



Giant hydatid lung cyst in non-endemic area

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ABSTRACT

Echinococcosis is a parasitic zoonosis in which humans are involved as an intermediate host. It is a very common pathology in the areas of Eastern Europe and Asia, where the main activity is represented by pastoralism and animal breeding. In humans, the most frequently affected sites are the liver (60–70%) and lungs (20–30%), manifesting themselves with symptoms such as abdominal pain, coughing, dyspnea, hemoptysis, up to cases of anaphylactic shock. In Italy it is not a common disease with an incidence of 2:100.000 inhabitants, and it's extremely rare in pediatric population. In this work we presented a case of a 11-years male patient came for a persistent cough, to whom was found a giant cystic mass in the thorax. Serological tests confirmed the diagnosis of echinococcosis. Patient underwent to thoracotomy to remove the cyst and he begun the antiparasitic therapy. Post-operative outcome was good and the patient is, until now, free of problems. Echinococcosis is a challenging problem for a pediatric surgeon, especially in non-endemic area, not only because it is not a common disease in pediatric population, but also for the surgical management that can presents some difficulties and complications in the post-operative course.

1. Introduction

Echinococcosis is a parasitic disease caused by the larva of *Echinococcus granulosus*. The adult echinococcus lives in the small intestine of the definitive hosts, canids (dogs, wolves, jackals, foxes, coyotes) that infest themselves by feeding on viscera contaminated by hydatid cysts and through the feces of the latter the eggs of the parasite are scattered in environment [1]. The man behaves like an accidental intermediate host, ingesting eggs. The larva is released in the intestine, enters the circulation and, via the portal venous or lymphatic route, reaches the liver, where it is retained by the liver filter and develops a hepatic hydatid cyst. From here, it can reach the pulmonary circle, localizing itself to the lung, or, by skipping the lung filter, reach the systemic circle, being able to establish itself in any organ. In fact, clinically, the most frequently affected sites are the liver (60–70%) and lungs (20–30%), manifesting themselves with symptoms such as abdominal pain, coughing, dyspnea, hemoptysis, up to cases of anaphylactic shock following breakage internal cyst [2]. Usually, however, they are asymptomatic and accidentally discovered. This happens, in particular, in the lung, that is an elastic tissue and can permit the grow of the cyst without symptoms. When an hydatid cyst measures more than 10 cm of diameter, it is defined as giant [3].

Surgery represents the first therapeutic choice in cases of large and symptomatic hydatid cysts. In case of intervention is important the choice of which kind of resection to perform (radical or partial), also in

order to manage the possible post-operative complications.

We present the case of an 11-year-old patient in whom, in the absence of obvious risk factors, an echinococcosis with the presence of hepatic and a giant pulmonary hydatid cysts was diagnosed. We focused our attention to describe the surgical and post-operative management, highlighting in particular the complications occurred.

1.1. Case report

At the beginning of March 2020 an 11-year-old male patient arrived at the Emergency Room. For rough cough for about two weeks, treated with antibiotic therapy without resolution of symptoms. Due to the respiratory symptoms and since the fact that the patient came from an endemic area of Sars-Cov-2 (Pesaro-Urbino area), he underwent our protocol (pharyngeal swab and isolation) until he was proven negative. Chest x-ray was performed highlighting the presence of left lung lesion. For this reason, chest-abdomen CT was performed with evidence of a cystic aspect lesion of 13 × 9 cm at the level of the upper lobe of the left lung (Fig. 1A and B) and of calcific lesion of 6 mm at the level of the IV hepatic segment. The patient had a negative history of risk factors for parasitosis, but in the presence of the radiological findings, serology for echinococcosis was performed and it showed a positive result. Albendazole therapy was initiated and it was decided to perform surgery for pulmonary cyst resection. A brain MRI was performed showing no lesions. We decided for an open thoracotomy due to the dimension of the

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cyst which made difficult a safe resection with a minimal invasive intervention.

1.2. Surgical intervention

In general anesthesia, through a left posterolateral thoracotomy at the IV intercostal space, a large, thick, whitish lobar pulmonary cyst was identified (Fig. 2). About 140 ml of clear liquid were extracted from the cyst through 14 G needle and 150 ml of 10% sodium chloride solution was introduced (Fig. 3). By opening the cyst wall, the presence of germinative membrane was identified, removed, and placed in an endoscopic bag (Fig. 4A and B). A large bronchopleural fistula was identified and closed by single stitches. Finally, the cyst wall was closed by a partial capitonage. Two 16 F thoracic drainages were positioned, respectively in the residual cyst cavity, anteriorly and in the cost-phrenic space, posteriorly. After 4 days the posterior drainage was removed.

On the 8th post-operative day, the patient presented a mild dyspnea with the radiological evidence of an atelectasis of the left lung. An urgent chest CT was performed which confirmed the presence of a left pulmonary atelectasis. In operative room was performed a bronchoscopy which found voluminous mucus plug which obstructs 80–90% of the lumen of the left main bronchus. The plug was diluted with washing and lung was inflated with positive pressure showing a good distension and patency of left bronchus, and then 80 mg surfactant is administered in 10 ml saline. Then a posterior drainage (20 Ch) was positioned. On 12th post-operative day, after a chest x-ray and an ultrasound that showed the absence of pleural effusion, the drainages were removed. The next days the course was regular and the patient was discharged in good general conditions on the 17th day.

At a 6 months follow up the patients is asymptomatic. The CT scan shows a residual pneumatocele (Fig. 5).

2. Discussion

Cystic echinococcosis is a zoonosis known since ancient times, with references even in the Babylonian Talmud in which is reported the discovery of hydatid cysts in the bowels of animals sacrificed during religious rites. It has a global diffusion, with endemic areas in Asia and Mediterranean Europe, with particular concentration in the areas with predominantly pastoralism activities [4]. In Italy it is not a common disease with an incidence of 2:100.000 inhabitants, with the exception of Sardinia and Sicily where the incidence is higher [5]. Although considerable progress has been made over time in the prevention and control of this pathology, in the endemic countries it represents a health problem of significant socio-economic impact [6]. The diagnosis can be challenging for a pediatric surgeon, especially in non-endemic area,

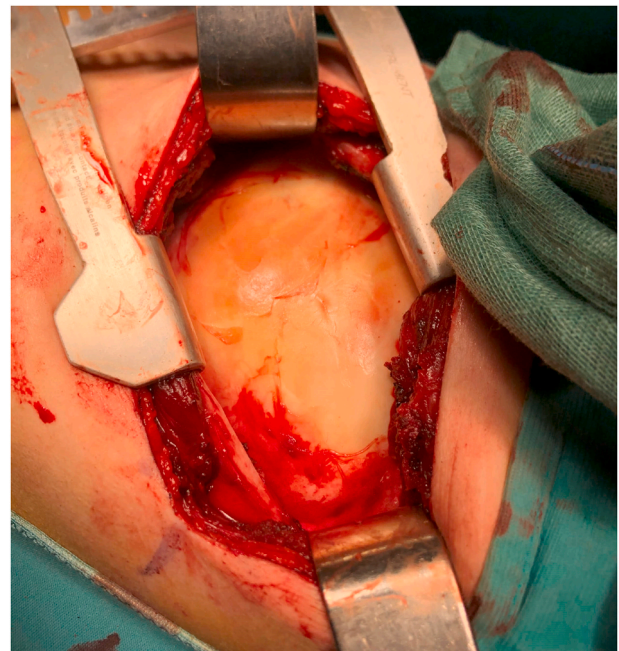


Fig. 2. Left posterolateral thoracotomy at the IV intercostal space and intra-operative finding of a large, thick lobar pulmonary cyst.

since the fact that it is not a common disease in pediatric population. The primary infection is usually asymptomatic and has an incubation period that can range from a few months to a few years. Cyst can be found accidentally during radiological examinations, performed for other reasons, or for the presence of symptoms in the case of cysts that reach significant dimensions (>5 cm) [7].

Nevertheless echinococcosis does not often represents a pediatric clinical entity, two series of cases are reported [8,9] in which the pathology presents itself as symptomatic in patients with an age range from 4 to 15 years.

In the presence of symptomatic and large cysts, surgery assumes a role of primary importance and has as objectives those of eliminating all the vital elements of the hydatid cyst (scolix, proligera membrane), avoiding the spillage of the cystic content and eliminating the capsulated elements of the cyst in order to resolve the mass effect on the surrounding tissues [10].

The major technical problem in surgical treatment is the complete resection, and, since these are cystic lesions solidary with the tissue in



Fig. 1. A–B: Chest CT-scan showing cystic aspect lesion at the level of the upper lobe of the left lung.



Fig. 3. Extraction of about 140 ml of clear liquid from the cyst through 14 G needle and introduction of 150 ml of 10% sodium chloride solution.

which they are formed (hepatic or lung parenchyma in the large percentage of cases), it is difficult to completely remove the neoformation without risking to damage seriously the organ.

So the surgical approaches of choice are essentially two, the conservative one and the radical one. The first aims to remove only the parasitic component (proliger membrane) and as much capsular cystic component as possible, while respecting the native tissue (tissue sparing technique). The second involves the complete removal of the hydatid cyst with sacrifice of part of the surrounding parenchyma (pericystectomy + hepatic/pulmonary resection) and greater onset of post-operative complications [11]. In pediatric surgery, the treatment of choice, especially in the case of cysts larger than 5 cm, is partial resection. In fact, in addition to being technically simpler, it guarantees the preservation of organ function which is one of the first objectives in pediatric patients. Although safe, this technique, however, exposes to risks such as that of recurrence, which in Literature is reported in a percentage ranging from 2% to 25% [12], and of fistula formation.

In 2015, Ran et al. [2] compared one group of pediatric patients

operated on with radical technique (n = 26) and one with conservative technique (n = 86), not showing a statistically significant difference in terms of recurrence.

In the case of pulmonary cyst, choice of the partial excision it could be useful to perform a capitonnage of the cyst, in order to fold with sutures the pericystic tissue after irrigation of 10% saline solution with scolicalidal effect. Infact pericystectomy in its entirety is not necessary and may cause complications such as severe air leak. On the other capitonnage can avoid the formation of a bronchopleural fistula or abscess in the remnant cavity [13].

When there is the need to treat an echinococcosis localized in the lungs, one of the major post-operative complications, especially in pediatric patients, is atelectasis. In a study of 2012 [14] this complication was present in the 17.5% of the patients. The causes of atelectasis in pediatric patients include the following: a lack of patient compliance and lack of appropriate analgesia resulting in retention of the secretion, dysfunction of the respiratory muscles, chest wall instability, and dislocation of the endotracheal tube during surgery. Oozing of blood and other fluids from bronchial openings in the cyst cavity into the bronchial system may appear as postoperative edema and atelectasis. To treat this condition is advisable to use ventilatory physiotherapy and inhalation of

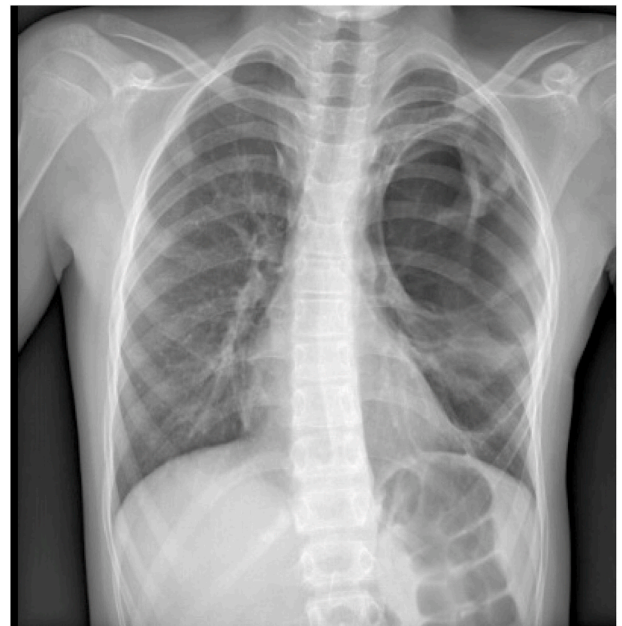


Fig. 5. Chest CT scan at 6 months follow-up highlighting a residual pneumatocele.

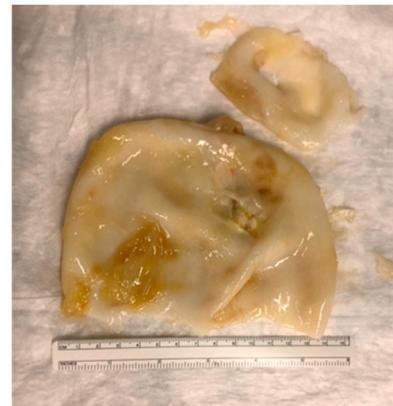
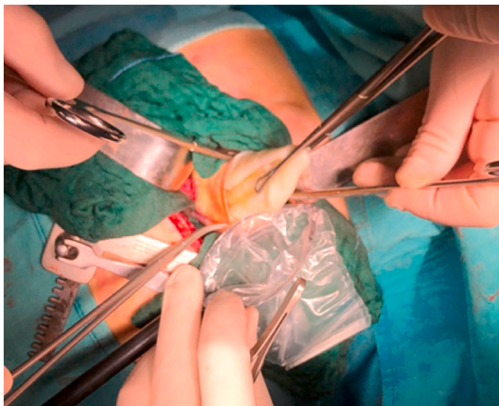


Fig. 4. AB: By opening the cyst wall, the germinative membrane was identified, removed, and placed in an endoscopic bag.

vapor and bronchodilator agents. However, unresolved atelectasis warrants a bronchoscopy [15] as in our experience.

In our case, the large size of the hydatid cyst, as well as making an open approach compared to the thoracoscopic one, have determined the choice of conservative technique at the outset. In fact, the complete removal of the cyst would have required the resection of an important portion of lung parenchyma, an unacceptable choice given the patient's age. In addition we performed the closure of the fistula with only a partial capitonnage in order to minimize the risk of air leak or abscess and, on the other side, to have a lower risk to cause atelectasis by obliterating, with sutures, the major bronchi surrounding the cyst cavity and present a consequent pulmonary parenchymal distortion. In our patient atelectasis was caused by a mucus plug and bronchoscopy gave us good results. The secretions were cleaned via a flexible fiberoptic bronchoscopy under general anesthesia and after that, a good expansion in the lung was achieved.

The choice of a partial capitonnage was, in our opinion, the safest one, in order to reduce the risk of air leak and infection and of atelectasis due to bronchial entrapment. Some authors noticed that capitonnage could cause atelectasis by obliterating the bronchus surrounding the cyst [16], and for this reason we decided to perform a partial closure of the cyst wall.

Presence of a pneumatocele caused by air trapping in the residual cavity, highlighted during the follow up, it's something encountered in Literature and usually it's free of risks [17]. There is no consensus agreement for the definition of a giant pulmonary hydatid cyst size. In terms of chest tube removal time, hospital stay, and complications such as pneumothorax and emphysema, atelectasis, and infection, there was no statistically significant difference between the patients with giant cysts undergoing cystotomy and the patients with giant cysts undergoing capitonnage. But the advantage was given to the capitonnage method in terms of residual cavity in the long term with good results and without any complications [18,19].

Despite the fact that currently the preferred approach in pediatric surgery is the minimally invasive one [20,21], this is not always suitable for all cases. In this type of pathology the open technique allowed us a good view of the cyst and allowed us to perform the partial enucleation without the risk of spillage.

We believe that our approach to this case, also considering the low Italian number of cases for this kind of disease, brought to good outcomes. In fact, the choice to eliminate the proliferating part, after irrigation of 10% saline solution with scolicedal effect, and the resection of an important portion of the pericystium represented a sensible choice in the management of the patient. In our experience, in fact, the radical nature in the removal of the parasitic part of the cyst and the control of any spillage represents an important guarantee in minimizing the risk of recurrence, also guaranteeing a low percentage of peri and post-operative complications.

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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