2001) Gigantic intracranial mass of hydatid cyst. Childs Nerv Syst 17:623–625
77. Pamir MN, Akalan N, Özgen T, Erbengi A (1991) Spinal hydatid cysts. Surg Neurol 21:53–57
78. Paşaoğlu A, Orhon C, Akdemir H (1989) Multiple primary hydatid cysts of the brain. Turk J Pediatr 31:57–61
79. Peker S, Çelik H, Eryılmaz M, Gürçay Ö (1991) Iohexol CT ventriculogram of multiple cerebral hydatid cysts. AJNR Am J Neuroradiol 12:797
80. Postalcı L, Demirgil B, Dalgıç A, Oral Z (2001) Porencephalic cavity case with mass effect after excision of hydatic cyst (in Turkish). Türk Nöroşir Derg 11:140–144
81. Saltuk E, Yalçınlar Y, Aral Ö (1968) A cerebral cyst hydatic case treated by operation showing extrapyramidal symptoms (in Turkish). Nöro-Psikiyatri Arşivi 5:182–184
82. Sarıbaş S (1947) Cases of hydatid cysts in the spine (in Turkish). Ankara Üniv Tıp Fak Mecmuası 1:127–183
83. Sarıbaş S (1948) Cases of hydatid cysts in the spine. Acta Med Turcica 1:51–58
84. Saygı G (1996) Hydatidosis in Turkey within the last fourteen years (1979–1993). Cumhuriyet University Press, Sivas
85. Şener RN (1996) Thalamic hydatid cyst: contrast-enhanced MR imaging findings. Comput Med Imaging Graph 20:395–398
86. Şener RN, Savaş R, Yalman O, Alper HH (1998) Cerebral hydatid cysts in children: unusual imaging findings. Kuwait Med J 30:117–120
87. Singounas EG, Leventis AS, Sakas DE, Hadley DM, Lampadarios DA, Karvounis PC (1992) Successful treatment of intracerebral hydatid cyst with albendazole: case report and review of the literature. Neurosurgery 31:571–574
88. Talaslıoğlu A, Ersoy AÖ, Soyuer A, Köksal E, Tomar A (1994) A case of cerebral embolism caused by a cardiac hydatid cyst (in Turkish). XXX. Ulusal Nöroloji Kongresi, 9–14 Ekim, Adana
89. Talı ET (1999) Cerebral hydatid cysts. Riv Neuroradiol 12 [Suppl 3]:121–123
90. Tarcan B (1961) Hydatid cyst of the brain. A study of eight cases. J Int Coll Surg 36:334–335
91. Tevrüz M, Bertan V (1968) Intracranial hydatid cysts with different anatomic location and secondary dissemination (in Turkish). Çocuk Sağ Hast Derg 10:84–89
92. Todorov T, Vutova K, Petkov D (1988) Albendazole treatment of multiple cerebral hydatid cysts. Case report. Trans R Soc Trop Med Hyg 82:150–152
93. Tümer B, Yolaş C, Könte H, Aydın IH, Aydın Y (1983) Intracranial hydatid cysts (in Turkish). Atatürk Üniv Tıp Fak Bülteni 15:73–83
94. Turgut M (1997) Hydatid disease of the spine: a survey study from Turkey. Infection 25:221–226
95. Turgut M (1997) Primary intracranial extradural hydatid cyst extending above and below the tentorium (letter). Br J Neurosurg 11:587–589
96. Turgut M (1998) Multiple primary cerebral hydatid cysts (letter). Br J Neurosurg 12:72–73
97. Turgut M (1998) The role of mebendazole in the surgical treatment of central nervous system hydatid disease (letter). Br J Neurosurg 12:289
98. Turgut M (1999) Reply to the letter from Dr. Erşahin and Dr. Çağlı (letter). Childs Nerv Syst 15:435
99. Turgut M (2001) Intracranial hydatidosis in Turkey: its clinical presentation, diagnostic studies, surgical management, and outcome. A review of 276 cases. Neurosurg Rev 24:200–208
100. Turgut M, Bayülkem K (1998) Cerebrovascular occlusive disease: hydatidosis. Childs Nerv Syst 14:697–699
101. Turgut AT, Turgut M (2002) Infected or complicated hydatid cyst case mimicking a primary brain tumor (letter). Neurosurg Rev 25:191

102. Turgut AT, Turgut M (2002) Vertebral hydatid cyst infection (*Echinococcus granulosus*): case report (letter). Eur Spine J 11:393–394
103. Turgut M, Benli K, Eryılmaz M (1997) Secondary multiple intracranial hydatid cysts caused by intracerebral embolism of cardiac echinococcosis: an exceptional case of hydatidosis. J Neurosurg 86:714–718
104. Tüzün M, Hekimoğlu B (1998) Hydatid disease of the CNS: imaging features. AJR Am J Roentgenol 171:1497–1500
105. Uğur HÇ, Attar A, Bağdatoğlu C, Erdoğan A, Egemen N (1998) Secondary multiple intracranial hydatid cysts caused by intracerebral embolism of cardiac echinococcosis. Acta Neurochir (Wien) 140:833–834
106. Vatansever M, Biliciler B, Aladağ MA, Çolak A (1996) A huge cerebral hydatid cyst associated with small liver cyst: a comparison of growth rates and sizes. Neurosurg Rev 19:123–126
107. Yıldızhan A, Özkul MH, Uluer S, Özcan H, Özkul N, Aral Ö (1989) Primary hydatid disease of cervical region: a case report (in Turkish). Bezm-i Alem Valide Sultan Vakıf Gureba Hastanesi Dergisi 16:61–65
108. Yılmazlar S, Aksoy K (1999) Approach via the floor of the fourth ventricle for hydatid cyst of the pons. Pediatr Neurosurg 31:326–329
109. Yolaş C, Gündoğdu C, Yakut C (1993) Dead cerebral hydatid cyst. A case report (in Turkish). Türk Nöroşir Derg 3:38–40
110. Yüceer N, Gökalp HZ (1998) Periventricular hydatid cyst. J Neurosurg Sci 42:173–175
111. Yüceer N, Güven MB, Yılmaz H (1998) Multiple hydatid cysts of the brain: a case report and review of the literature. Neurosurg Rev 21:181–184