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GASTROENTEROLOGY TRAINING DURING COVID 19 PANDEMIC

Dr Nandana Dinamithra

It is important to study on how this pandemic has impacted gastroenterology training across the globe. Most surveys demonstrate that GI training has been affected to various extents and encompasses all aspects – clinical, endoscopy, MDT meeting exposure, examinations and research. It has been demonstrated that the training was affected to various extents during the first wave of the pandemic, with respondents replying that their training was moderately to severely affected across the various aspects of training, these being: clinical gastroenterology training (67.4%), outpatient (75.5%) didactic teaching (88.3%), MDT meeting (65.2%) and endoscopy training (75.6%).

Didactic teaching was the aspect most strongly affected. This could be due to trainers having less time for teaching, trainees not having the opportunity to attend lectures and reluctance for all trainees to meet in one room. Online learning is not a new concept, and several societies have been offering online courses and lectures even before the pandemic. In a hospital setting, however, not all trainers might have the necessary apparatus, setting and know-how to deliver online lectures. Financial investment and upgrades in hardware and/or software might be needed in a time when resources are being directed elsewhere.

The failure to achieve the required skills within the time schedule has to be addressed. This may imply both a pre-empting of extension of training time as well as restructuring of the programme. Reasons for restructuring of the training programme include changes in the amount of exposure to gastroenterology training as well as new realities such as the introduction of online phone/video consultations. One has to question whether remote consultations have the same learning value as face-to-face consultations. Trainees not only have to consider the clinical question posed by the consultation; they also have to decide whether a physical examination is required. It also changes the concept of the usual supervised training in clinics. Thus, young trainees should be taught how to obtain reliable clinical data through online consultations.

The postponement of relevant gastroenterology examinations may also result in training extension as in some countries this would be a requirement for completion of training. Furthermore, the fact that 20.9% had fellowships postponed might lead to delay in training as well as the loss of acquisition of new skills among our future gastroenterologists. This is particularly a major concern in Sri Lankan trainees.

On a positive note, the trainees' accessibility to European gastroenterology conferences has remained there and possibly has become more accessible as these have gone both online and their prices have been reduced. Thus, the lower cost for the conference registration as well as the absence of travelling and accommodation costs should make such resources more readily accessible to all trainees and should also be considered and continued once the pandemic is over.

In conclusion these are difficult times ahead, and a training analysis will need to be done by each training centre in order to identify new lacunae in training, both in terms of quantity and quality. Improvements could include using online platforms for MDTs, increased use of simulators during endoscopy, organizing targeted training and utilizing alternative methods for didactic teaching. Furthermore, the psychological impact of all this on our trainees should also be taken care of. Our main aim as specialists is to defend the trainee's interest and provide the best and safest environment and resources for training as these are both our future gastroenterologists and trainers.

Epidermolysis Bullosa Acquisita as a rare cause of dysphagia: a case report

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Abstract:
Epidermolysis bullosa acquisita (EBA) is a rare, chronic relapsing-remitting autoimmune blistering skin disorder with potential mucosal involvement. Rarely dysphagia and oesophageal strictures have been reported resulting in significant weight loss and nutritional deficiencies. Involvement of mucosal membranes is associated with higher morbidity with life threatening complications. EBA is notoriously difficult to treat as there is no definitive treatment. EBA patients require lifelong regular follow up with increased vigilance for malignancy.

Here we describe a 50-year-old female with EBA, who presented with progressive dysphagia, found to have a benign upper oesophageal stricture, a rare complication of EBA.

Keywords: Epidermolysis Bullosa Acquisita, Dysphagia, Stricture, Case report

Introduction

Epidermolysis bullosa acquisita (EBA) is a relapsing-remitting, acquired autoimmune blistering skin disorder. Rarely dysphagia and oesophageal strictures have been reported in EBA causing significant weight loss and nutritional deficiencies. Here we describe a case of a patient with EBA, presenting with dysphagia, who was later found to have a high, benign oesophageal stricture which resulted in significant loss of weight and reduced quality of life.

Case presentation

A 50-year-old Sinhala, Sri Lankan female, previously diagnosed with a blistering skin disorder two years back presented to the Gastroenterology Outpatient Clinic at the Colombo North Teaching Hospital, Ragama, Sri Lanka, with progressive dysphagia to solids over a period of one year. She complained of "lump in throat" feeling with discomfort associated at mealtimes. Despite a normal appetite, there was a gradual weight loss of 10 kg in the preceding three months. There was no odynophagia, regurgitation, nausea or vomiting experienced. At presentation, she was managing to consume pureed food with liquid nutritional supplements. Following a skin biopsy for recurrent blistering rash in the preceding year, she had been diagnosed with EBA. She had no known allergies, previous autoimmune conditions or malignancies. There was no family history of autoimmune conditions or skin disorders.

On examination, she was pale with a body mass index (BMI) of 18.7kg/m2. She did not exhibit any other features of significant nutritional deficiencies. There were no neck lumps, lymphadenopathy or thyroid enlargement noted. Chest, abdomen and cardiovascular examinations were normal. She had significant scarring and millia on extensor surfaces of both lower limbs and upper limbs.

Her routine blood investigations revealed Hb 10.7 g/dl, MCV 89.9 fL, WBC 8.34 x 109/L, platelet 258 x 109/L with AST 38 U/L, ALT 39 U/L, Total bilirubin 1.9 mg/dl. Her renal function including serum electrolytes was normal, as were C-reactive protein, thyroid function, calcium, phosphate and magnesium levels. A routine chest x-ray was normal in appearance.

A barium swallow showed persistent narrowing of the upper oesophagus at C6/C7 with no obstruction to liquid passage or regurgitation (Images 1 & 2). The rest of the oesophagus, stomach and proximal bowel loops were normal. A contrast enhanced Computer Tomography (CT) chest did not reveal any significant extrinsic obstruction to the oesophagus. An upper gastrointestinal (UGI) endoscopy performed under sedation, demonstrated a tight stricture at 15cm of oesophagus, at the level of C5/C6 (Image 3). The endoscope was unable to negotiate beyond the stricture. The background mucosa appeared normal, healthy but was fragile to scope manipulation. Hence biopsy was not attempted. The stricture was dilated with CRE balloon to 6-8mm (Images 4 & 5). Post-procedure she was able to consume regular meals consisting of liquids and soft solids comfortably. A short course of antibiotics was used following dilatation and discontinued promptly. Additionally, she was commenced on regular proton pump inhibitor, with intravenous omeprazole 40mg twice daily administered during the in-patient stay and continued as per oral 20mg twice daily for two weeks afterwards.

A repeat UGI endoscopy with dilation to 10-12mm was performed two weeks later. The patient was able to maintain a soft diet consisting of small frequent meals on the advice of the nutritionist, without any further recurrence of symptoms following the dilation. As part of cancer surveillance, myeloma screen, mammogram, ultrasound neck and abdomen, were done, which were all normal. In the subsequent weeks, her symptoms, energy levels and weight improved. On Dermatology advice, regular immunosuppressive treatment (Prednisolone and MMF) was commenced. Image 1 & 2: Barium swallow appearance showing persistent narrowing of the upper oesophagus at C6/C7

Much attention was given to optimizing her nutritional status. Following review by the Clinical Nutritionist, whey protein and essential micronutrients were added to her diet. The patient was advised to consume a soft diet consisting of regular, small, frequent meals. Additionally, she was counselled and further educated on EBA and its management. Given the high risk of perforation, following improvement of her symptoms, further dilation was deemed of low priority at present. If symptoms recurred, repeat dilatation up to 15 mm was planned. A blood picture, iron studies and colonoscopy were planned with regular follow up in Gastroenterology outpatient clinic in one month. On follow up visits, the patient had maintained a soft diet and had gained weight (up to 7kg in six weeks). She denied having any dysphagia or odynophagia. Her skin lesions had improved as well, since the commencement of regular immunosuppressive treatments.

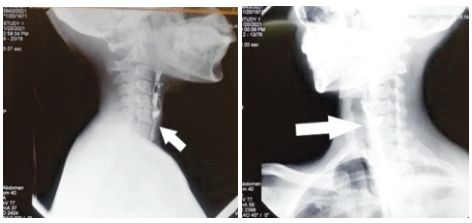

Image 1 & 2: Barium swallow appearance showing persistent narrowing of the upper oesophagus at C6/C7

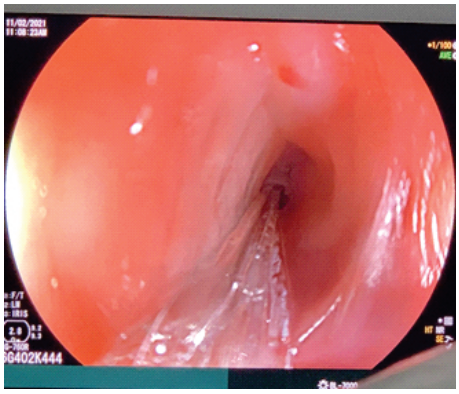

Image 3: Oesophageal stricture with CRE balloon

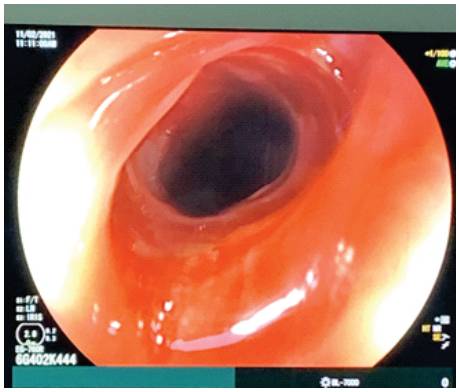

Image 4: Post dilation of stricture

Discussion

EBA is a rare, relapsing-remitting, acquired, autoimmune blistering skin disorder. It is a subepidermal bullous disorder with autoimmunity to type VII collagen, which is a major component of the anchoring fibrils of the dermal-epidermal junction 1. It is usually seen between the fourth and fifth decades of life 1. Tense vesicles and bullae are usually localized to the extensor surfaces of hands, feet, knees and elbows – areas which are most at risk of trauma. When heal, these lesions result in significant scarring and millia. Rarely, there is mucosal involvement in the mouth, larynx, oesophagus resulting in laryngeal stenosis and oesophageal strictures 1, 2. Moreover it is associated with other autoimmune conditions including inflammatory bowel disease, systemic lupus erythematosis, myeloma, rheumatoid arthritis and thyroiditis 1. EBA has known associations with malignancies such as lung, haematological and bowel cancers 2. In this patient, as the overlying mucosa appeared healthy and was not ulcerated, biopsies were not taken. Absence of histological exclusion is therefore a limitation of this case report. Furthermore, endoscopic ultrasound (EUS) facilities are not available at our centre to perform EUS fine needle aspiration for acquiring biopsies. Hence it too is a further limitation in this case report. Diagnosis is by demonstrating the presence of subepidermal blistering with presence of circulating IgG auto antibodies targeting the basement membrane on immunofluorescence 1,3.

Since there is no definitive treatment, the main aim of EBA management is to protect the skin, by minimizing blister formation and preventing complications. Involvement of mucosal membranes is associated with higher morbidity with life threatening complications 2, 4. Treatment options for EBA include use of topical or systemic steroids, steroid-sparing immunosuppressive agents, immunomodulators and biological agents 1,2. Thorough education of patients on the potential side effects of such therapies ought to be done prior to initiating therapy. Those with reflux disease should be optimally treated with anti-reflux therapy to prevent complications such as laryngeal injury 5. Promising results have been noted with the use of intravenous immunoglobulins and biologics in patients remaining refractory to immunosuppressive treatments 6, 7.

In EBA with mucosal involvement, there is repeated trauma at the high end of oesophagus as a result of solid, poorly chewed food items. Hence it causes repeated inflammation and fibrosis at the upper end of the oesophagus resulting in narrowing of oesophageal lumen. Endoscopic balloon dilation is a safe and effective method to use in those with an oesophageal stricture 8, 9. However it carries the risk of bullae formation, bleeding and cervical oesophageal perforation 9. Hence great deal of care must be taken when balloon dilation is attempted. Additionally, patients must be investigated for nutritional deficiencies that could occur due to poor oral intake as a result of dysphagia. Consequently, patients should be adequately educated on diet, lifestyle modification and use of anti-reflux treatment. Owing to the high risk of malignancies associated with EBA, early and regular cancer surveillance must be instituted.

Conclusion

EBA is a chronic relapsing-remitting autoimmune blistering skin disorder with potential mucosal involvement. If there is mucosal involvement, these patients are at high risk of life-threatening complications. Vigilance for malignancies must be maintained at all times. If there is mucosal involvement, these patients should be treated with long term immunosuppression. Here we described a patient with EBA, and an associated rare, benign upper oesophageal stricture managed with dilatation followed by immunosuppression.

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An unusual presentation of Hepatocellular carcinoma: a case report

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Abstract:
Hepatocellular carcinoma (HCC) is a leading cause of cancer mortality and morbidity in the world. Patients are largely asymptomatic at the early stages of HCC. It can be sometimes difficult to differentiate from liver abscess by imaging when in early stages of the disease. Hence it can result in delayed diagnosis of HCC, thereby leading to poor prognosis. Here we present a case where a focal lesion in the liver was misdiagnosed initially as an abscess, which failed to reduce in size despite adequate antibiotics and was later diagnosed as HCC on biopsy with corresponding high alpha feto protein.

Keywords: Hepatocellular carcinoma, Abscess, Case report

Introduction

Hepatocellular carcinoma (HCC) is a leading cause of cancer mortality and morbidity1. With rising prevalence of diabetes and obesity, non-alcoholic steatohepatitis accounts for most cases of Cirrhosis, thus inevitably to most cases of HCC2. Patients are largely asymptomatic at the early stages of HCC and there are no tell-tale signs specific for HCC as well3. Unfortunately, it is not uncommon for a fraction of patients to be treated initially as a liver abscess prior to diagnosing HCC thereby leading to poor prognosis.

Case presentation

A 63-year-old, Sri Lankan female previously diagnosed with type 2 diabetes mellitus, hypertension, hypothyroidism, dyslipidaemia, presented with a tender neck lump over 1 month. She complained of low-grade fever in the preceding month along with loss in appetite and increased fatigue. On examination she appeared well and was afebrile. There was no pallor or ictera present. There was a tender enlarged mass of 4x5cm at the right submandibular triangle. There was no palpable goitre or lymphadenopathy noted. The cardiovascular, chest and abdomen system examinations were normal. Her routine blood investigations revealed raised white blood cell (WBC) at 13x 109/L, with haemoglobin of 11.7 g/dL and platelet count of 289 x 109/L. Erythrocyte sediment rate 60mm/hr and C-reactive protein was 80. Her Liver, Renal functions including serum electrolytes were normal. An ultrasound (US) of the neck revealed an enlarged submandibular gland with abscess formation and local lymphadenopathy. A routine US abdomen was done, which revealed the presence of a focal lesion at segment 3 (2.7 cm by 3cm), with possible early abscess formation as seen in image 1. Blood cultures including tuberculosis cultures and fungal cultures were negative. A routine chest x-ray was normal in appearance. Antibodies for Melioidosis was negative. Hepatitis screen, retroviral studies and VDRL were negative.

Following the above findings, she was treated with broad spectrum intravenous antibiotics, for a total of 18days. Incision and drainage revealed presence of chronic sialoadenitis. With antibiotics, her inflammatory markers improved, and the neck lump soon disappeared. However, the patient continued to complain of loss of appetite and fatigue. A month later, her repeat liver functions revealed AST 26 U/L, ALT 18 U/L, ALP 137 U/L, GGT 225 U/L, total bilirubin 6.3mg/dl. A repeat US abdomen showed the continued presence of focal lesions in both segment 2 and 3, with no change in size compared to previous. Therefore, a contrast enhanced computer tomography (CECT) abdomen was done. It revealed a normal liver size with multiple low attenuating focal lesions involving both lobes of the liver, with largest lesion in segment 7 measuring 5.8cm by 5cm by 5.6cm with a thick wall with smaller lesions noted in segments 2 and 6 of the liver (Image 2). There was no thrombosis within portal vein or inferior vena cava. Additionally, there was no free fluid noted. Repeat blood, urine cultures as well as Melioidosis antibodies were all negative. An upper gastrointestinal endoscopy and colonoscopy were done, both of which were normal examinations. The patient underwent a liver biopsy of the largest lesion which revealed metastatic deposits of adeno carcinoma with immunohistochemistry positive for HepPar 1, indicating presence of hepatocellular carcinoma. An alpha feto protein (AFP) was raised at 669. Hence hepatocellular carcinoma was diagnosed. The patient was counselled and educated on HCC. A multidisciplinary team discussion was conducted involving hepatobiliary surgeons, interventional radiologists, pathologists and hepatologists. Given the patient's current state with good performance status, it was deemed that she was best served with transcatheter arterial chemoembolization (TACE). She underwent TACE procedure with no post procedure complications.

Discussion

HCC is an aggressive tumour which can directly invade portal and hepatic veins. In early stages patients may appear asymptomatic and are unlikely to have distant metastases. Hence when identified, depending on the patient's performance status, underlying liver status and co-morbidities, appropriate curative treatments can be initiated early.

This case highlights the difficulty in diagnosing HCC in early stages, when focal lesions in liver can be misdiagnosed, which in turn delays the initiation of appropriate management. Whilst the patient presented to us with a submandibular mass with low grade fever, which later turned out to be chronic sialoadenitis, it was on further evaluation of this presentation that we came across the focal lesions in the liver on imaging. Owing to the clinical presentation, the focal lesions were initially suspected to be multiple liver abscesses. However, as there was no reduction in size in subsequent scans despite having received adequate antibiotic usage, further evaluation was warranted. Liver biopsy is usually reserved for those with atypical presentation. In this case a liver biopsy ultimately yielded the final diagnosis of HCC. Therefore, the diagnosis of HCC was delayed owing to the atypical appearance in initial US and CT imaging.
The clinical picture of low-grade fever with submandibular lump further misdirected the clinicians towards a possible diagnosis of liver abscess. Typically, liver abscesses appear as well circumscribed ring lesions on US abdomen but they can appear as heterogenous mass like lesions which are indistinguishable from HCC. Likewise, HCC can have variable appearances, especially on US abdomen depending on lesion size and echogenicity of background liver. Hence while the sensitivity of US in diagnosing liver abscesses ranges from 66-90% 4,5 there are studies reporting high false positive rates (up to 55%) for US in detecting liver abscesses as well 6.

Therefore, this case further highlights the importance of adequately evaluating focal liver lesions. If in doubt of the characteristics of the focal liver lesion, it is important to consider other alternatives. In such situations, a raised alpha feto protein level could help direct the clinicians towards a diagnosis of HCC as when raised its specificity is close to 100% for HCC 3. Furthermore, it is important to appreciate that HCC tumour volume doubling time of small HCC (< 5cm) is usually around 110-200 days, which warrants for repeat evaluation and follow up of focal lesions via imaging modalities 7.

HCC is usually rare in patients with non-cirrhotic livers and when present may have atypical presentations. Typically, HCC presents as a large solitary or dominant mass with peripheral satellite lesions in a non-cirrhotic liver 9. Our patient had presence of multiple focal lesions, largest of which was in segment 7. However, despite initially not having clinically significant or biochemical markers of cirrhosis as well as a FIB-4 score of 1.34, few weeks later she presented with episodes of decompensation with ascites. Given her long history of type 2 diabetes mellitus, it is likely that she had underlying non-alcoholic steatohepatitis, which contributed to development of HCC and cirrhosis. Once diagnosis was established, surgical resection was not possible and TACE was considered as a potential treatment to slow the progression of the disease.

Conclusion

HCC must be suspected in patients presenting with focal liver lesions. It commonly occurs in the background of cirrhosis. Early involvement and discussion within a multidisciplinary team must be instituted.

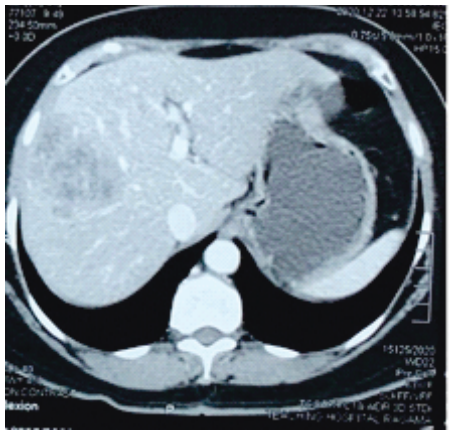

Image 1: Moderate sized mixed echogenic focal lesion noted in segment 3 as seen on US abdomen

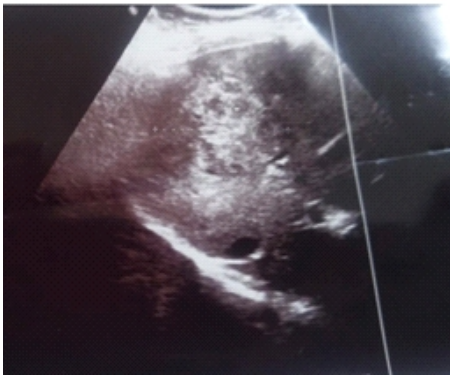

Image 2: Largest lesion in Segment 7 with thick wall with heterogenous contrast enhancement as noted on CECT abdomen (arterial phase)

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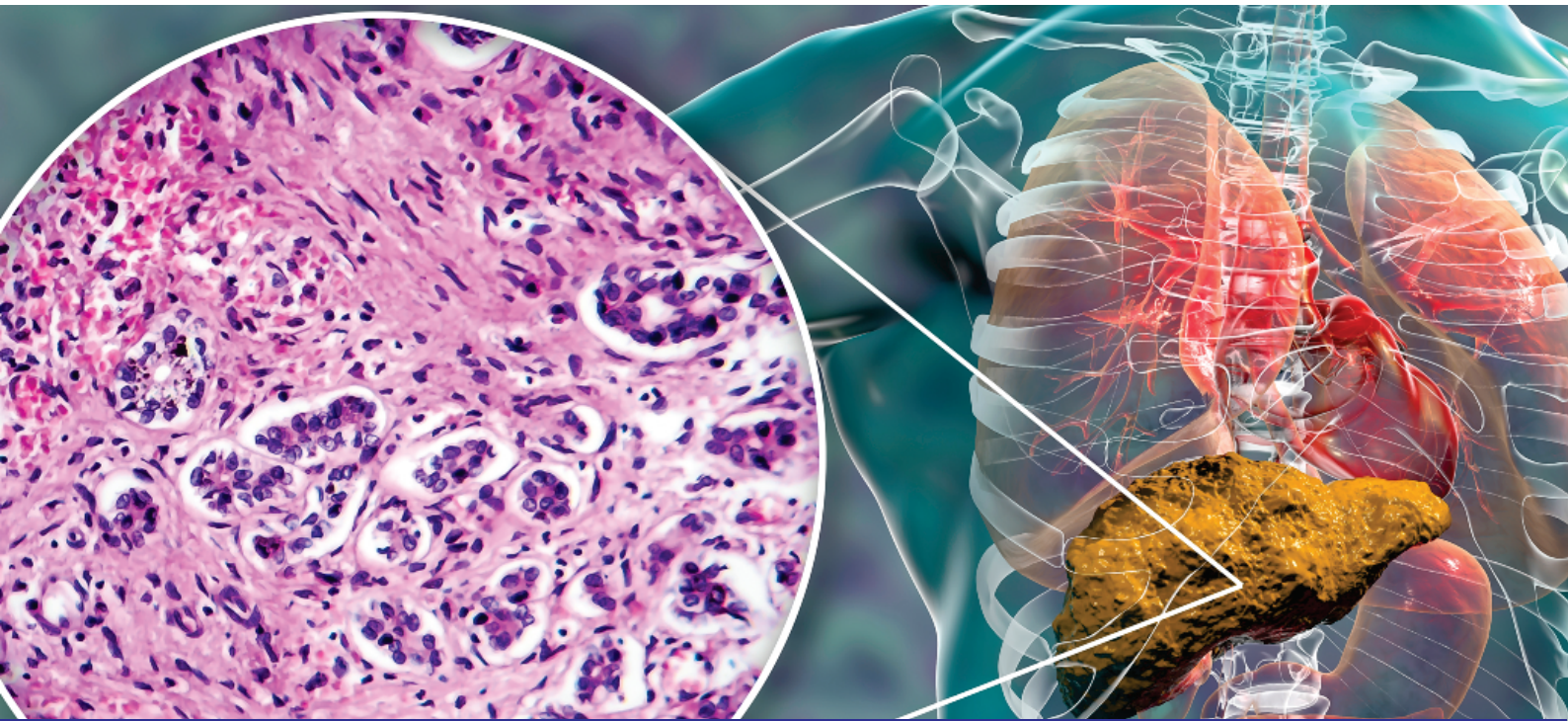

A Case of Primary Hyperparathyroidism presented as Recurrent Acute on Chronic Pancreatitis

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Ranjith Peiris, Hishali Jayasundara, Tilan Aponso

Abstract
Primary Hyperparathyroidism (PHPT) is an uncommon cause for Acute Pancreatitis (AP) or Chronic Pancreatitis (CP). Although the exact etiology for the development of CP in PHPT remains controversial, it is thought to be due to the hypercalcemia which occurs in PHPT. Other than the CP there can be several other organ involvements in PHPT such as renal calculi, gallstones, bone disease and psychiatry disorders.

Here we present a case of a lady who presented with recurrent episodes of acute on chronic pancreatitis. On etiological investigations she was found to have hypercalcemic and on further evaluation there was elevated serum Parathyroid Hormone (PTH) levels. Contrast CT scan was suggestive of a left inferior parathyroid adenoma and following successful surgical resection her PTH and serum calcium levels were normalized and reduction of AP episodes were also observed.

Although the association of CP and PHPT is rare, proper etiological assessment would be beneficial for the prompt diagnosis and timely management of the disease.

Keywords: Acute Pancreatitis, Chronic Pancreatitis, Primary Hyperparathyroidism, Parathyroid Adenoma

Introduction

Chronic Pancreatitis is a progressive inflammation in the pancreas which has a worldwide prevalence of 2-14/ 100000 population. Among all the etiological factors which contribute for the development of CP, Alcohol is dominating. PHPT is a rare cause for pancreatitis which can be manifested as Severe acute, Recurrent acute or Chronic pancreatitis. CP can present with pain syndrome, exocrine failure and/or endocrine failure. In PHPT, in addition to CP several other organs can be affected such as kidneys and bones. Cause for the development of CP in PHPT is thought to be elevated serum calcium levels which will act in several ways for the development of pancreatitis. Main principals in the management of such condition are prompt management of acute episodes of pancreatitis along with chronic pancreatitis, management of hypercalcemia and surgical resection of secreting tumor.

Case report

Here we present a case of a 44-year-old female patient from Angoda, with a history of recurrent episodes of epigastric pain for two years duration presented with a similar pain which radiate to the back for 2 weeks. She had suffered from few attacks of acute pancreatitis with elevated serum amylase level from time to time during that period where she got admitted to the hospital and managed conservatively. At the same time, she was diagnosed with diabetes mellitus which was poorly responded to oral hypoglycemic agents and eventually changed in to insulin with a fair control. During the same period, she was investigated for altered bowel habits with predominant constipation where she underwent colonoscopy which was a normal study. Apart from that she has got no significant family history of note.

Abdominal examination revealed mild epigastric tenderness with no palpable lumps.

On investigations her serum amylase level found to be high which was 198 U/l. Ultrasound of the abdomen showed evidence of chronic pancreatitis and mild main pancreatic duct dilatation with no features of pseudocyst formation. Abdominal CT scan was performed which was suggestive of chronic pancreatitis with pancreatic duct dilatation at the body with multiple ductal strictures. There were no calcifications (Fig:1). These findings were confirmed by the subsequent MRCP (Fig:2). She underwent an ERCP, during which the pancreatogram also showed irregular tortuous main pancreatic duct with several strictures and a pancreatic stent has been inserted (Fig:3).

Given the history of recurrent acute on chronic pancreatitis and the significant constipation, serum calcium level has been performed which was elevated up to 15.2 mg/dl. So, on further evaluation her serum parathyroid hormone level was found to be elevated to 454.5 pg/ml with a low total vitamin D level 5.88 ng/ml. DEXA scan was performed to evaluate the bone involvement which showed a discrepancy between proximal and distal bone mineral densities with a T score of -2.3 and -2.8 in proximal femur and a T score of -5.7 and -5.9 in distal forearm which was suggestive of hyperparathyroidism (Fig:4). Her very high serum calcium level was managed with intense hydration and intravenous Zolindronic acid. With that she achieved normocalcemia. Ultrasound scan of the neck showed evidence of solid hyperechoic nodule with internal vascularity in the lower pole of the thyroid suggestive of a parathyroid adenoma. Fine needle aspiration cytology of the lesion revealed cellular smears with cells representing parathyroid origin with hyperplastic/ neoplastic changes. CECT scan of the neck revealed an exophytic left lower thyroid nodule suggestive of a parathyroid adenoma (Fig:5). With the normalization of the serum calcium level with medical management she underwent a surgery for left inferior parathyroidectomy. During the surgery intraoperative parathyroid hormone assay was performed, in which preoperative PTH value of 441.9pg/ml dropped down to 27.8pg/ml post operatively. Histology confirmed parathyroid adenoma and eventually her serum calcium level normalized and reduced frequency of abdominal pain.

Fig :1

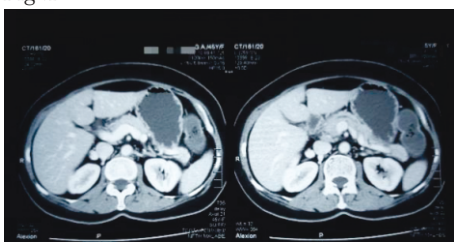


Fig :2

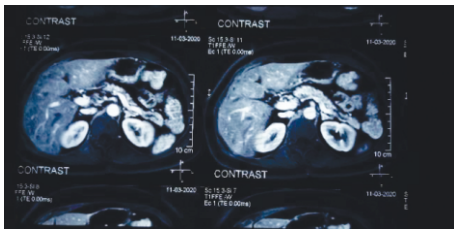


Fig :3

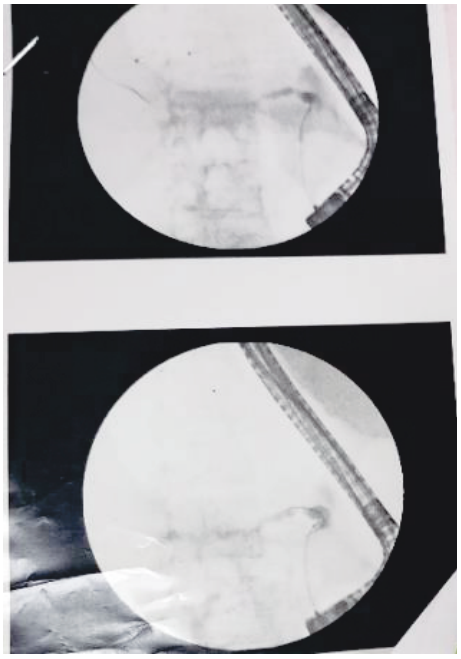


Fig :4

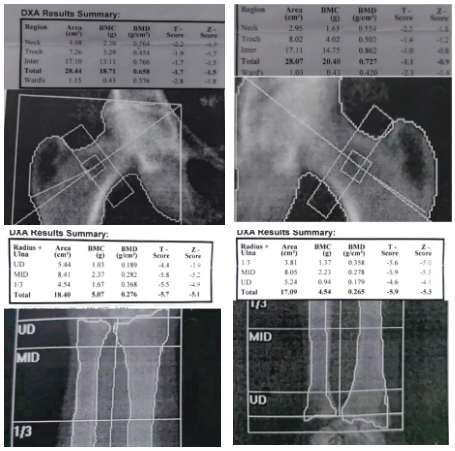
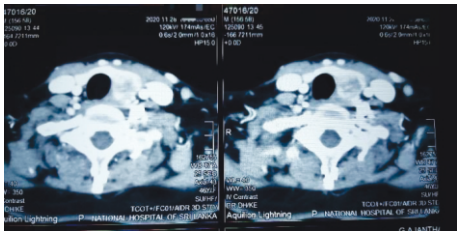


Fig :5



Discussion

Chronic Pancreatitis (CP) is defined as a pathologic fibro-inflammatory syndrome of the pancreas, which occurs mainly in individuals with genetic, environmental and/or other risk factors and among them who develop persistent pathologic responses to parenchymal injury or stress. (1)

The overall incidence of CP ranges from 2-14/100,000 population and shows some variability according to the study design and country with a male predominance (2). Among the various causes for the development of CP, alcohol is the dominating factor which accounts for approximately one-half of all cases (3). Most of the patients have consumed more than 150 g of alcohol per day for more than 10 years (4). TIGAR-O classification classify by risk modifiers causing pancreatic disease as toxic-metabolic, idiopathic, genetic, autoimmune, recurrent and severe AP-associated CP, and obstructive (5).

Primary Hyperparathyroidism accounts for about 0.1-4% of all CP cases (5). Some countries show less prevalence of association which may be due to the occurrence of pancreatitis in the advanced stage of hyperparathyroidism (6). In most of the areas this association is predominant in young males where as some other areas show otherwise (7).

The cause for the occurrence of CP in the setting of hyperparathyroidism remains controversial but there is a significant association of hypercalcemia with CP (8). Significantly high serum calcium levels were observed in patients with CP due to hyperparathyroidism compared to other etiologies. Hypercalcemia affects through several mechanisms for the development of pancreatitis although the exact mechanism remains controversial. During hypercalcemia there is increased intracellular calcium concentration within the acinar cells of the pancreas, which is a vital intracellular second messenger for the release of pancreatic enzymes (9). Thus, the high levels of intracellular calcium cause damage to the pancreatic tissues by unopposed activation of pancreatic enzymes particularly trypsinogen (10). Other than that deposition of calcium and formation of protein plugs which are responsible for upstream pancreatitis and direct toxic effect of PTH on pancreas also play a role for the development of pancreatitis (11).

Jacob et al. reports four types of associations of PHPT and pancreatitis, which are PHPT revealed by AP, PHPT revealed by recurrent AP without CP, PHPT revealed by a CP with or without pancreatic calcifications, or PHPT complicated by AP in the postoperative period (7). One meta-analysis concluded that there is 15% chance of developing CP in PHPT (6).

There are other organs involvement reported in CP due to PHPT such as Nephrolithiasis, Bone diseases, Psychiatry disorders and Gallstone disease (12), which we couldn't observe in our patient although she was extensively investigated.

Regardless of the PHPT, the episode of acute pancreatitis must be treated initially with supportive care before any intervention for PHPT (13) so as the hypercalcemia with adequate hydration and bisphosphonates as in this patient, because the results can be detrimental. After the initial stabilization patients should be subjected to surgery for the removal of secreting lesion and the surgery will result in intraoperative drop in PTH concentration (14) as we observed in this patient. Some studies showed that 42%-100% resolution of pancreatitis episodes following surgery even the pain in CP (12). In our patient there was reduction of pain episodes and normalization of PTH levels along with serum calcium was observed following surgery.

Conclusion

Primary hyperparathyroidism is a rare cause for the development of CP. Diagnosis of such association is also challenging due to non-specific symptoms. After diagnosing the condition, we have to treat promptly for episodes of acute pancreatitis and hypercalcemia followed by surgical resection of the PTH secreting tumor which will cause the improvement of symptoms.

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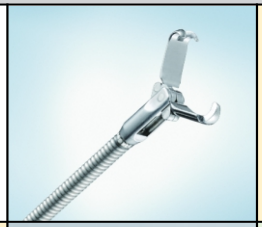
GI ENDOSCOPY TOOLS AND ACCESSORIES

A VISUAL CATALOGUE FOR GASTROENTEROLOGISTS, NURSES, & TECHNICIANS

BIOPSY FORCEPS
To obtain biopsy specimens

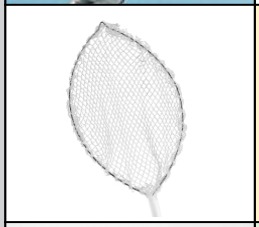
Diameter: Giant Jumbo 5.5mm, Jumbo 3.2mm, Regular 2.3mm, Pediatric 1.8mm



RAT TOOTH GRASPING FORCEP
For retrieval of foreign objects

ALLIGATOR GRASPING FORCEP
For retrieval of foreign objects; with wider grasp

3-PRONG GRASPING FORCEP
For retrieval of large polyp or foreign objects



RETRIEVAL NET
For retrieval of large polyp or foreign objects

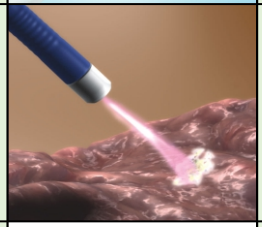
HEMOCLIP
For mechanical hemostasis, closure of mucosal defects, marking or anchoring

INJECTION NEEDLE
For sclerotherapy for hemostasis; also used for submucosal injection to lift lesion prior to EMR/ESD

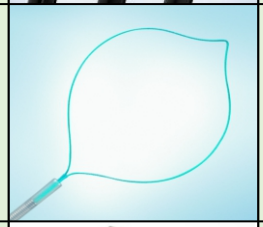
HEMOSTATIC FORCEPS
Targeted coagulation for hemostasis at the precise site of bleeding

HEATER PROBE
For thermal coagulation via heated tip and pressure application

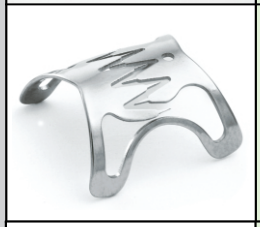
BIPOLAR COAGULATION PROBE
Bipolar spiral tip provides coagulation at any angle



ARGON PLASMA COAGULATION
Monopolar, noncontact probe which emits ionized argon gas for hemostasis



SINGLE-USE LIGATION DEVICE
For polyp ligation prior to polypectomy; prevents post-polypectomy bleeding

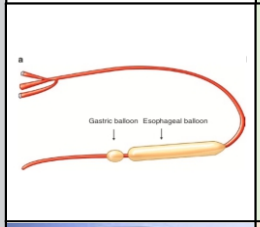


OVER-THE-SCOPE CLIP
Clipping device that provides a strong tissue grasp and compression

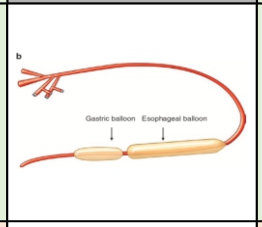
HEMOSTATIC SPRAY
Inorganic powder administered endoscopically which promotes hemostasis

BAND LIGATOR
For endoscopic ligation of esophageal varices and anorectal hemorrhoids

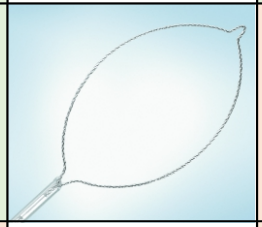
SUTURING DEVICE
For correction of defects such as fistula, GI bleeding, perforation or tears using sutures



SENGSTAKEN BLAKEMORE TUBE
Device used for tamponade of bleeding esophageal varices (3 ports)



MINNESOTA TUBE
Device used for tamponade of bleeding esophageal varices (4 ports to allow aspiration of both gastric and esophageal contents)



POLYPECTOMY SNARE
For polyp removal via cold or hot resection

ESD KNIFE
For lateral incision and submucosal dissection of large lesions

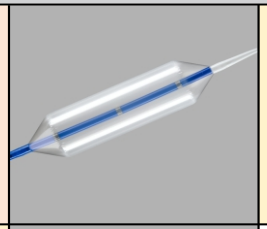
DISTAL CAP
To improve visualization and maintain constant distance while resecting large surface areas

MUCOSAL TATTOO
For marking and identifying lesions along the GI tract

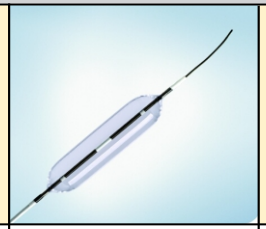
LIFTING AGENT
Injected in the submucosal layer to lift lesions and create a plane for dissection prior to EMR/ESD

POLYP TRAP
Tissue container for easier colonoscopy specimen retrieval

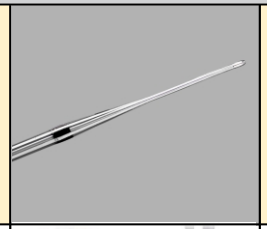
ENDOCUFF
Device attached to distal end of colonoscope designed to maintain and maximize viewable mucosa



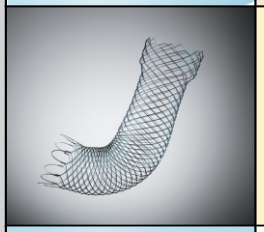
ACHALASIA BALLOON
For dilation of esophageal strictures; specifically indicated for patients with achalasia



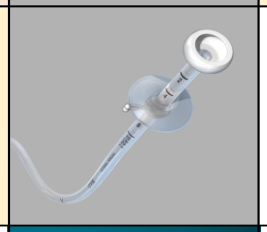
ESOPHAGEAL BALLOON DILATOR
For dilation of esophageal strictures



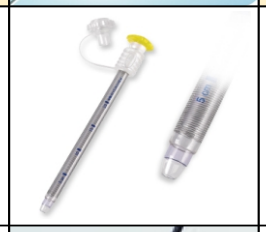
SAVARY GILLIARD DILATOR
For dilation of esophageal strictures



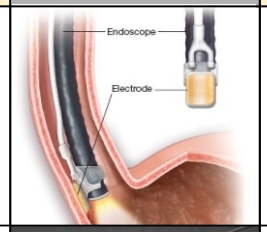
ESOPHAGEAL/ DUODENAL/ COLONIC STENT
To expand and open obstructed sections of the GI tract; often used for palliative treatment



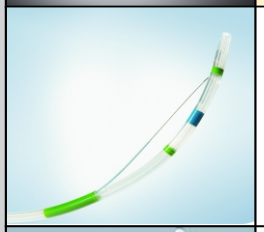
PEG TUBE
For percutaneous endoscopic placement to provide enteral nutrition for patients requiring nutritional support



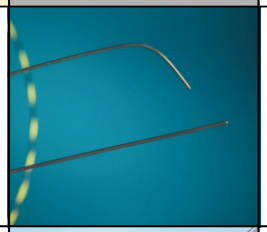
OVERTUBE
Used to prevent aspiration or mucosal laceration in upper GI endoscopy



RADIOFREQUENCY ABLATION DEVICE
Attached to the end of an endoscope which delivers radiofrequency energy or heat to cause tissue death



SPHINCTEROTOME
For cutting the papilla and to assist in cannulation



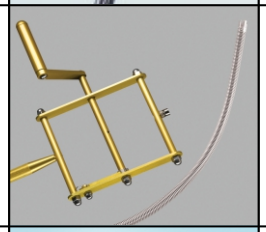
GUIDEWIRE
Facilitates access to both biliary and pancreatic ducts, maintain cannulation, and allow passage of instruments in ERCP

STONE EXTRACTION BALLOON
Facilitates stone extraction via sweeping of the biliary tract with an inflated balloon

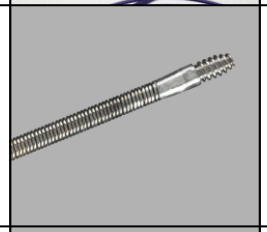
BILIARY BALLOON DILATOR
For biliary dilatation and sphincteroplasty

STONE EXTRACTION BASKET
For retrieval of stones in the bile duct or pancreatic duct

MECHANICAL LITHOTRIPTOR
For mechanical crushing of stones within the duct

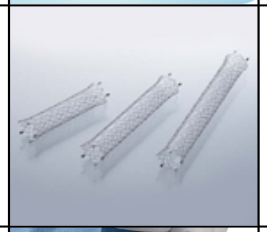


SOEHENDRA LITHOTRIPTOR
For mechanical crushing of stones in the bile duct when other methods of endoscopic removal have failed

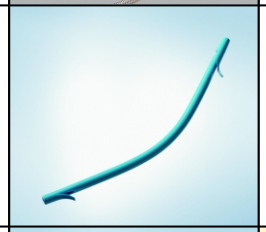


SOEHENDRA STENT RETRIEVER
Used for endoscopic removal of stents from the biliary and pancreatic ducts

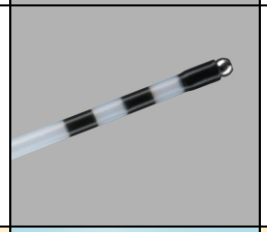
CYTOLOGY BRUSHES
For collection of biliary specimen and detection of malignant neoplasms within the duct



BILIARY METALLIC STENTS
Flexible metallic tube to keep the bile duct open, which has been blocked or partially blocked



BILIARY AND PANCREATIC PLASTIC STENTS
Flexible plastic tube to keep the bile duct open, which has been blocked or partially blocked

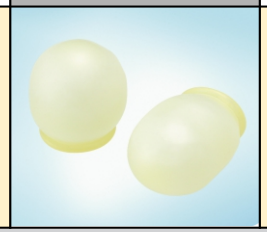


METAL BALL TIP CANNULA
Used to aid in difficult cannulation of the ductal system

CANNULA FOR ENTEROSCOPY-ASSISTED ERCP
Used for cannulating the papilla in enteroscopy-assisted ERCP

CHOLANGIO-SCOPE
Enables direct visualization of the pancreatic and bile duct

EUS FNB DEVICE
Device used for EUS-guided fine needle biopsy of suspicious lesions



EUS BALLOON
Used to provide clearer views in EUS procedures

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Concomitant Guillain–Barré Syndrome in a Young Sri Lankan Male with Severe Ulcerative Colitis: a Case Report

J. M.H. D. Jayasundara 1 (MBBS MD Senior Registrar in Gastroenterology) corresponding author,

Vajira Tharanga Samarawickrama 1, Ranjith Peiris 1, Tilan Aponso 2, Danushi Abeynayake 2

Abstract

Background

Guillain–Barré Syndrome is an immune mediated polyneuropathy. Ulcerative Colitis is an immune mediated chronic inflammatory condition mainly of the large intestine. Guillain–Barré Syndrome can present as a rare extraintestinal manifestation of Ulcerative Colitis when in remission or in a relapse. However, the concomitant presentation of Guillain-Barré Syndrome during a relapse of Ulcerative Colitis is very rare and only a few cases are reported to date.

Case presentation

A 24 year old young male diagnosed of Ulcerative Colitis presented with bloody diarrhea of frequency more than six times a day. He had been in clinical remission even after defaulting treatment for more than a year. He had also noted difficulty in walking prior to admission to the hospital. He was managed as for a severe relapse of Ulcerative Colitis and Guillain–Barré Syndrome. Appropriate management of both the illnesses helped him to recover.

Conclusion

Immune mediated diseases can have rare coexisting presentations. We report a case of Ulcerative Colitis with concomitant Guillain–Barré Syndrome. It is essential to be open minded and timely, appropriate treatment led to successful management of both the illnesses.

Keywords: Ulcerative Colitis, Guillain– Barré Syndrome, Acute Motor Axonal Neuropathy, Inflammatory Bowel Disease, case report

Background

Ulcerative colitis is a chronic inflammatory disease of the colon with relapses and remissions. Its incidence and prevalence are increasing globally including Asia [1]. Neurologic complications of inflammatory bowel disease are not very common [2]. Guillain–Barré Syndrome is considered as one of the rare extraintestinal manifestations of inflammatory bowel disease [3]. Guillain–Barré Syndrome is an immune mediated, monophasic acute paralyzing illness usually provoked by a preceding infection. Out of the different variants of Guillain– Barré Syndrome, Acute Motor Axonal Neuropathy (AMAN) is a primary axonal form of Guillain–Barré Syndrome. It is thought that genetic susceptibility, aberrant self-recognition and immunopathogenic autoantibodies against organ-specific cellular antigens shared by the colon and extra-colonic organs play a role in contributing to the pathogenesis and development of the extra intestinal manifestations [4].The first documentation is in 1985 by Zimmerman where Guillain–Barré Syndrome had occurred in patients who were in remission [5]. To date, only about 9 cases are reported according to the best of our knowledge. We present a young patient who had not received any TNF alpha therapy previously presenting with Acute Motor Axonal Neuropathy (AMAN) variant of Guillain–Barré Syndrome along with a relapse of Ulcerative Colitis, a combination which has not been reported in literature.

Case presentation

Our patient is a young, 24 year old male, diagnosed with extensive colitis of Ulcerative Colitis in mid- 2020. He had defaulted treatment after three months following the diagnosis. However, he had been in clinical remission for almost a year. He was initially treated with oral prednisolone, sulfasalazine and azathioprine. This time he presented with clinical features suggestive of a relapse of severe ulcerative colitis. There was no evidence of toxic megacolon or other detrimental complications. Cessation of smoking was identified as a potential precipitating factor in addition to the poor compliance. He did not have any concomitant extra intestinal manifestations. There had been a marked unintentional weight loss of almost twenty kilograms within a month. One week prior to admission he had noted difficulty in walking but was able to mobilize with support.

He had marked bilateral lower limb edema with general unwellness. He was afebrile and did not have tachycardia, hypotension or a tender abdomen. Lower limb examination revealed bilateral symmetrical proximal more than distal weakness with diminished reflexes and preserved sensation without sphincter involvement. There was no significant muscle wasting or fasciculations. Also, he had symmetrical proximal more than distal upper limb weakness with diminished reflexes and all sensory modalities were intact. His cranial nerves, higher functions and cerebellar examinations were normal. He had a weak neck muscle power and cough effort but was not in respiratory distress. He maintained his vital parameters with no desaturation or fluctuation of blood pressure or pulse rate.

Full blood count revealed a moderate hypochromic microcytic anemia. His inflammatory markers were elevated with very low albumin. His renal functions were normal and urine full report did not reveal albuminuria. He had a mild hypokalemia and thyroid functions were normal. Fecal calprotectin was positive. Stool cultures excluded other enteric infections and Clostridium difficile toxins were not detected. Flexible sigmoidoscopy revealed severe mucosal inflammation (Mayo score of 3 on endoscopic appearance) and histology excluded concomitant CMV colitis and confirmed Ulcerative Colitis flare. Nerve conduction study revealed Acute Motor Axonal Neuropathy type of Guillain–Barré Syndrome. CSF analysis confirmed protein cell dissociation with absent cells and protein of 62mg/dl. Covid 19 infection was safely excluded as well as other viral aetiologies as CMV, EBV, HIV.

The relapse of Ulcerative Colitis was managed with intravenous hydrocortisone of 100 mg every 6 hourly, subcutaneous enoxaparin as for DVT prophylaxis, intravenous fluids and albumin. He showed a significant clinical improvement by day 3

along with a rapid decline of CRP being less than 45. Subsequently, he was managed with oral prednisolone and later on with sulfasalazine 1 g bd and azathioprine 50 mg daily. Since he had many poor prognostic factors such as young age of onset, extensive colitis requiring hospitalization, low albumin, high CRP, Tofacitinib 10mg twice a day was commenced as he was not a suitable candidate for TNF alpha blockers due to concomitant GBS.

Guillain–Barré Syndrome was managed with IV immunoglobulin 0.4g/kg for five days by which his lower limb proximal muscle weakness improved. His vital capacity and other vital parameters were monitored daily. He received regular chest and limb physiotherapy. By day five there was an improvement in the proximal lower limb weakness. He was followed up at the clinic and Ulcerative Colitis was in remission with Tofacitinib, prednisolone tapering regime and sulfasalazine.

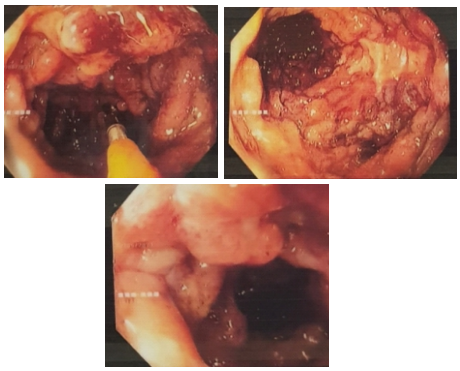

Fig 1

Motor Nerve Conduction Study

Site	Latency (ms)	Amplitude (mV)	Area (Vms)	Segment	Distance (mm)	Interval (ms)	NCV (m/s)	NCV N D
Ulnar R								
Wrist	2.37	2.86	25.54	Wrist		2.37		
Elbow	6.69	2.82	23.01	Wrist-Elbow	270	4.32	62.5	
Ulnar L								
Wrist	2.49	2.42	12.55	Wrist		2.49		
Elbow	6.81	2.31	12.41	Wrist-Elbow	270	4.32	62.5	
Peroneal R								
Ankle	2.85	900.00uV	6.78	Ankle		2.85		
Head of Fibula	10.25	880.00uV	7.92	Ankle-Head of fibula	380	7.40	51.4	
Tibial R								
Ankle	6.95	2.92	11.65	Ankle		6.95		
Peroneal L								
Ankle	2.6	820.00	5.44	Ankle		2.6		
Head of Fibula	10.05	780.00	5.33	Ankle-Head of fibula	380	7.45	51	
Tibial L								
Ankle	4.25	4.56	21.58	Ankle		4.25		

F Wave Study

Nerve	Stim.site	F-Lat	F-Lat N.D.	M Lat	F-M Lat	F Occurr			
Tibial R	Ankle	47ms		5.2ms					absent
Tibial L	Ankle	5.2ms		5.2ms					absent
Ulnar R	Wrist	2.6ms		2.6ms		0/9.0%			repeaters
Ulnar L	Wrist	2.75ms		2.7ms		0/8,0%			

Sensory Nerve Conduction Study

Site	Latency (ms)	Amplitude	Area	Segment	Distance (mm)	Interval (ms)	NCV (m/s)	NCV N D
Ulnar, R								
Wrist	1.56 ms	26.30uV	1.33uVms	Wrist		1.56ms		
Ulnar, L								
Wrist	1.79ms	31.10uV	0.68uVms	Wrist		1.79ms		
Sural, R								
Sural	2.42ms	7.40uV	0.10uVms	Sural		2.42ms		
Sural,L								
Sural	2.21ms	9.40uV	0.56uVms	Sural		2.21ms		
Radial,R								
Forearm	1.15ms	33.40uV	0.75uVms	Forearm		1.15ms		

Fig 2

Discussion and Conclusions

We present a young male who presented with a relapse of Ulcerative Colitis subsequently developing Guillain–Barré Syndrome. Also he had a significant weight loss and generalized body weakness. Timely, appropriate diagnosis and management aided in marked improvement and recovery. This case demonstrates a rare clinical presentation of coexistent Ulcerative Colitis with AMAN variant of Guillain–Barré Syndrome.

Extra intestinal manifestations occur in 5% to 50% of all patients with inflammatory bowel disease. The severity and occurrence of extra intestinal manifestations and their correlation with intestinal-inflammatory bowel disease activity vary, but most extra intestinal manifestations are directly associated with an ongoing intestinal flare [6].

The association of Ulcerative Colitis with neurologic involvement is rare and often controversial [7]. According to A. Lossos out of the IBD patients who developed neurologic manifestations; 74 % had developed after a mean of 5.7 years following development of IBD and only 10% during an IBD exacerbation. The neurologic manifestations documented were in the form of myelopathy, myopathy ,myasthenia gravis and cerebrovascular disorders[8].Peripheral neuropathies related to IBD seems to be more frequent in Ulcerative Colitis, with a reported incidence of 1,9% but it seems that it is associated with a lower rate of demyelinating forms as compared to Crohn's Disease [9].

Guillain–Barré Syndrome is one form of neurological manifestations which can occur both in remission or a relapse of Ulcerative Colitis[10].The exact pathogenesis of Ulcerative Colitis with Guillain–Barré Syndrome is unclear.It may be related to the following factors: Ulcerative Colitis-associated vasculitis, post infection immunity, malnutrition, toxic metabolites, vitamin deficiency, and thrombotic disease[11]. It is also postulated that since both Ulcerative Colitis and Guillain–Barré Syndrome are autoimmune diseases there may be similar autoimmune mechanisms in the development of both these diseases. However ,association of Guillain–Barré Syndrome and Ulcerative Colitis is extremely rare and only a few cases have been reported [12,13]. There are different variants of Guillain–Barré Syndrome of which Acute Motor Axonal Neuropathy (AMAN) is one such type. Our case was an Acute Motor Axonal Neuropathy (AMAN) form of Guillain-Barré Syndrome with a relapse of Ulcerative Colitis which has not been reported up to date so far. Most cases have antecedent infection with Campylobacter jejuni and many have antibodies directed towards GM1 ganglioside-like epitopes.The mechanism of nerve-fiber injury has not been defined yet. Acute Motor Axonal Neuropathy (AMAN) is a novel disorder caused by an antibody- and complement-mediated attack on the axolemma of motor fibers [14]. The nerve conduction study was in favor of axonal injury in our patient.

Infliximab, a Tumor Necrosis Factor (TNF) alpha blocker, is known to be an effective treatment for Ulcerative Colitis. There are many cases documented in the literature mainly by the US Food and Drug Administration where Guillain–Barré Syndrome had developed after the initiation of anti- Tumor Necrosis Factor (TNF) alpha therapy [15,16]. Interestingly, our patient had not received infliximab therapy at any time. Our next choice was Tofacitinib which is an orally administered small molecule and a Janus kinase inhibitor. Tofacitinib is one of the drugs emerging into the limelight for the management of moderate to severe Ulcerative Colitis. It is known to be cost effective and also effective in achieving endoscopic response, endoscopic remission, and mucosal healing [17]. It is important to exclude other causes of weakness in a patient presenting with diarrhea; mainly electrolyte imbalances, endocrine disorders as hypothyroidism, thyrotoxicosis and iatrogenic Cushing's syndrome. Finally, the clinical picture and the relevant investigations directed us for

appropriate and timely management of our patient. Thus, our case highlights the importance of thorough clinical examination and being keen on the rare manifestations of common illnesses.

Abbreviations

DVT: deep vein thrombosis

CSF : cerebro spinal fluid

IBD : inflammatory bowel disease

CRP : C reactive protein

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Contributions

JMHDJ VT TA DA were involved in managing the patient and gathering of data. JMHDJ and VT did the literature review and writing of the initial manuscript was done by JMHDJ. RP finalized the manuscript and gave expert opinion in management issues. All authors read and approved the final manuscript.

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Ethics declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Informed written consent for the publication of details and pictures was obtained from the patient. Consent form can be made available to the editor on request.

Competing interests

The authors declare that they have no competing interests.

HIGHLIGHTS
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Highlights of Endoscopy Assistants Training








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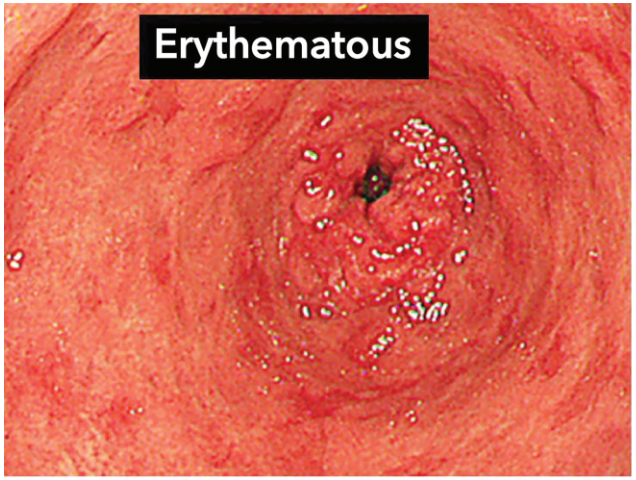

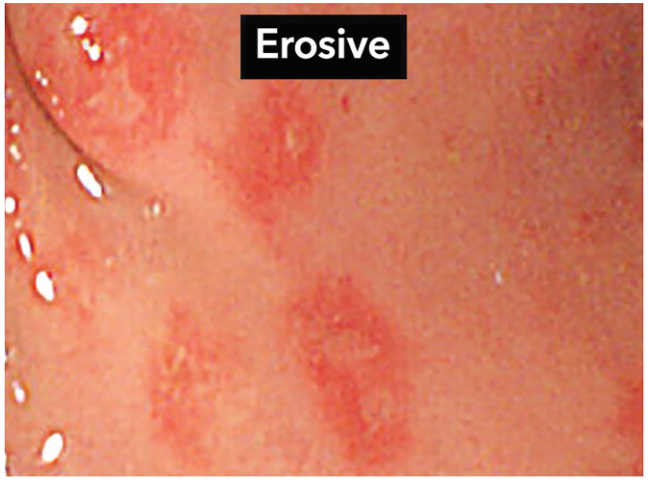

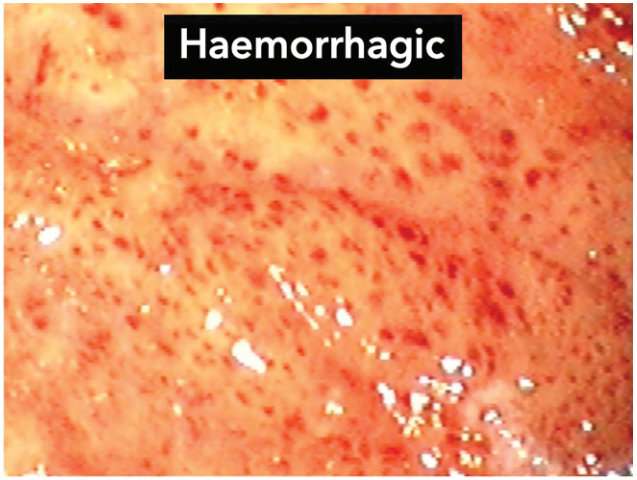

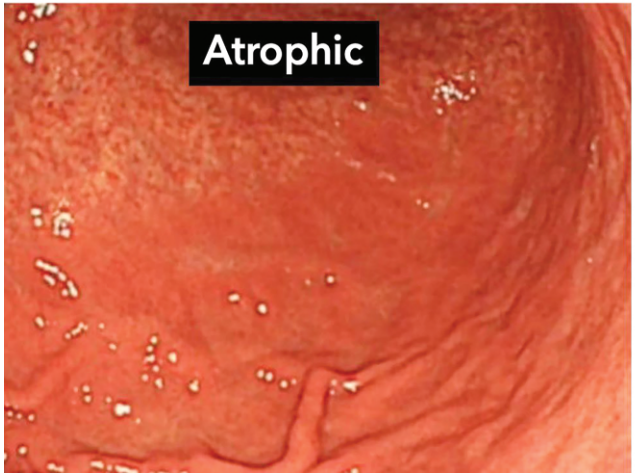

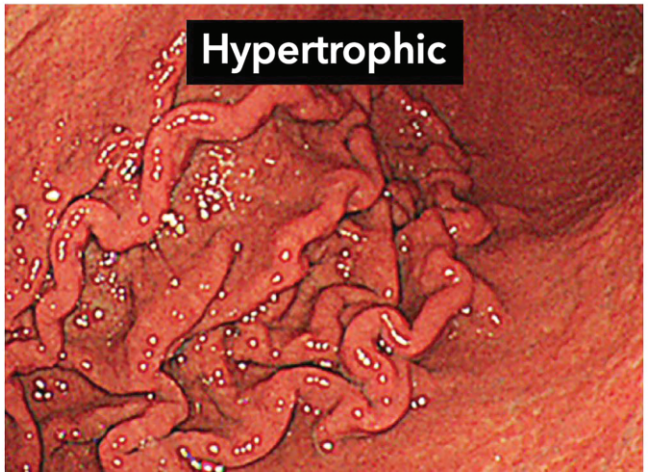

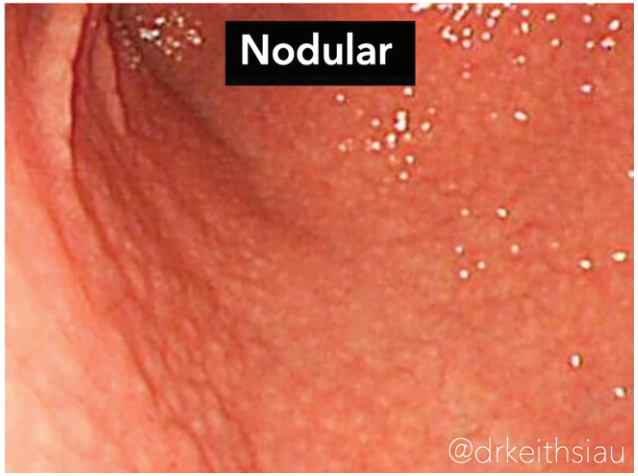

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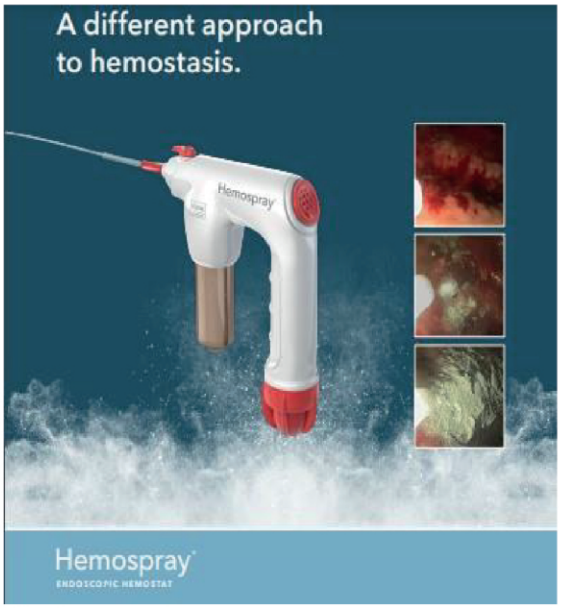

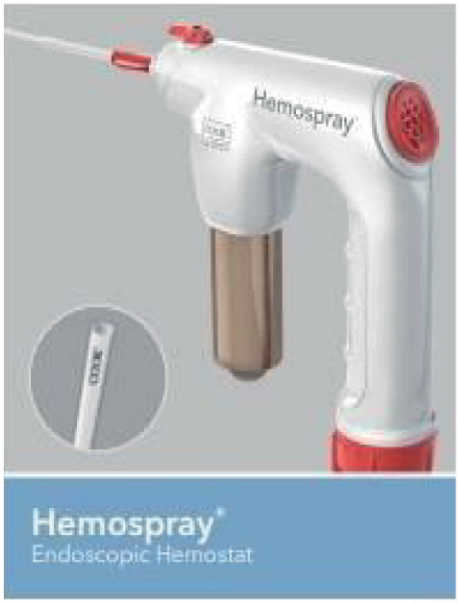

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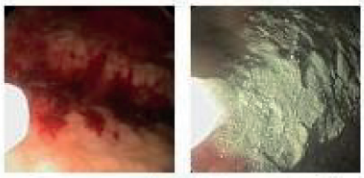

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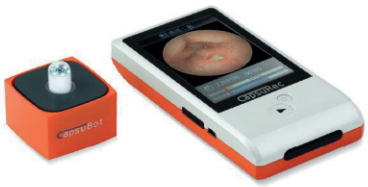

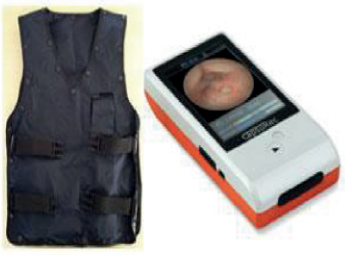

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