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ORIENTAL CHOLANGIOHEPATITIS : A RARE CASE REPORT WARRANTING SURGICAL ACCESS PROCEDURE

General Surgery

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ABSTRACT

Oriental cholangiohepatitis (OHC) aka Recurrent Pyogenic Cholangiohepatitis or Hong Kong Disease is a pathology predominantly found and reported in south east asian population. The disease manifests as recurrent right upper quadrant abdominal pain, fever and jaundice of obstructive profile. The disease is characterised by intra- and extra- hepatic bile duct strictures and dilatations with intraductal pigment stone formation. Diagnosis is based on typical clinical features and specific radiological findings.

Management consists of percutaneous biliary drainage procedures, biliary stone extraction and complex pathologies may require surgical access procedures.

KEYWORDS

oriental, cholangiohepatitis, arrowhead sign, access procedure, percutaneous biliary drainage, hong kong disease

INTRODUCTION:

Oriental cholangiohepatitis (OHC) aka Recurrent Pyogenic Cholangiohepatitis is a disease entity although not restricted to but found with staggering predominance in south east asian population. The exact etiology is still elusive although Clonorchis sinensis infestation, Ascaris lumbricoides infestation, Ascending cholangitis from gut E.coli flora, poor nutritional status and poor socio-economic status have been implicated with high index of suspicion. Clinical profile includes recurrent right upper quadrant abdominal pain, fever and jaundice of obstructive pattern.

Characteristic pathology consists of multiple strictures and dilatations of intra- and extra- hepatic bile ducts with intraductal pigment bile stones. There is bile duct proliferation and inflammatory cell infiltration along peri portal spaces and hepatic parenchyma. The bile collections in dilated ducts may get secondarily infected and patient may present with liver abscess.

MRCP provides the most specific imaging technique demonstrating multiple biliary strictures and bile collections; reduced arborisation of peripheral ducts known as "ARROWHEAD SIGN".

Management is within the increasing purview of Interventional Radiology. Most cases are managed by percutaneous biliary drainage and clearance of stones, balloon dilatation of biliary strictures and repeat percutaneous procedures to clear stones and mud-like biliary sludge.

Difficult pathologies , where endoscopic and percutaneous procedures do not suffice, the only resort that remains is surgical biliary access procedure.

Complications associated with the disease itself are biliary cirrhosis and cholangiocarcinoma (in ~5% cases).

CASE REPORT: A 26 year old female (resident of jammu and kashmir state of india) presented to the surgical department of Indraprastha Apollo Hospitals, New Delhi with chief complaints of recurrent right upper quadrant abdominal pain, fever and jaundice since 1998. She underwent cholecystectomy for gall bladder calculi and Choledochoduodenostomy for choledochal cyst in 1998. The symptoms have plagued her ever since in a periodic pattern. She gives a history of liver abscess 3 months before she presented to us which was treated conservatively. Patient underwent investigations at our institute on which LFT demonstrated an obstructive pattern with raised ALP (alkaline phosphatase) and 5'-nucleotidase. ELISA Assays for HBsAg (hepatitis B surface antigen), HIV-I & II and HCV (hepatitis C virus)were negative.

USG whole abdomen demonstrated dilated IHBR (intrahepatic biliary radicals); CBD Sludge and Pneumobilia.

MRCP demonstrated multiple biliary strictures and dilatations with bile filled spaces, biliary tract calculi and CBD stent in situ.

UGIE (upper GI Endoscopy) was done which demonstrated posterior wall opening in the first part of duodenum (of choledochoduoden ostomy) with stone impacted in the opening.

ERCP for biliary drainage could not be done due to stone in the opening.

In view of inability to manage the condition endoscopically, patient was considered for surgical management.

She underwent Roux-en-Y Hepaticojejunostomy with access loop under general anesthesia.

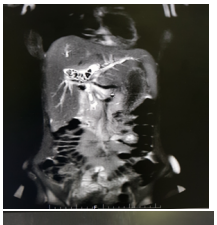
No significant intra-operative complicatons were encountered.

Post-operative recovery was uneventful and patient was managed on gradual diet escalation.

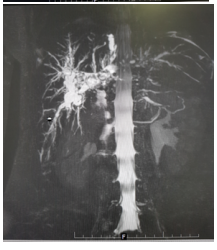
She was discharged tolerating oral feeds.

Post operative assessment showed symptom free patient and normal LFT's.

A)



B)



MRCP IMAGES

A) Dilated IHBR (intra hepatic biliary radicals) with multiple intrahepatic calculi
B) Multiple dilatations in intra hepatic biliary tree with reduced arborization of peripheral ducts (ARROWHEAD SIGN)

A)

B)

c)

USG ABDOMEN IMAGES

A) Pneumobilia
B) CBD Sludge
C) Dilated IHBR (intra hepatic biliary radicals)

DISCUSSION: Oriental cholangiohepatitis as a disease entity was first described in 1930.

Since then only it has undoubtedly shown a preponderance for the south east asian population.

It has been called by different names viz.Hong Kong disease, Oriental Cholangitis, Oriental infestational cholangitis. Its major etiological factors have been helminthic infestation (Ascaris lumbricoides, Clonorchis sinensis, Opisthoorchis viverrini); Gut Coliform infection and Malnutrition.

Malnutrition inhibits the deconjugation inhibiting enzymes such as the beta glucuronidase inhibitor glucaro-1,4-lactone. This causes uninhibited bilirubin deconjugation, which complexes with calcium to form calcium bilirubinate which in turn acts as a nidus for pigment stone formation. Clinical features consist of the same triad of pain, fever and jaundice as seen in cholangitis, the difference being jaundice of obstructive pattern and clinical features of recurrent nature. MRCP remains the most specific investigation for the disease and being helpful in assessing post intervention/surgery changes of disease pathology. Treatment will start with minimal access interventions by interventional radiologist for biliary drainage, stone extraction and stricture dilatation. Treatment for failed minimal access techniques or complex pathologies will ultimately lead to surgical access procedures.

CONCLUSION: OHC is a disease entity not so uncommon in the south east asian countries, due to relatively poor nutrition, socio-economic status and hygiene. Due to the progressive nature of the disease and cholangiocarcinoma being a well associated complication, a high index of suspicion should be exercised while coming across patients of cholangitis and liver abscess in this geographical belt. With evolving technologies in health care, minimal access procedures are gaining popularity but this case teaches us that complex disease will still require formal surgical management.

Undue repeated attempts of minimal invasive non surgical procedures in such complex cases may delay formal surgical management but cannot avoid them. Large scale studies must be under taken at high volume centres for the disease to ascertain if advanced cases should be tried first with repeated percutaneous techniques or should be offered appropriate formal surgical management at the first go to reduce the procedure related and financial burden on the patient.

REFERENCES

1. Aloreidi, K., Sethi, P., Yeager, T., & Atiq, M. (2017, August 27). Percutaneous Endoscopic Management for Oriental Cholangiohepatitis: A Case Report and a Brief Review of the Literature. https://www.ncbi.nlm.nih.gov/pubmed/28932607
2. Verweij, K. E., & Van, H. (2016, November). Oriental cholangiohepatitis (recurrent pyogenic cholangitis): a case series from the Netherlands and brief review of the literature. https://www.ncbi.nlm.nih.gov/pubmed/27905307
3. Parray, F. Q., Wani, M. A., & Wani, N. A. (2014, June 08). Oriental cholangiohepatitis - is our surgery appropriate?
4. https://www.ncbi.nlm.nih.gov/pubmed/24918695
5. Garg, D., & Gupta, M. (2012, December). MRI features of oriental cholangiohepatitis. https://www.ncbi.nlm.nih.gov/pubmed/22921337
6. Singla, S., Warner, A. H., Jain, A., Thomas, R. M., & Karachristos, A. (2012, March 16). Oriental cholangiohepatitis masquerading as cholangiocarcinoma: A rare presentation that surgeons need to know. https://www.ncbi.nlm.nih.gov/pubmed/22503913
7. Tsui, W. M., Lam, P. W., Lee, W. K., & Chan, Y. K. (2011, July). Primary hepatolithiasis, recurrent pyogenic cholangitis, and oriental cholangiohepatitis: a tale of 3 countries. https://www.ncbi.nlm.nih.gov/pubmed/21654363
8. Wani, N. A., Robbani, I., & Kosar, T. (2011, February). MRI of oriental cholangiohepatitis.
9. https:// www.ncbi.nlm.nih.gov/pubmed/21216332
10. Leow, C. K., & Lau, W. Y. (1998, January). Re: Biliary access procedure in the management of oriental cholangiohepatitis. https:// www.ncbi.nlm.nih.gov /pubmed/9457046
11. Gott, P. E., Tieva, M. H., Barcia, P. J., & Laberge, J. M. (1996, November). Biliary access procedure in the management of oriental cholangiohepatitis. https:// www.ncbi.nlm.nih.gov/pubmed/8895715
12. Lim, J. H. (1991, July). Oriental cholangiohepatitis: pathologic, clinical, and radiologic features. https://www.ncbi.nlm.nih.gov/pubmed/2048504
13. Lim, J. H., Ko, Y. T., Lee, D. H., & Hong, K. S. (1990, September). Oriental cholangiohepatitis: sonographic findings in 48 cases.https:// www.ncbi.nlm.nih.gov/ pubmed/2117346