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ISOLATED VASCULITIS OF THE HEPATIC ARTERY: A RARE CAUSE OF POST-CHOLECYSTECTOMY ABDOMINAL PAIN IDENTIFIED IN INDIA

General Surgery

Dr. Monika Joshi Senior Resident, Department Of General Surgery, Apollo Hospitals, Navi Mumbai

Dr. Deepak Gupta Senior Consultant, Department Of Medical Gastroenterology, Apollo Hospitals, Navi Mumbai

Dr. Nitish Jhawar Senior Consultant And Head Of The Unit, Department Of General Surgery, Apollo Hospitals, Navi Mumbai

ABSTRACT

Post-cholecystectomy pain (PCP) is not uncommon, but many times the cause may be difficult to determine. Herein, we present a case of a rare isolated hepatic artery vasculitis, which had led to PCP. A 68-year-old female had presented to hospital, on day 13 after cholecystectomy, with complains of right upper quadrant pain and shortness of breath. The laboratory parameters were normal, with only c-reactive protein raised. Autoimmune work-up for antibodies was also negative. Analgesics failed to provide any relief. Ultrasonography of abdomen showed findings indicative of hepatic artery vasculitis with left mild effusion. Effusion was managed by pleural tapping, followed by computed tomography (CT)-angiography which confirmed the diagnosis of hepatic artery vasculitis. The patient was initiated on oral steroid, and the pain subsided. On a one-month follow-up post-surgery, USG-abdomen showed regression of vasculitis and subsidence of pleural effusion. This case highlights the importance of detailed work-up for abdominal pain, with rare causes like isolated vasculitis also being considered.

KEYWORDS

Post-cholecystectomy pain, hepatic artery vasculitis, single organ vasculitis.

BACKGROUND

Gallstone disease continues to be one of the major reasons of abdominal morbidity as well as mortality all over the globe. The clinical presentations comprise of acute cholecystitis as well as febrile illness with pain and tenderness in the right upper quadrant (known as Murphy sign). The typical surgical management for gall stones is cholecystectomy.[1] While most patients have pain alleviation after cholecystectomy, over 40% of people are expected to still experience discomfort after the procedure.[2] This represents a substantial percentage of individuals who continue to have health issues. Post-cholecystectomy pain (PCP) can manifest as either a continuation of the pain that was there before the cholecystectomy, or as a new discomfort altogether. Single-organ vasculitis (SOV) is defined as the localized inflammation of a blood vessel, specifically targeting one organ, such as the skin, genitourinary system, or the aorta, without any accompanying systemic characteristics. Gastrointestinal SOV involvement is uncommon, with hepatic artery involvement documented in only a couple of published cases. Here, we present a case wherein an isolated hepatic artery vasculitis was the reason behind PCP.

Case Report:

A 68-year female, who had undergone cholecystectomy, presented at the surgery outpatient department on day 13 after the surgery, with severe right upper quadrant pain and shortness of breath. The temperature and vital signs were within normal range. On examination, the tight side lower chest air entry was noted to be decreased. Routine laboratory investigations were run, which came out normal. Other laboratory tests were also run in the meantime, like anti-nuclear antibody (ANA), and antineutrophil cytoplasmic antibodies (ANCA), which came out to be negative, while c-reactive protein (CRP) was raised (98 mg/dl).

Chest x-ray showed left side mild pleural effusion. The ultrasonography of the abdomen was conducted, which showed cord like thickening of hepatic artery without evidence of thrombosis was seen with left mild effusion (Image 1a). In the meantime, the patient was started on analgesics, but did not show any improvement.

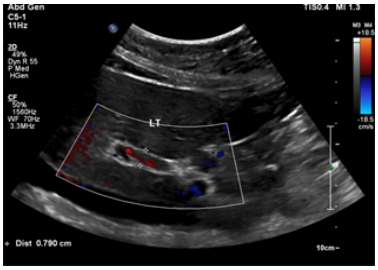

Image 1a shows the USG abdomen finding before steroid was initiated

On the next visit (day 15 after cholecystectomy), pleural effusion had worsened, and hence, pleural tapping was done. The evaluation of the pleural fluid showed protein levels of 2.1 gm/dl, and a leucocyte count of 80%. The patient was posted for computed tomography (CT)-angiography, and the results showed thickened intimal wall of hepatic artery up to 12 mm patent vessel with no thrombosis. The diagnosis of vasculitis of hepatic artery was noted.

The patient was started on oral prednisolone 1 mg/kg for 1 month, on day 17 after cholecystectomy. The oral steroid was then tapered with time for a total duration of 2.5 months. The patient also started on oral paracetamol with steroid, prescribed for a period of one week. The pain subsided by day 20 post-cholecystectomy, and the patient was called for follow-up after three weeks of starting oral steroid. A USG-abdomen was conducted at the follow-up, which showed near-complete regression of intimal thickening with symptomatic improvement and disappearance of pleural effusion (Image 1b).

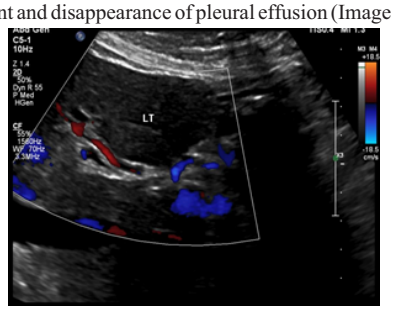

Image 1b shows the USG abdomen finding at follow-up, after steroid was administered

DISCUSSION

The usual causes of PCP were first considered for the mentioned case. A recent systematic review on the symptoms following cholecystectomy revealed several factors contributing to these symptoms. These factors include the presence of remaining or newly formed gallstones, the coexistence of other diseases such as irritable bowel syndrome, gastro-esophageal reflux, gastritis, peptic ulcer, anterior cutaneous nerve entrapment syndrome (ACNES), gastroparesis, and fatty liver disease. Psychological distress and dysfunction of the sphincter of Oddi (SOD) were also identified as potential causes.[3]

Since hepatic artery vasculitis is on its own a rare occurrence, and GI-SOV is not a common cause of PCP, it was very important to do thorough workup of the patient. To diagnose vasculitis, comprehensive

clinical assessment, together with a mix of laboratory tests and sophisticated imaging investigations, is necessary. In certain cases, a tissue biopsy may also be required to rule out other possible illnesses.[4] The most appropriate diagnosis for the case reported is SOV, which refers to inflammation of the arteries or veins in a specific organ, without any signs of involvement in the entire body.[5] Primary vasculitis frequently exhibits gastrointestinal (GI) tract involvement, notably in polyarteritis nodosa (PAN), ANCA-associated vasculitis, IgA vasculitis, and Takayasu arteritis. To rule these primary reasons out, we had run ANCA tests which came out negative.

The management of the patient involved the administration of steroids based on the general principles of the gastrointestinal (GI) tract's SOV. In our case, just like other published evidence, oral steroid was effective in tackling hepatic artery vasculitis. A case series undertaken by the Mayo Clinic over a span of 12 years discovered 18 instances of vasculitis that affected the GI tract.[6] All patients in this series underwent a comprehensive autoimmune investigation, which yielded negative results. The predominant symptom experienced by almost all patients was abdominal pain, accompanied by additional symptoms such as abdominal angina, nausea or vomiting, diarrhea, hematochezia, or melena. Out of the 18 patients, ten were administered prednisone, starting at a median dose of 60 mg per day (ranging from 30 to 100 mg per day). Meanwhile, three patients were given methylprednisolone pulse treatment, with a dosage of 1 g per day for 3 to 5 days, before switching to oral prednisone (median duration of 7.5 months). Out of the 10 patients who received therapy, eight showed a positive response. Among them, two achieved remission, five had recurrence, and three unfortunately passed away. The causes of death were hepatic abscesses, hepatic infarcts, and intestinal perforation, each accounting for one case. These findings show that if not managed adequately and in time, hepatic artery vasculitis can also lead to lethal outcomes. In a case study conducted by Mali et al., a patient suffering from hepatic artery vasculitis received treatment with methylprednisolone at a daily intravenous dose of 1000 mg for three consecutive days. This was followed by the administration of prednisone, which was gradually reduced over time.[7] During the six-month follow-up, the patient said that they experienced alleviation from abdominal discomfort, and there was an improvement in the levels of CRP. The abdominal CT scan showed a notable reduction in the density of the soft tissue around the common hepatic artery. Quiceno et al. recently reported an isolated vasculitis case of the hepatic artery. 18-fluorodeoxyglucose positron emission tomography (FDG-PET) was performed to identify inflammation in the hepatic artery and ascending colon by distinguishing it from other vascular problems based on metabolic activity. It facilitated additional diagnostic procedures and therapy by identifying the scope of the illness and prospective locations for biopsies. The patient received a diagnosis of SOV, affecting the hepatic artery. They had a three-day treatment with high-dose methylprednisolone, followed by a steroid-sparing regimen consisting of weekly doses of prednisone and methotrexate, each at a dosage of 15 mg. The patient's symptoms subsided, and she was released with ongoing gradual reduction of steroid dosage.[8] A follow-up for six months is very important for SOV cases as 26% of these GI cases may progress to a systemic manifestation.[5,7,9]

CONCLUSION

Hepatic artery vasculitis is a rare form of SOV, seldom reported in scientific literature. On literature search, this may be the first published case report from India, of a hepatic artery vasculitis. Moreover, no other published report mentions the possibility of hepatic artery vasculitis being the reason for pain after cholecystectomy. This underlines the importance of rare causes, which can be identified by thorough clinical examination and investigations. In line with the available evidence, immunosuppression is effective to reduce morbidity and mortality.

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