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# Diagnosis and surgical treatment of cysto-gastric fistula out of an hepatic hydatid cyst.

Unusual case

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## Diagnosis and surgical treatment of cysto-gastric fistula out of an hepatic hydatid cyst. Unusual case.

BACKGROUND: Hydatid cyst is an endemic zoonotic infection that annual incidence ranges from <1 to 200 per 100,000 individuals. The most common complication of hepatic hydatid cyst reported is rupture of the cysts, most commonly intrabiliary rupture. Direct rupture to hollow visceral organs is rarely seen. We describe here an unusual cystogastric fistula in a patient with liver hydatid cyst.

CASE PRESENTATION: The 55-year-old male patient presented with right upper quadrant abdominal pain. After radiological imaging studies, the diagnose was of hydatid cyst involving the left lateral segment of the liver ruptured into the gastric lumen and resulted in a cystogastric fistula. Gastroscopy revealed that the cyst and its contents protruding from anterior wall to the gastric lumen. Partial pericystectomy and omentopexy were performed and the gastric wall was primarily repaired. There were no complications in the postoperative period and 3-month follow up.

CONCLUSION: This case, to our knowledge, is the first reported case of cystogastric fistula surgically treated in a patient with liver hydatid cyst in the literature. Our clinical experience shows that, although it is a benign disease, complicated hydatid cysts should be evaluated in detail preoperatively, and after the detailed diagnostic work-up, surgical therapy might be planned individually for each case.

KEY WORDS: Cysto-gastric fistula, Hydatid Cyst, Liver hydatidosis

### Introduction

Hydatid cyst is an endemic zoonotic infection caused by Echinococcus species. The most frequent one is Echinococcus granulosus causing cystic echinococcosis. High prevalence rates are reported in parts of Eurasia (mostly in Central Asia and Mediterranean region),

Africa (northern and eastern regions), Australia and South America. The annual cystic echinococcosis incidence ranges from <1 to 200 per 100,000 according to endemicity of the region <sup>1</sup>.

Many patients remain asymptomatic for years. Those patients are diagnosed either by radiological

studies performed for other indications or after becoming symptomatic due to the complications.

Intrabiliary rupture of a hepatic hydatid cyst is the most common complication with a reported

incidence of 3-17%. The other less common complications are cyst rupture to the peritoneal or pleural cavities, fistulization to adjacent organs, and secondary bacterial infection <sup>2</sup>.

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Rupture into the hollow visceral organs are very uncommon. A cystoduodenal and a cystocolic fistula cases were reported in the literature <sup>3,4</sup>. This is, to our knowledge, the first case demonstrating the direct cystogastric fistula which was surgically treated.

#### Case Presentation

A 55-year-old male patient presented with pain in right upper abdominal quadrant for 40 days. The only comorbidity was hypertension. Vital signs were within the normal range. In the physical examination, there was a minimal sensitivity in the right upper abdominal quadrant. In blood workup, complete blood count, ALT, and AST levels were within normal limits, ALP and GGT levels were 120 U/L and 220 U/L, respectively. The bilirubin values were within normal range. Upper abdominal CT scan and MRI were performed. The abdominal CT scan revealed the hydatid cyst which was 12x6x6 cm in

dimension completely involving segment 2 and 3 of the liver. The CT scan also revealed the cystogastric fistula which was in 5 mm in 2 diameter and 8 mm in length, opening to the anterior wall of the stomach. The MRI revealed similar findings to the CT scan in addition to absence of cystobiliary fistula (Fig. 1).

Gastroscopy was performed and a sac thought to be hydatid cyst was seen on the anterior wall of lesser curvature. Fundus and cardia mucosa were observed to be normal (Fig. 2).

With these findings, surgical therapy was planned to the patient. In the exploration, the fistula tract was observed between the left lobe lateral segment of the liver and the anterior wall of the lesser curvature of the stomach. Partial pericystectomy and omentopexy were performed, fistula tract was excised and the fistula opening on the gastric wall was repaired (Fig. 3). The postoperative course was unremarkable, the patient was discharged without any complication. At the 3- month follow-up he did not report any problem.



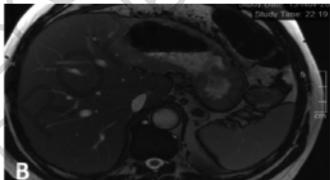


Fig. 1: A) Upper abdominal CT image of the patient. The arrow indicates that hydatid cyst involving segment 2 and 3 of the liver resulted in cystogastric fistula. Oral contrast matter filling to the cyst cavity can be seen. Upper abdominal MRI of the patient; B) The MRI revealed similar findings with CT in addition to absence of cystobiliary fistula.



Fig. 2: Endoscopic view of gastric hollow. The arrow indicates the fistula opening on the gastric wall. The cyst wall and the cystic contents inside are seen.

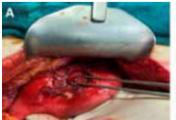




Fig. 3: A) The arrow indicates the cystogastric fistula opening on the anterior surface of the gastric wall. B) Intraoperative appearance of the cyst cavity.

#### Discussion

Treatment of complicated hepatic hydatid cysts is still controversial in the literature. Surgical therapy is advised by former authors in hepatic hydatid cysts communicating with the biliary tree, cysts fistulizing to adjacent organs and cysts exerting pressure on adjacent vital organs. There are many different surgical techniques described in the literature that range from simple cyst evacuation to pericystectomy and hepatic resections. It has been reported that radical surgical procedures may have lower recurrence and morbidity rates, yet they are also associated with higher intraoperative risk <sup>5</sup>.

We preferred to perform partial pericystectomy and omentopexy in this patient. Partial pericystectomy is defined as a safe surgical approach that not only minimizes blood loss and operative time, but also the recurrence and morbidity rates are quite low <sup>6</sup>. Omentopexy is advised in addition to partial pericystectomy procedure to prevent deep abdominal complications by filling the residual cavity after resection <sup>7</sup>. The patient was discharged without any complications and he didn't report any problem in 3-month follow up.

To our knowledge, this is the first reported hepatic hydatid cyst case with cystogastric fistula so there is not a specific treatment suggestion in the literature. In our opinion, in case of a fistula between hepatic hydatid cyst and gastrointestinal tract, the main goals of surgical treatment are complete removal of parasite, excision of the fistula tract and maintaining the continuity of the gastrointestinal tract.

#### Conclusion

A patient with a hydatid cyst complicated by cystogastric fistula and the treatment modality were described in this case report. Treatment of hydatid cyst disease should be tailored to the patient on a case-by-case basis. In complicated hydatid cyst cases, preoperative evaluation should be done meticulously and the surgical treatment should be carefully planned.

#### Riassunto

L'idatidosi è un'infezione zoonotica endemica che ha un'incidenza annuale varia da <1 a 200 per 100.000 individui. La complicazione più comunemente nota di una cisti idatidea epatica gnalata è la rottura delle cisti, più comunemente intrabiliare. Raramente si osserva la rottura diretta negli organi cavi viscerali. Descriviamo qui una fistola cistogastrica insolita in un paziente con cisti idatidea del fegato.

Si tratta di un uomo di 55 anni che si presentava con dolore addominale al quadrante superiore destro dell'addome. Dopo studio di imaging radiologico, la diagnosi era di cisti idatidea del segmento laterale sinistro del fegato rotto nel lume gastrico con fistola cistogastrica. La gastroscopia ha rivelato che la cisti e il suo contenuto sporgeva dalla parete anteriore dello stomaco.

È stata eseguita una pericistectomia parziale, seguita da omentopessia e la parete gastrica è stata suturata. Assenza di complicazioni nel postoperatorio e nel follow up a 3 mesi

CONCLUSIONE: Questo caso, a nostra conoscenza, è il primo caso riportato in letteratura di fistola cistogastrica trattata chirurgicamente in un paziente con cisti idatidea epatica. La nostra esperienza clinica mostra che, sebbene sia una malattia benigna, le complicazioni del le cisti idatidee dovrebbero essere valutati in dettaglio prima dell'intervento e dopo l'iter diagnostico accurato, e la terapia chirurgica potrebbe essere pianificata individualmente per ciascun caso.

#### References

- 1. Wen H, Vuitton L, Tuxun T, et :: Advances in the 21st Century. Clin Microbiol. Rev. 2019 Feb 13;32(2): e00075-18.
- 2. Alexiou K, Mitsos S, Fotopoulos A, et al. *Complications of Hydatid Cysts of the Liver: Spiral Computed Tomography Findings.* Gastroenterology Res. 2012;5(4):139-143.
- 3. Daldoul S, Moussi A, Zaouche A. Spontaneous fistulization of hepatic hydatid cyst into the duodenum: an exceptional complication. J Coll Physicians Surg Pak. 2013;23(6):424-6.
- 4. Greco, S., Cannella, R., Giambe*lluca, D. et al.* Complications of hepatic echinococcosis: multimodality imaging approach. Insights Imaging. 2019; 10: 113.
- 5. Sayek I, Tirnaksiz MB, Dogan R. Cystic hydatid disease: current trends in diagnosis and management. Surg Today. 2004;34(12):987-96.
- 6. Sozuer E, Akyuz M, Akbulut S. Open surgery for hepatic hydatid disease. Int Surg. 2014;99(6):764-769.
- 7. Dziri C, Paquet JC, Hay JM, et al. *Omentoplasty in the prevention of deep abdominal complications after surgery for hydatid disease of the liver: a multicenter, prospective, randomized trial. French Associations for Surgical Research.* J Am Coll Surg. 1999;188(3):281-89.