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Case Report

Recurrent pneumococcal meningitis secondary to a congenital anatomical defect in cribriform plate

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Abstract:

Recurrent meningitis is rare and accounts for approximately 1% of all cases of meningitis. Recurrent pneumococcal meningitis is often associated with underlying predisposing factors such as immunodeficiency and skull base anatomical defects and carries a high risk of life-threatening long-term sequel. An 11-year-old child presented with recurrent meningitis associating with cerebrospinal fluids (CSF) rhinorrhea. Blood and CSF cultures confirmed the growth of Streptococcus pneumoniae which was sensitive to Cefotaxime. CSF rhinorrhea was supported by a positive glucostix test. The defect in the cribriform plate was confirmed by MRI CSF cysternogram. The child responded to a 21-day course of intravenous Cefotaxime and the cribriform plate was repaired by Functional endoscopic sinus surgery (FESS). Appropriate early diagnosis and treatment of pneumococcal infections are a challenge to all clinicians in order to minimize immediate mortality and long term neurological outcomes especially, hearing impairment, and acquired hydrocephalus. Recurrent pneumococcal meningitis warrants further evaluation of underlying pathology either anatomical or immunodeficiency.

Keywords: Recurrent meningitis, streptococcus pneumonia, cribriform plate, Cefotaxime.

Introduction

Infections of the central nervous system in children are associated with high morbidity and mortality and the management often needs early attention and specialist input by a multidisciplinary team to prevent unacceptable long-term complications [1]. Recurrent meningitis is rare in children [2] and accounts for approximately 1% of all children with bacterial meningitis [3]. Recurrent meningitis is usually associated with underlying predisposing factors such as primary and secondary immunodeficiencies, presence of indwelling devices in the ventricular system, and congenital [4] or acquired anatomical abnormalities in the anterior cranial fossa that cause CSF rhinorrhea. Underlying anatomical defects are the aetiology in approximately 30-50% of the cases of recurrent bacterial meningitis [5]. Congenital anatomical abnormalities often cause management challenges as they are underdiagnosed, difficult to be accurately located, and carry a high cumulative risk of recurrent meningitis and related life-threatening sequel [6]. We report the case of an 11-year-old boy who presented with recurrent pneumococcal meningitis in a background CSF rhinorrhea since the age of 4 years following a congenital defect in the right cribriform plate.

Case history

An 11-year old boy second born to non-consanguineous healthy parents from a low socioeconomic community and who had a history of recurrent acute pyogenic meningitis presented with sudden onset of headache, high fever, and vomiting for 2 days and drowsiness for the one-day duration. He had several previous admissions with headache and fever to local hospitals for management of recurrent acute pyogenic meningitis but defaulted follow up due to social circumstances. The child did not report photophobia and phonophobia. There was no preceding history of trauma, surgery, or seizures, but he had been having intermittent, watery, and smelly nasal discharge from the right nostril since the age of 2 years for which no treatment was sought. Past medical history revealed that he had eight episodes of bacterial meningitis and one episode of bronchopneumonia since the age of 4 years. During the last three episodes, he had presented with a high fever of short duration, headache, vomiting, and a stiff neck and had been admitted to the local hospital and transferred to tertiary care hospital for further investigations and management. There was no history of early-onset severe infections or hospitalizations before the age of 4 years. The birth was uneventful and early developmental milestones had been achieved age appropriately. His school performance has been below average due to

frequent school absence for medical reasons. He was vaccinated according to the local EPI (Expanded program of immunization) schedule and therefore, he had not been vaccinated for capsulated organisms apart from receiving pneumococcal vaccine once following recurrent infections at the tertiary care hospital. There was no history of active infections following live attenuated vaccines.

Physical examination revealed an ill-looking, febrile (105F), and drowsy child with positive neck stiffness and Kernig's sign. BCG scar was present. He had watery stickily discharge from the right nostril and it was foul-smelling. He was examined for a foreign body in the nostril and no foreign bodies were found. Neurological and other systems examination was normal.

Investigations revealed high white blood cells (WBC) with predominant neutrophils (WBC-18X103/μL, neutrophils - 90%), low platelets (120x103/μL), high C-reactive protein (CRP-196mg/dl) and high Erythrocyte sedimentation rate (ESR- 120mm/hour). The blood picture revealed increased neutrophils with left shift and the presence of toxic granules compatible with a bacterial infection. The renal function had been within normal limits except borderline high blood urea (62mg/dL). The liver function had been elevated (ALT- 8 4 IU/L, AST-102 IU/L). Serum D-dimers were normal (120ng/dL). Serum fibrinogen had been within the normal limit (256 mg/dL). Bacterial meningitis was confirmed on all three occasions with strongly positive CSF reports for microscopy, high protein, and low sugar. Blood and cerebrospinal fluids cultures (CSF) grew Streptococcus pneumonia which was sensitive to Cefotaxime and resistant to penicillin in all episodes. CSF rhinorrheal fluid grew no organisms. Ultrasound abdomen revealed splenomegaly of 2cm below the left costal margin. EEG (electroencephalogram) demonstrated focal encephalitis (Figure-1). Non-contrast CT (computed tomography) of the brain showed changes of cerebritis and encephalitis (Figure-2). The ophthalmological evaluation was normal. He was treated for pneumococcal meningitis with intravenous Cefotaxime for 21 days. Repeated CSF and blood cultures upon completion of treatment revealed normal findings.

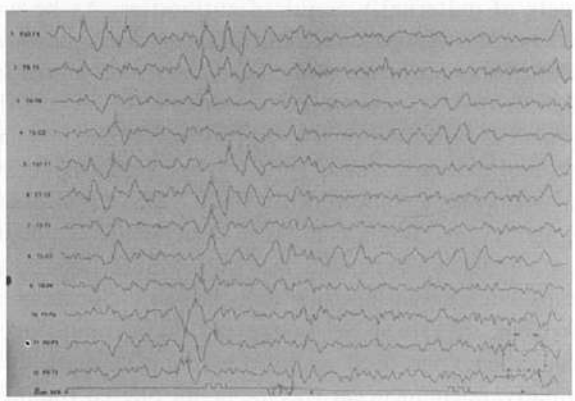

Figure-1: Slow waves which are compatible with the focal enchaphalitis (Red arrows)

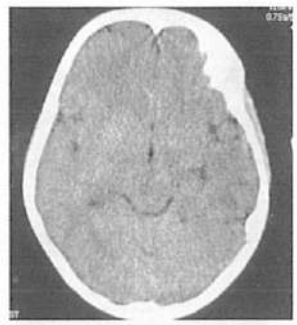

Figure-2: Hypodense area in the non-contrast computed tomography of the brain shows changes of cerebritis and encephalitis (red)

He was subsequently evaluated for recurrent bacterial meningitis. He was investigated for hypogammaglobulinemia and hypocomplementaemia with normal findings. Table 1 shows the findings of the immunodeficiency screen to rule out these causes.

Table 1 – Findings of the immunodeficiency screen

Investigation	Value on admission	Value at one month review
Total IgG	1440 mg/dL	1234 mg/dL
IgG1	-	1008 mg/dL
IgG2	-	300 mg/dL
IgG3	-	94 mg/dL
IgG4	-	24 mg/dL
IgA	280 mg/dL	120 mg/dL
IgM	252 mg/dL	100 mg/dL
Complement factor 3	250 mg/dL	-
Complement factor 4	70 mg/dL	-

Human immunodeficiency virus (HIV) screening was normal. CSF rhinorrhea was clinically suspected by the otorhinolaryngologist based on clinical characteristics of the discharge, absence of upper respiratory tract infections, and normal rhinoscopy findings. The nasal discharge was analysed by Glucostrix test and the findings supported CSF rhinorrhea. CT cisternogram was normal. However, an MRI CSF cysternogram was performed subsequently and a defect in the right cribriform plate was diagnosed. Based on investigations CSF rhinorrhea secondary to a congenital defect of the cribriform plate was confirmed. Figure 3 shows the findings of the CSF cisterogram which confirmed CSF leakage through the defect in the right cribriform plate.

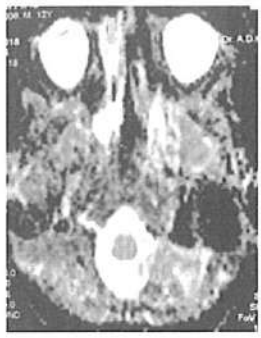

Figure 3- Findings of MRI cisterngram showed CSF leakage into right nostril

He underwent FESS (Fenestrated endoscopic sinus surgery) for repair of the defect in right cibriform plate. MRI cisternogram was not repeated following surgery to confirm the closure of defect as MRI studies were not freely available. Currently, he is being followed up for 24 months following repair of the defect and no recurrence was reported during this period. His school performance improved with improved school attendance. Hearing assessment during follow-up revealed normal findings.

Discussion

Recurrent bacterial meningitis is defined as the reemergence of signs and symptoms three weeks after a negative CSF culture with the same pathogen or at any time following a different CSF pathogen [7]. Most children with recurrent pneumococcal meningitis have underlying predisposing factors and it is crucial to diagnose these factors early to prevent and control further recurrences. Often it is difficult to diagnose the underlying aetiology for recurrent meningitis [8]. Primary and acquired immunodeficiency syndromes that are associated with increased risk of pneumococcal meningitis include complement defects, asplenia, HIV infection, interleukin-1-receptor-associated kinase-4 (IRAK-4) deficiency, and humoral immunodeficiencies (IgG subclass deficiency, agammaglobulinemia, common variable immunodeficiency). Congenital anatomical abnormalities that predispose to pneumococcal meningitis include meningoceles, skull base defects, inner ear abnormalities (Mondini dysplasia), neuroenteric, dermoid and epidermoid cysts [9]. Trauma [10] neurosurgery and infections account for acquired skull base defects.

The possibility of immunodeficiency was ruled out in this child by a negative immunodeficiency screen. The presence of CSF rhinorrhea assisted in clinically suspecting an anatomical defect in the cribriform plate. The type of pathogen isolated also helps in narrowing down the differential diagnosis. Ventricular devices, postcranial surgeries, acquired defects, and dermoids often predispose to Staphylococcal meningitis whereas pneumococcal meningitis recurs in children with underlying immunodeficiency and certain congenital anatomical defects. The diagnosis of this child was delayed till 12 years due to the lack of a proper referral system for follow-up and defaulting of medical care due to adverse socioeconomic circumstances in the family.

The traditional method of identifying glucose in nasal discharge using glucostrix would be supportive for the identification of CSF rhinorrhea if it is more than 30mg/dL in the absence of any blood contamination [11]. The test was positive (64mg/dl) in this child for CSF rhinorrhea. Other rhinorrhea fluid analysis methods include absorbent fillet test (double-ring sign), chloride, and total protein estimation in fluids although the specificity of these tests is low. The gold standard method had been detecting Beta-2 transferrin by immunofixation electrophoresis, but at least 2ml of nasal fluids is required for assay and clinical use is limited by high cost11. Neuroimaging is vital in those with unilateral leakage suggestive of CSF rhinorrhea. Both HRCT (high resolution computed tomography) scan and MRI cisternogram are equally effective in detecting CSF leakages [11]. Although HRCT failed to demonstrate the defect in the cribriform plate in this child, the defect was clearly evident in MRI cisternogram.

Endoscopic evaluation and management via endonasal approach have been popular in recent times with the acceptable morbidity for the correction of leaks in both ethmoid and sphenoid regions; however, the procedure requires exploration for the frontal sinus [12,13]. The success rate after first and second endoscopic surgery ranges from 87 to100% and 94 to 100% respectively. Late recurrences had been reported and they should be anticipated during long term follow up [14, 15]. Our child had been free of recurrences following surgery for the last 24 months although he needs further long term to follow up to detect potential long term complications of surgical procedure and recurrences.

Although the reported child had been having CSF rhinorrhea since the age of 2 years from the right nostril, he did not have a significant infection until 4 years at which he started attending preschool. This could be due to the absence of exposure to the infection and protective effects of maternally acquired antibodies. As he was born before the introduction of the Hib vaccine for Haemophilus influenza in the Sri Lankan immunization schedule, he had not been immunized for any of the capsulated organisms such as Streptococcus pneumoniae, Haemophilus influenza, and Neisseria meningitides. He had recurrent meningitis only by Streptococcus pneumoniae and this was in favor of an anatomical defect over complement deficiency that leads to infections by all encapsulated organisms. It is thought that meningococcal infections are rare in Sri Lanka and the meningococcal vaccine is not part of the national immunization schedule. Similarly, pneumococcal vaccine is not offered routinely in the national immunization program, and the vaccine is offered only in special circumstances where children have a high predisposition to invasive pneumococcal infections. The emergence of pneumococcal infections highlights the importance of the incorporation of pneumococcal vaccines

into the national immunization schedule in Sri Lanka.

Given the hardship of diagnostic and management challenges, it is crucial that children with recurrent pneumococcal meningitis secondary to anatomical abnormalities are dealt with by paediatrician, radiologist, microbiologist, otolaryngologist, and neurosurgeon in close collaboration. All children with undiagnosed aetiology need CT or MRI imaging of temporal bones, skull base, and paranasal sinuses to look for underlying anatomical abnormalities. All children need the evaluation of hearing during follow-up and 23 valent polysaccharide pneumococcal vaccine to reduce the risk of recurrences.

Conclusion

Appropriate early diagnosis and treatment of pneumococcal infections are a challenge to all clinicians in order to minimize immediate mortality and long term neurological outcomes especially, hearing impairment, and acquired hydrocephalus. Recurrent pneumococcal meningitis warrants further evaluation of underlying pathology either anatomical or immunodeficiency.

Acknowledgment

We would like to thank Director, T H Batticaloa, Dr. V.R Francis, Consultant Microbiologist and Dr V Jeevathas Consultant ENT Surgeon as they all contributed to diagnose the disease.

Availability of Data

The data that support the findings of this case report are available from Medical Records Department, Batticaloa Teaching Hospital, but restrictions apply to the availability of these data, which were used under license for the current report and so are not publicly available. Data are, however, available from the authors upon reasonable request and with permission of Medical Records Department, Teaching Hospital, Batticaloa, Sri Lanka.

Conflict of interest

The authors declare that there is no conflict of interest in publication of this article.

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Case Report

Methicillin Resistance Staphylococcus aures (MRSA) induce cavitary Pneumonia following Dengue Haemorrhagic fever: A Case report

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Abstract:

Dengue fever most common infection in Srilanka[6]. Dengue patients are vulnerable to secondary bacterial infections due to leucopenia[2,4]. Here we report a case 19 year old girl presented with haemoptysis and cavitary lung lesion following Dengue haemorrhagic fever, managed Methicillin Resistance Staphylococcus aures (MRSA) induce cavitatry lung pneumonia.

Keywords:
Dengue haemorrhagic Fever, MRSA, Cavitary lung lesion.

Introduction

Dengue fever most common infection in Srilanka. Dengue patients are vulnerable to secondary bacterial infections due to leucopenia (especially neutropenia), impairment of antigen-presenting cells' functions, reduction of the phagocytic and migratory capacities of macrophages, impairment of the interferon signaling pathway, impairment of the integrity of skin barrier against bacteria and other transient changes in the immune system caused by dengue virus[2,4,5].

MRSA stands for methicillin-resistant Staphylococcus aureus, a type of bacteria that is resistant to several antibiotics. In the community, MRSA most often causes skin infections. In some cases, it causes pneumonia (lung infection) and other infections. If left untreated, MRSA infections can become severe and cause sepsis. Common mode of transmission of this MRSA is skin wounds as well as if the patients in ward most mode of transmition is cannula site infection[1,2,4].

Case Presentation

19-year-old girl previously healthy a week back treated with dengue haemorrhagic fever (DHF). Presented with 2 days history of high grade fever, chills and rigors after discharged from the hospital. Associated with productive cough with yellow colour sputum with several episode of mild haemoptysis and lethargy. No features loss of weight loss of appitite or evening pyrexia and no contact history of Tuberculosis.

On admission she was febrile 1020F and ill look, not pale, no rashes, no ankle oedema no clubbing, evidence of cannula site tenderness was there and no palpable lymph node. Her vital parameters were blood pressure was 110/70mmhg, pulse rate 98 beats per minutes. Abdomen was soft and bilateral lung few coarse crepitation and bronchial breathing was audible at left upper zone of the lung.

Her initial laboratory workup full blood count(FBC) reveled white cell count (WBC) 18 x 103, neutrophil (9.5x103u/L 75%), lymphocyte (6.5x103u/L 20%), eosinophil (0.41x103u/L 3%) haemoglobin 13.5g/dl and platelets 144x 109, C-Reactive protein (CRP) 180mg/l, AST 197U/l, ALT 112U/l, Creatinine 67umol/l, Na+ 135mmol/l, K+ 4.1mmol/l, Erythrocyte Sediment Rate (ESR) 80mmin 1st hour, Urine full report shows only trace albumin and pus nil. Chest xray shows left side lung upper lobe cavitary shadow (figure 1). CECT chest revealed left side upper lobe cavity (Figure 2). Blood culture was found Methicillin Resistance Staphylococcus aures (MRSA) sensitivity to clindamycine, Erythromycin, Teichoplanin, Vancomycin and Linozolid. Urine and sputum culture was no growth. Sputum Acid fast bacilli (AFB) three samples negative, TB PCR gene xpert was negative She was managed with oral linezolid 600mg tds for a week. She underwent flexible bronchoscopy examination that was completely normal study and bronchial wash samples took for TB PCR, Culture ABST, AFB stains, Fungal studies and TB Culture those investigations were normal her 2D ECHO also normal, Antinuclear antibody and p-ANCA and c-ANCA were unremarkable.

After six day of the admission her FBC reveled WBC 6.4 x 103 neutrophil 5.8 x103 (64%) lymphocyte 3.8 x 103 (28%), CRP 35mg/l, ESR 30mm 1st hour and repeat blood culture was no growth and her haemoptysis settled and recorved fully. Repeat chest x-ray in four-week cavity completely resolved(Figure 3).

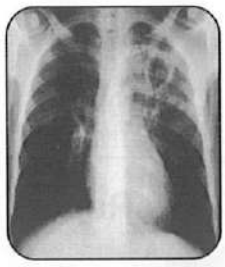

Figure 1

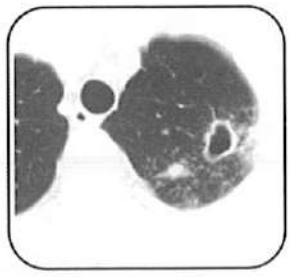

Figure 2

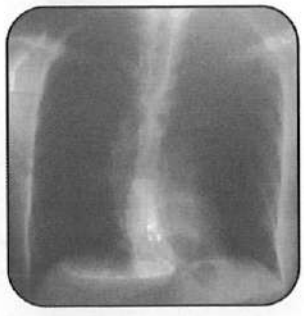

Figure 3

Discussion

Dengue patients are vulnerable to secondary bacterial infections due to leucopenia (especially neutropenia), impairment of antigen-presenting cells' functions, reduction of the phagocytic and migratory capacities of macrophages, impairment of the interferon signaling pathway, impairment of the integrity of skin barrier against bacteria and other transient changes in the immune system caused by dengue virus [2,3,4,5]

Development of an abscess at the site of the first intravenous cannula indicated that as a potential site of MRSA entry. If that was the case MRSA that has been on his skin or from the hands of the nursing staff (who inserted the cannula) could have been the source.

Two percent of the population carries MRSA [4]. A study from another Sri Lankan teaching hospital shows that >10% of the nursing staff were at risk of transmitting MRSA [4].

Here our patient recently treated with dengue hemorrhagic fever associated with cannula site infection that was the source of the entry of the infection MRSA and blood culture also strongly evidence of this.

Treatment option according to the sensitivity if patient severely ill start empirical antibiotics after blood cultures mainly with gram positive cover[1].

Conclusion

MRSA is commensal in skin if skin is breached that is the potential site of inversion. When insert cannula always stick to the aseptic techniques that prevent the secondary infections.

Acknowledgement

We would like to thank Dr. N. Egodawela consultant respiratory physician for his valuable input on managing this patient.

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6. Dengue national guideline

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Case Report

Severe Anaphylaxis with Kounis Syndrome following Covid-19 Vaccination

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Abstract:

Severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) infection which is also designated as COVID-19 results in a global pandemic. Vaccines to prevent SARS-CoV-2 infection are considered to be the most promising approach for curbing the pandemic. Kounis syndrome is the occurrence of acute coronary syndromes in the setting of allergic or hypersensitivity insults. This syndrome is caused by inflammatory mediators released during the allergic activation process.

Keywords:
Kounis syndrome and hypersensitivity reaction

Introduction

Severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) infection which is also designated as COVID-19 results in a global pandemic. Vaccines to prevent SARS-CoV-2 infection are considered to be the most promising approach for curbing the pandemic.

Given the importance of the vaccine in fighting this public health crisis, understanding the allergic reactions with the approved COVID-19 vaccines is crucial. The vaccines against COVID-19 are new and some have a novel mechanism of action. Moreover, it is not surprising that anaphylaxis has not been reported in the clinical trials to-date, given the very low incidence and the exclusion of individuals with a history of hypersensitivity reactions in most studies. [1].

Kounis syndrome is the occurrence of acute coronary syndromes in the setting of allergic or hypersensitivity insults. This syndrome is caused by inflammatory mediators released during the allergic activation process [2]. Kounis Syndrome is known to occur as a result of hypersensitivity reaction to vaccines. We herewith report a case of severe anaphylaxis complicated with Kounis syndrome following ChAdOx1nCoV-19 Corona Virus Recombinant (Oxford Aztrazeneca) Vaccine.

Case Presentation

A 47 years-old-female health staff attendant with a history of anaphylaxis for contrast media received her first dose of ChAdOx1 nCoV-19 Corona Virus Recombinant Vaccine. Following few minutes after the vaccination she developed severe tightening chest pain which progressed to shortness of breath. Though there was no urticarial rash, she developed intense itching of body. Examination revealed a dyspnoeic patient with respiratory rate of 32/min. Her blood pressure was unrecordable and she was tachycardiac with pulse rate of 120 bpm. Lung examination revealed diffuse rhonchi. Saturation was not detected by the pulse oximeter. She was immediately attended by the medical team and intramuscular 1:1000 adrenaline 0.5ml was given.

Subsequently, she received another 3 boluses of intramuscular adrenaline, IV Chlorphenamine 10mg and IV hydrocortisone 200mg which improved the blood pressure to 70/50 mmHg. She was started on adrenaline infusion with rate of 0.1 mcg/kg/minute. With infusion her blood pressure was maintained around 100/70. Her pulse rate stabilized around 100 bpm. The lung ronchi were cleared and saturation was improved to 98%. Gradually her adrenaline infusion was tailed off over 12 hours and she could maintain her BP around 100/60 mmHg without inotrope support. Figure1 is the first ECG on admission (1.5hr after anaphylaxis)

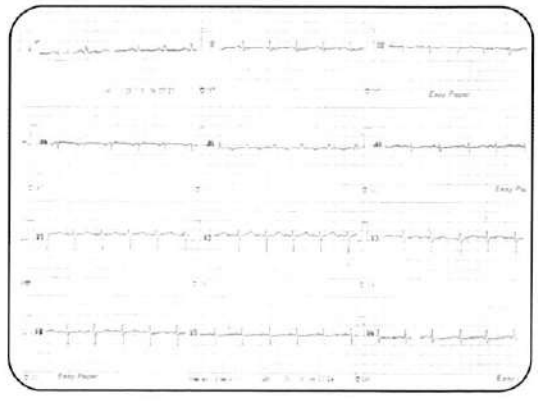

Figure 1: The first ECG on admission (1.5hr after anaphylaxis). ECG showed sinus rhythm with marginal GT prolongation (QTc with Bazzet formula 465ms)

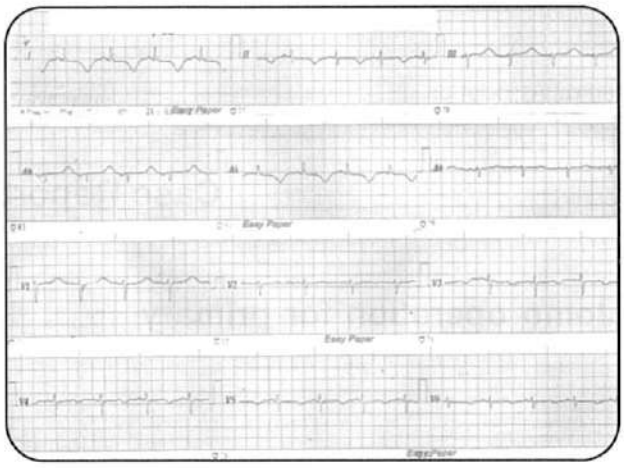

Figure 2 : The repeat ECG following 24 hours. There were dynamic ECG changes with development of deep T waves in lead I,II,aVL,V3,V4, V5,V6. QTc with Bazzet formula 581ms.

Her troponin I titre was progressively rising from 3.5 ng/ml, 5.76 ng/ml 7.8 ng/ml (<0.5). Her trans-thoracic echo showed inferolateral hypokinesia with ejection fraction of 50%. Her serum electrolytes were normal. She was referred to cardiology team for coronary angiogram.

Because of the past history of anaphylaxis with contrast, it was decided by the cardiology team to manage the patient medically without coronary angiogram. She was treated with Enoxaparin and dual antiplatelets. The long QT in the ECG was attributed to intravenous chorperanamine which she received for 2 days. Subsequently it was corrected once culprit drug was withdrawn. She was discharged with good recovery after one week of inward care.

Discussion

Out of the first 4,041,396 doses of Moderna vaccine administered in the United States, 10 cases were determined to be anaphylaxis, with a rate of 2.5 anaphylaxis cases per million. Nine of these anaphylaxis cases, included a patient history of allergies or allergic reactions [3]. Out of first 1,893,360 doses of Pfizer-BioNTech vaccine administered in the United States, 21 cases were determined to be anaphylaxis (a rate of 11.1 per million doses administered) [4]. The interim analysis of four randomised controlled trials in Brazil, South Africa, and UK out of 12021 ChAdOx1 nCoV-19 vaccine received, only 1 case of anaphylaxis was reported [5]. As for our knowledge, this is the first case report of a severe anaphylaxis following covid-19 vaccination in Sri Lanka.

In addition to anaphylaxis, our patient experienced an acute coronary syndrome following the vaccination. Two mechanisms can be used to explain the occurrence of acute coronary syndrome in this patient. The first being the Allergic vasospastic angina, also termed as Kounis syndrome. In Literature number of cases have been reported in vaccine associated with Kounis. Kundi et al. report a patient who developed Kounis syndrome following an allergic reaction to a tetanus vaccine [6]. Our patient might have experienced Kounis syndrome following covid-19 vaccination. In current literature there are no case repots Covid-19 Vaccine associated with Kounis syndrome

The other mechanism explaining the acute coronary syndrome is the effect of multiple bolus doses of adrenaline. This could have precipitated the coronary vasospasm which lead to subsequent acute coronary syndrome.

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Case Report

Outcome of artery-sparing modified Palomo operation for primary varicocele in a cohort of soldiers

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Abstract:

Tuberculosis is one of the commonest communicable diseases in the world. The incidence has been rising specially in Africa and Asia due to the association with HIV and other immunosuppressive conditions such as diabetes mellitus and chronic kidney disease. Clinical manifestations of tuberculosis may range from commonly seen pulmonary tuberculosis to the rarely seen vasculitis with peripheral neuropathy.

This case report illustrates a patient who presented with combined chronic severe sensory motor axonal polyneuropathy and digital gangrene with positive PR3 anti neutrophil cytoplasmic antibody (PR3 ANCA) and was found to have tuberculosis.

Keywords:
Tuberculosis Vasculitis Broncho alveolar lavage

INTRODUCTION

Tuberculosis is caused by mycobacterial species of Mycobacterium tuberculosis complex (MTb). There are four main mycobacterial species, namely ***Mycobacterium tuberculosis, Mycobacterium bovis, Mycobacterium africanum*** and ***Mycobacterium microti***. The infection develops due to inhalation of bacteria. However, the hosts immune response can lead to the development of latent tuberculosis, primary active tuberculosis and reactivation tuberculosis.

The majority of tuberculosis cases are due to reactivation of latent infection. Reactivation is usually due to immunosuppression as in HIV coinfection, immunosuppressive therapy including corticosteroids, diabetes mellitus, end stage chronic kidney disease, malnutrition and aging. The typical appearance of post-primary tuberculosis are patchy consolidations or linear and nodular opacities[1].

Screening for pulmonary tuberculosis is done using sputum for microscopy, smear and culture. If the patient is unable to produce sputum, induced sputum or bronchoscopy and lavage can be used to obtain respiratory secretions[2].

The association of tuberculosis with ANCA associated vasculitis is considered unusual and is a diagnostic challenge, as both share similar features clinically, radiologically and histologically. However when both these conditions are associated, the possibilities are the coexistence of both tuberculosis and vasculitis, antigenic exposure to tuberculosis triggering ANCA vasculitis, incidental finding of ANCA in patients with tuberculosis without pathogenic value and development of tuberculosis while on immunosuppression for vasculitis[3].

CASE HISTORY

The patient was a 73-year-old female who got admitted following bilateral upper and lower limb arthralgia and impaired hearing for ten days' duration. She has been a non-vegetarian and was on treatment for anaemia. She had no contact history of tuberculosis. Examination revealed bilateral upper limb wrist drop with sensory motor polyneuropathy and bilateral lower limb stocking type sensory loss with distal muscle weakness.

She had reduced breath sounds over the right lower chest and had saturation of 98% on air. During the hospital stay the patient developed dry gangrene of distal phalanges of left second, third, fourth and fifth fingers. (Figure 1)

Figure 1

The patients' investigation findings are summarized in table 1

Full blood count	White cell count 13.41*109/l Neutrophils 79.9% Lymphocytes 12.8% Haemoglobin 9.8g/dl Mean corpuscular volume 79.1fl Platelet 512*109/l
Erythrocyte sediment rate	130mm/1st hr
C reactive proteins	186.6 mg/l
Liver profile	Alanine aminotransferase 39u/l Aspartate aminotransferase 42u/l Alkaline phosphatase 138u/l Total protein 72g/dl Albumin 20g/dl Globulin 52g/dl Total bilirubin 5.1µmol/l
Serum urea	2.1 mmol/l
Serum sodium	136mmol/l
Serum potassium	4.2mmol/l
Serum creatinine	39µmol/l
Urine full report	Protein + WBC 1-2/hpf RBC 8-10/hpf
Serum calcium	2.4 mmol/l
Serum magnesium	0.92mmol/l
Serum phosphorous	1.51mmol/l
Serum uric acid	99µmol/l
Retulocyte count	3.37%
Fasting serum sugar	5.29mmol/l
Blood culture	No growth
Urine BenceJones protein	Negative
Immunofixation / BenceJones	No abnormal bands in the separation
VDRL	Non reactive
Retroviral screening	negative
Thyroid function tests	TSH 6.51mIU/l Free T4 0.89ng/dl
Rheumatoid factor	32u/l
APLS screening	DRVTT positive for lupus anticoagulant
Anti nuclear antibody	1:80 negative
Cryoglobulin	Negative
Mantoux test	Negative
Bronchoalveolar lavage for cytology	Acute inflammatory smear
Fasting lipid profile	Total cholesterol 3.13mmol/l Triglyceride 1.43mmol/l HDL 0.34mmol/l LDL 2.14mmol/l
ANCA	pANCA negative PR3ANCA positive

Table 1

The patient's chest X ray revealed right lower zone consolidation. (Figure 2)

The blood picture was suggestive of anaemia of chronic disease with evidence of iron deficiency anaemia. Serum protein electrophoresis was suggestive of chronic inflammation with increased acute phase reactants with polyclonal increase in gamma globulins. The skeletal survey revealed no lytic lesions and PR3-ANCA was positive.

The contrast enhanced computed tomography of neck, chest, abdomen and pelvis (CECT-NCAP) showed right middle lobe consolidation and circumferential thickening of mucosa in the second part of the duodenum. Subsequent upper gastrointestinal endoscopy reported oesophageal candidiasis, gastric antral vascular ectasia (GAVE) with normal first and second parts of the duodenum.

The skin biopsy from the edge of the gangrenous digit of the middle finger was not suggestive of vasculitis and the patient did not consent for the repeat biopsy.

The electrophysiological findings were more in favour of combined chronic severe sensory-motor axonal polyneuropathy of lower limbs, whereas the upper limbs studies demonstrated electrophysiological evidences of mononeuritis multiplex. The possibilities were vasculitis, secondary to infective pathology (tuberculosis or leprosy) and "burnt-out" chronic inflammatory demyelinating polyradiculoneuropathy (CIDP).

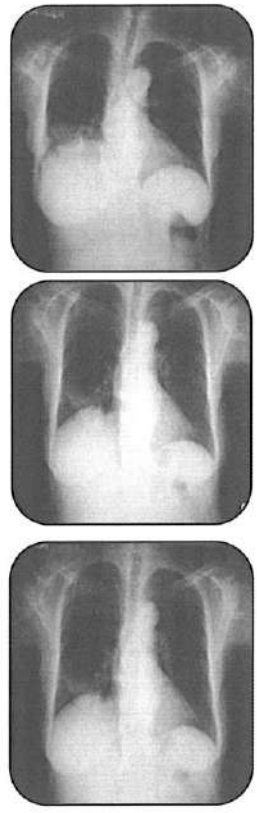
The pure tone audiometry revealed right sided conductive hearing impairment.

Although her urine protein creatinine ratio (UPCR) was in nephrotic range, the patient did not consent for renal biopsy.

She was clinically diagnosed with possible ANCA associated vasculitis and she was commenced on intravenous methylprednisolone 1g pulses daily for three days when she developed gangrene of her fingers and converted to oral prednisolone for vasculitis. Additionally, she was started on bosentan daily. Since there was renal involvement, intravenous cyclophosphamide was also initiated.

Although she showed only partial response initially, her condition started to decline thereafter.
At eight weeks since admission, the broncho-alveolar lavage sample for Mycobacterium tuberculosis culture became positive. Anti-tuberculosis treatment was started in addition to oral steroids.

The patient's clinical status improved markedly following the commencement of treatment for vasculitis. Her general well-being, appetite and hearing were also improved. The follow up chest X rays revealed improvement of the consolidation.



(Figure 2)

DISCUSSION

Pulmonary tuberculosis can present in the same way of systemic vasculitis4. In these instances, the differentiation of tuberculosis from systemic vasculitis necessitate biopsy, serological marker detection and genetic testing for tuberculosis. In the presence of strong clinical suspicion of tuberculosis, the investigations would range from detection of acid fast bacilli in sputum, mantoux test, culture and genetic testing. Tuberculosis related vasculitis is one of the rare clinical manifestations5. The unusual tuberculous granulomatous vasculitis is generally associated with meningitis and retinitis5. Earlier published case reports on tuberculosis and vasculitis has shown cutaneous leukocytoclastic vasculitis[5].

In this case, the diagnosis of systemic vasculitis became challenging due to the unavailability of renal biopsy and an inclusiveness of the skin biopsy. Even though PR3-ANCA is considered a sensitive marker of granulomatosis with polyangiitis6 it can be present in patients with tuberculosis without a pathological significance[3].

A few case reports revealed tuberculosis presenting as peripheral gangrene, cutaneous ulcers with absent peripheral pulses6and rarely nasal septal perforation[7].

CONCLUSION

It is of great importance to consider tuberculosis among patients in highly prevalent regions with clinically and diagnostically non-specific presentation and urge for a high index of suspicion.

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Case Report

FENOFIBRATE INDUCED MIXED CHOLESTATIC HEPATITIS;

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Abstract:

Fenofibrate, a fibric acid derivative generally presumed to be a fairly safe drug in comparison to older fibrates. Even though mild elevation of liver enzymes is common following fenofibrate administration we could find only few case reports describing severe mixed cholestatic hepatitis in PubMed and Google scholar search. We present 38-year-old man presenting with mixed cholestatic hepatitis following Fenofibrate, probably the first of its kind in Sri Lanka.

Keywords:

Fenofibrate, drug induced hepatotoxicity, mixed cholestatic hepatitis

INTRODUCTION

Fenofibrate, a fibric acid derivative currently recommended to treat dyslipidaemia and hypertriglyceridaemia. Fenofibrate; which has replaced older fibrates such as gemfibrozll and bezafibrate. It is generally presumed to be a fairly safe drug in comparison to the former group. It's an agonist for the nuclear transcription factor peroxisome proliferator-activated receptor- alpha (PPAR-alpha). Even though mild elevation of liver enzymes is common following fenofibrate administration we could find only few reports describing severe mixed cholestatic hepatitis. DILI (Drug Induced Liver Injury) from fenofibrate is a rare event probably arising in fewer than 1:10,000 exposed persons. [1]

Fenofibrate is associated with idiosyncratic acute liver injury with a typical latency of 5–8 weeks although this can be more prolonged. [1] The pattern and severity of injury is variable but a chronic progressive cholestatic disease can occur. [1] Although rare, the injury can be severe and patients treated with fenofibrate should be alerted to the possibility of liver injury and importance of discontinuing therapy for unexplained symptoms or signs of possible liver injury. [1]

CASE PRESENTATION

A 38 years old man admitted with generalized body weakness, loss of appetite, yellowish discolouration of the eyes and urine for one-week duration. He denied abdominal pain or pale stools. There was no history of a viral illness or alcohol consumption. He denied a history of joint pains or rashes. His drug history revealed that he was on Fenofibrate 200 mg daily for recently diagnosed Dyslipidemia, which had been continuing unsupervised for the last two months prior to presentation. He denied history of intravenous drug abuse, sexual promiscuity, blood transfusions, consumption of native medications and any family history of liver diseases.

He was conscious and alert. GCS 15/15. BMI 22 kg/m2. Physical examination revealed deep icterus and tender hepatomegaly. Rest of his system examination was unremarkable.

His lipid profile was as below.

Total cholesterol	615 mg/dl
Triglycerides	281 mg/dl
HDL cholesterol	11 mg/dl
LDL cholesterol	547 mg/dl
Cholesterol/HDL	55.91

Investigations on admission

WBC 8.96 x 109 l-1 (4-11)	AST 204 U/L (0-40)
Hemoglobin 13.2 g/dL (11-16)	ALT 253 U/L (10-40)
Platelet 312 x 109 l-1 (150-450)	ALP 199 U/L (30-120)
CRP 4 mg/L (<6)	Total Bilirubin 14.9mg/dL (0.3-1.2)
Direct 10.5 mg/dL	
Serum Sodium 138 mmol/L (136-145)	Gamma GT 341 U/L (11-50)
Serum Potassium 4.8mmol/L (3.5-5.5.)	Serum Albumin 4g/dL (3.7-5)
Serum creatinine 83 µmol/L (80-115)	Serum Amylase 50 IU/L (0-80)
Hepatitis A IgM Negative	Serum Transferrin Saturation 25%
Hepatitis C IgM Negative	Prothrombin time (PT) 11S INR 1.03
Hepatitis B Surface Ag Negative	ESR 18 mm/Hr.
Anti-Nuclear Antibody (ANA) Negative	Serum ceruloplasmin levels 32mg/dL (20-35)
Anti-Mitochondrial Antibody Negative	HbA1C 5.8%
HIV I/II Negative	

Ultrasound scan of the abdomen revealed grade I fatty liver. There was no dilated biliary tract or visible biliary calculi. Eye examination failed to reveal Kayser-Fleischer rings suggestive of Wilson's disease.

With the available investigations tentative diagnosis of mixed cholestatic hepatitis induced by fenofibrate was made. Fenofibrate was withheld. Ursodeoxycholic acid 500mg bd was started. Diagnosis of Definite familial hypercholesterolaemia (FH) also made according to Dutch lipid clinic network diagnostic criteria scoring 9(>8). Ezetimibe 10mg mane added due to presence of high fasting cholesterol and LDL levels. Statin was not added due to possible further alteration of liver enzymes. Patient was educated regarding condition. He was discharged and reviewed after three weeks.

After three weeks following treatment he remained icteric with mild improvement in his liver profile. Ultrasound guided liver biopsy was carried out at the end of the third week and same treatment was continued.

Biopsy revealed portal inflammation, prominent cholestasis and hepatocellular injury. Hepatocyte injury localized to the zones of cholestasis. Pearl stain for iron was negative. Patient was reviewed after 4 weeks with a lipid profile and liver profile.

AST 55 U/L (0-40)	Total cholesterol 190mg/dl
ALT 110 U/L (10-40)	Triglycerides 130 mg/dl
ALP 115 U/L (30-120)	HDL cholesterol 35 mg/dl
Total Bilirubin 1.1mg/dL (0.3-1.2) Direct 0.4 mg/dL	LDL cholesterol 150 mg/dl
Gamma GT 55 U/L (11-50)	Cholesterol/HDL 5.4
Serum Albumin 3.9g/dL (3.7-5)	

He was advised regarding life style changes including exercise. Family screening carried out to detect other affected family members with Familial hypercholesterolemia. Ursodeoxycholic acid was omitted and Ezetimibe was continued.

DISCUSSION

The diagnosis of Fenofibrate induced liver injury was made after excluding other possible causes and establishing its causal relationship with the event.

Identifying the pattern of liver injury can guide diagnostic approach to DILI, including appropriate further diagnostic testing necessary to rule out other causes of liver injury. Calculating the ratio of the alanine aminotransferase (ALT) value/ALT upper limit of normal (ULN) divided by the alkaline phosphatase (AP) value/AP ULN can help to differentiate hepatocellular from cholestatic liver injury. The result; R ratio is typically ≥ 5 in hepatocellular injury, < 2 in cholestatic liver injury, and between 2 and 5 in mixed hepatocellular/cholestatic liver injury. In index case ratio fell between 2-5 made it a mixed pattern.

Histologic findings are not diagnostic for a specific cause of DILI. In our patient we performed the liver biopsy as he had a lagged clinical and biochemical response. Biopsy was cholestatic in nature supporting our diagnosis, pearl stain was negative making haemochromatosis unlikely. Liver enzymes were gradually dropped following the cessation of the culprit drug reinforced the diagnosis.

There have also been multiple reports of clinically apparent liver injury in patients on Fenofibrate, but in our literature research we could find only few reports describing Fenofibrate induced Mixed cholestatic hepatitis. [2], [3]. For the best of our knowledge we couldn't find any report originated from Sri Lanka.

Fenofibrate accounted for 7 of 1229 patients (0.6%) with DILI enrolled during the first 12 years of the DILIN Prospective study. [1] The injury did not appear to be dose related.

CONCLUSION

Fenofibrate is associated with idiosyncratic acute liver injury with a typical latency of 5–8 weeks, although this can be more prolonged. This emphasize the importance of having the clinical suspicion of this potentially serious drug reaction and early withdrawal of causal agent to avoid disastrous outcomes.

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Case Report

A Case of Incomplete intestinal obstruction due to a duodenal web located in the 3rd part of the duodenum

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Abstract:

A duodenal web refers to a complete or incomplete obstruction at the duodenum due to a membranous web or intraluminal diverticulum. When the obstruction is incomplete, as it was in our case, the clinical symptoms are variable and the diagnosis can be difficult.

Keywords:
duodenal web

Introduction

A duodenal web refers to a complete or incomplete obstruction at the duodenum due to a membranous web or intraluminal diverticulum.

Webs and Atresias occur due to failure of duodenum to recanalize during the period of the 6th to the 8th weeks of gestation (1). This abnormal process leaves behind a web made out of only mucosal and submucosal layers. The muscularis layer is typically absent (2).

Duodenal atresia and duodenal web are reported causes of duodenal obstruction with the incidence that ranges between 1/10000 up to 1/40000 live births (2). Unlike the duodenal atresia which typically presents itself with the double bubble sign immediately after birth, duodenal web may seldom remain undiagnosed until beyond infancy (3).

When the obstruction is incomplete, as it was in our case, the clinical symptoms are variable and the diagnosis can be difficult.

Case History

We report a case of a 40 day old female premature infant who was born in Base hospital, Valaichenai, at 34 weeks of gestation by normal vaginal delivery. The baby was the 3rd of non-related parents and her brother (12yrs) and sister (15yrs) are healthy. The mother had an immediate past history of intrauterine death at the POA of 35 weeks in her previous pregnancy.

This unplanned pregnancy of the 41 year old elderly mother had been complicated with gestational diabetes as the mother was already diagnosed with chronic type 2 diabetes mellitus for 7 years and on oral metformin 500mg tds. There is a strong family history of type 2 diabetes from maternal side.

During antenatal period the mother was advised on proper diet and treated with subcutaneous insulin in addition to metformin but the control was not adequate. Apgar Scores were 9, 9, and 10 at 1, 5 and 10 minutes respectively. The birth weight was 2.38kg. The baby developed hypoglycemia on day 1 of life due to lack of milk flow in the mother. Formula feeding was initiated at hospital and discharged home on day 4 of life with the satisfactory weight gain.

The baby passed meconium within 24 hours of life and had regular bowel movements 5 – 6 times a day thereafter. From 1st week of life onwards some spitting and occasional vomiting was noted after feeds but the weight gain was adequate.

At 38 days of life the baby developed severe non bilious vomiting 8 -10 times a day. Urine output was reduced to 3 times per day. Bowel not opened for 2 days with increased irritability. There was no abdominal distension or fever. After admission to Teaching hospital, Batticaloa, baby has developed multiple episodes of bilious vomiting.

The baby was adequately grown for her age. Her vitals on admission were consistent with some dehydration.

Her blood tests at presentation shows hypochloremic metabolic alkalosis and mild indirect hyperbilirubinemia. Initial abdominal X-ray reveals double bubble sign consistent with duodenal obstruction and an Ultra sound scan abdomen reveals pre-stenotic dilatation of proximal part of the duodenum & stomach. An upper GI contrast study reveals delayed passage of contrast medium as well as change in caliber between part 3 & 4 of the duodenum.
On day 3 of admission baby was prepared for operative exploration. At laparotomy, normal rotation was found and there was no any associated annular pancreas or biliary tree abnormalities. Duodenostomy revealed a web with the pinpoint hole in the center. The web was completely excised and a duodenodudenostomy made in between 3rd and 4th part of the duodenum. Nasogastric tube inserted to decompress the stomach.

The baby was then admitted to NICU for pain control and observation for 24 hours. She was well covered with intravenous antibiotics (Cefuroxime & Metranidazole) and proton pump inhibitors. Despite the antibiotic treatment the baby developed post-operative sepsis but treated accordingly. Oral feeding was started gradually after 48 hours of bowel rest. Domperidone was started to help in gut motility. The baby fully tolerated oral feeds in 3 days and with the satisfactory weight gain for 3 consecutive days baby was discharged home.

During her 1st follow up visit in 2 weeks post discharge and second follow up visit in 10 weeks post discharge baby showed sustained growth and weight gain with no clinical signs of intestinal obstruction.

Discussion

Congenital duodenal obstructions might be complete or partial and can be classified as either intrinsic or extrinsic. The intrinsic lesions include duodenal atresia or web. While extrinsic lesions include anterior portal vein, duodenal duplication, malrotation with Ladd's bands and annular pancreas (3). During infancy, common causes primarily include duodenal atresia and/or webs. Hypertrophic pyloric stenosis occurs later around 6 weeks of age (1).

Most of the duodenal webs are located in the second part of the duodenum (85 – 90%), rare cases are reported beyond second part (as in our patient) (4). Because most duodenal obstructions occur distal to the ampulla, the vomitus is bile stained in more than two thirds of cases. Non bilious vomiting however, can occur when the ampulla of Vater is inserted distal to the web (4).

As our patient presented with severe non bilious vomiting with biochemical evidence of hypochloremic metabolic alkalosis and indirect hyperbilirubinemia we initially suspected pyloric stenosis but after admission the bilious vomiting was evident and the radiological investigations further confirmed a duodenal pathology.

Though several studies have confirmed the causal relationship between gestational diabetes and the duodenal atresia still there are limited studies for the same in case of duodenal webs.

Conclusion

Duodenal webs can present with wide range of clinical scenarios irrespective of age in which the diagnosis can be difficult. We need further studies to establish the relationship between the gestational diabetes and duodenal webs as our case is highlighting the necessity.

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Picturs

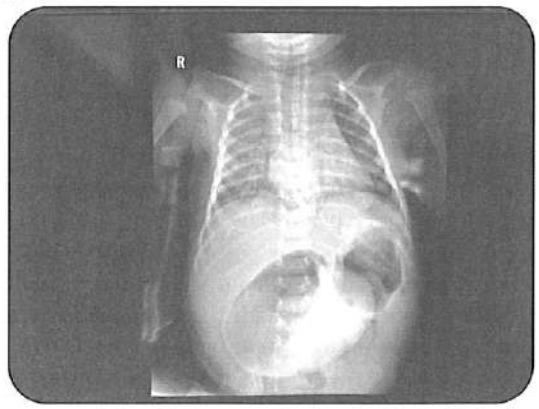

Figure 1 – Abdomial X-ray with the double bubble sign

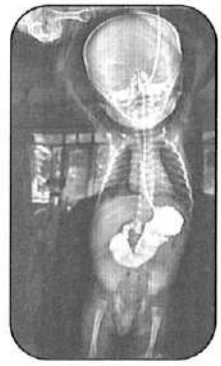

Figure 2 – Contrast study shows change in caliber between 3rd and 4th part of duodenum

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Case Report

Guillain-Barré syndrome with preserved reflexes; a not to miss diagnosis

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Abstract:

Guillain-Barré syndrome (GBS) is an acquired acute autoimmune polyradiculoneuropathy carrying a favourable outcome if intervene early. Weakness with Hyporeflexia or arereflxia considered as the hallmark of GBS. There have been studies, describing cases with acute motor axonal neuropathy AMAN that presented with preserved deep tendon reflexes (DTRs), or hyperreflexia [1–4] mainly in Chinese, Japanese, and European populations. Albeit rare, pure sensory symptoms, asymmetric weakness, and preserved or increased DTRs should not exclude the diagnosis of GBS. Clinicians should have a high clinical suspicion to diagnoses these heterogeneous presentations of GBS early to have a better outcome. We report a case of 26-year-old male admitted with bilateral lower limb weakness found to have an axonal type GBS with preserved DTRs probably the first of kind reported in Sri Lanka.

Keywords:
Guillain-Barré syndrome

Introduction

Guillain-Barré syndrome (GBS) is an acute monophasic illness causing a rapidly progressive polyneuropathy with weakness or paralysis. Diagnostic criteria for GBS include presence of progressive weakness and absence of deep tendon reflexes (DTRs) in weak limbs. Current GBS classification contains two major subtypes, acute inflammatory demyelinating polyneuropathy (AIDP) and acute motor axonal neuropathy (AMAN), based on the underlying pathogenesis. There have been studies, describing cases with AMAN that presented with preserved (DTRs), or hyperreflexia [1–4]. It is now well recognized that mainly AMAN and rarely AIDP can present with normal or exaggerated DTRs [5, 6]. This can make a delay in diagnosis which might later alter the prognosis. Thus clinicians should be aware of heterogeneous presentations of GBS including pure sensory symptoms, asymmetric weakness, and preserved or increased DTRs.

Case report

A 26-year-old previously healthy male was admitted with bilateral lower limb weakness for 3 days. His main complaint was inability to get up from sitting position. His symptoms were progressive making him unable to walk independently. There were no sensory, bladder or cranial nerve symptoms. There was no muscle pain. He denied a history of recent vaccination, antecedent illness or snake bite. On examination, limbs were flaccid, and power of the proximal muscles of the lower limb was 3/5 (Medical Research Council grading) and distal muscles was 4/5. DTRs were normal throughout the course of illness with bilateral flexor planters. Upper limb muscle power was normal. There was mild truncal weakness with preserved neck muscle power. Sensory and cerebellar system was normal. Cranial nerve examination was normal. Respiratory rate was 13/min. SPO2 98% on air. Full blood count was normal with total and differential counts, electrolytes were normal (Na,K,Mg,Ca), Creatinine phosphokinase, thyroid functions and ESR were within normal ranges. Magnetic resonance imaging (MRI) of brain and spine were normal. Nerve conduction study(NCS) on D3 showed F wave abnormalities with motor amplitude reduction suggestive of pure motor axonopathic variant of GBS (acute motor axonal neuropathy [AMAN]). CSF examination on D12 of the illness showed 2 cells (100% lymphocytes). CSF sugar was 97 mg/dL (plasma glucose 105 mg/dL) and protein was 30 mg/dL. He was treated with plasmapheresis. He had a rapid recovery, on discharge he could able to walk independently.

Discussion

GBS comprises group of diseases which describe immune mediated damage mostly to peripheral nervous system. Depending on the underlying pathology it is further divided into two categories; Demyelinating and Axonal. Axonal type further divided into AMAN and acute motor and sensory axonal neuropathy (AMSAN). Even though hyporeflexia or areflexia is characteristic of GBS there have been reports of GBS with preserved or exaggerated DTRs mainly in its axonal forms. [1–4]

Antecedent Campylobacter jejuni infection is commonly associated with GBS which can manifest as abdominal pain and diarrhoea. Our patient denied such history. He might had a subclinical infection which has gone unnoticed. Unfortunately, we could not do anti-Campylobacter jejuni antibodies, anti-GM1, GM1b, and GD1a ganglioside antibodies due to financial restraints. It is well recognized that one week after symptoms onset the CSF protein will be increased in only 50% of cases, and by the second week the percentage increases up to 80% [7]. Anyhow on day 10 of the illness index case didn't show albumin-cytologic dissociation.

Although preservation of reflexes may simply be due to sparing of the sensory afferent pathway, the occurrence of brisk reflexes suggests a central mechanism. Dysfunction of inhibitory systems in the spinal interneurons has been proposed [8] Generally AMAN has been associated with extensive axonal loss and poor outcome, this subgroup with preserved or exaggerated reflexes recovers rapidly. [8]. In our patient we repeated a NCS before discharge (D14) which showed improved peripheral motor responses consistent with above finding. No electrodiagnostic correlate of peripheral nerve demyelination was found.

Conclusion

GBS has heterogeneous presentations. Clinicians should have a high clinical suspicion to diagnoses these early to have a better outcome. Preserved or exaggerated DTRs should not exclude the diagnosis of GBS.

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Case Report

Hamman's syndrome (spontaneous pneumomediastinum with subcutaneous emphysema): A rare occurrence in a young male

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Abstract:

Pneumomediastinum (PM) is a rare clinical entity which describes the presence of free air in the anatomical space of mediastinum. Hamman's syndrome, also known as Macklin's syndrome is defined as subcutaneous emphysema with the presence of spontaneous pneumomediastinum (SPM). Although it is commonly described in females in association with labor. we describe a young male presented to our hospital with SPM with subcutaneous emphysema probably the first reported case in Sri Lanka. This unmarried healthy male admitted to our unit with acute onset pain and swelling of the lower part of the face and neck, feeling of neck crepitus, chest pain and difficulty in breathing without haemoptysis, sputum production or fever. Urgent chest Xray (Figure 1) confirmed the subcutaneous emphysema in the previously mentioned areas with radiolucent area in the mediastinum.

Keywords:
Hamman's syndrome

Introduction

Hamman's syndrome was initially described by Hamman in a postpartum female who developed pneumomediastinum with subcutaneous emphysema [1]. He also described the rare Hamman's sign (a.k.a. Hamman's crunch) which is a crunching sound over the precordium synchronous with the heart beats. It is best heard in the left lateral position but it is not always associated with Hamman's syndrome.

This entity is commonly seen in retching in vomiting, aggressive physical activity prolonged labor, forceful coughing from asthmatic bronchospasm or pulmonary infections, inhalational drug use [2]. It usually affects young males and pregnant females and disease course is usually benign and self-limiting.

Case report

23 year old unmarried healthy male admitted to our unit with pain and swelling of the lower part of the face and neck, feeling of neck crepitus, chest pain and difficulty in breathing without haemoptysis or sputum production. He was seemingly well before going for a nap in the afternoon. These symptoms has suddenly appeared when he got up. Chest pain was severe enough to disable him from getting up from the bed but there was no sweating, vomiting, dizziness, palpitation or radiation of the pain. He was brought to our hospital within one hour of the onset of the symptoms. He didn't have a significant past medical history. He denied recent onset fever, cough, vomiting. There was no history of heavy weight lifting, contact sports or trauma. He consumes alcohol occasionally last drink was about one month back. He doesn't smoke and denied inhalational drug use or other illicit drugs. There was no significant family history of similar illness or premature deaths.

He was averagely built (BMI 22.5 kg/m2) dyspnoic and tachypnoic. Lower part the face and neck was swollen with associated crepitus which extends down to the nipple level and right shoulder laterally. Blood pressure 135/85 mmHg pulse rate 110/min. Cardiac auscultation was normal without any additional sounds. Respiratory examination revealed bilateral symmetrical air entry without hyper resonance or added sounds. Sp02 recorded as 98% on room air. Arterial blood gas was normal except for a mild respiratory alkalosis. Urgent chest Xray (Figure 1) confirmed the subcutaneous emphysema in the previously mentioned areas with radiolucent area in the mediastinum. Patient was transferred to the HDU, he was closely observed while giving 2L/min 60% humidified O2. His vitals remained stable. ECG revealed only sinus tachycadia. There were no features of acute coronary event. His lab reports revealed

WBC 6000 x 103 ul-1 ,Neutrophils 76%, Lymphocytes 18%, Hb 13.9g/dL, Platelet 247 x 103 ul-1, CRP 8 mg/L, ESR 15mm/hr. Nasal and throat swab for Covid 19 PCR came Negative. Liver enzymes, renal functions and electrolytes were within normal ranges. Throat swab and sputum culture failed to reveal any pathogenic organisms. HIV I/II antibody was negative. High resolution CT scan of the chest revealed extensive PM with mild subcutaneous emphysema in anterior chest wall and neck. There was no pneumopericardium or pneumothorax. Both lung fields demonstrated mild bronchial thickening and distal centri lobular nodules. There were no consolidation, bronchiectasis, reticulation, honeycombing or mediastinal or hilar lymphadenopathy. His fiber optic bronchoscopy (FOB) and upper gastrointestinal endoscopy (UGIE) failed to reveal any abnomalities. Subcutaneous emphysema gradually resolved over the next two days which was confirmed by the repeat Chest Xray (Figure 2).

Discussion

SPM is believed to be a result of the spontaneous rupture of an alveolus and may also be called "respiratory pneumomediastinum" [3]. The pathophysiology was explained by M. T. Macklin and C. C. Macklin in 1944 [4]. Barotrauma causes alveolar rupture and the air tracks along the bronchovascular connective tissue planes into the mediastinum and hilum. This is also called the "Macklin Effect." Mediastinal air leaks into the subcutaneous tissue, resulting in subcutaneous emphysema. This combination of SPM and subcutaneous emphysema is known as Hamman's syndrome/Macklin's syndrome.

Even though this entity is originally described in Valsalva associated with vaginal deliveries later it has been also reported with retching in vomiting, aggressive physical activity prolonged labor, forceful coughing from asthmatic bronchospasm or pulmonary infections, inhalational drug use [2]. But our patient did not have any of the precipitating factors.

Most of the patients present with retrosternal chest pain, radiating to the back or neck. Dysphonia, dysphagia, and dyspnoea may be present. Extensive subcutaneous emphysema may distort the facial and neck anatomy. Resultant facial and vocal changes may mimic a neurological pathology [5]. Subcutaneous emphysema is the most common sign. Hamman's sign, a precordial crunching sound, synchronous with heartbeat may also be present, and loss of cardiac dullness will be present on percussion. We could not appreciate Hamman's sign in our patient.

Disease course is usually benign and self-limiting. But as Hamman's syndrome is diagnosis of exclusion need to investigate more serious conditions as Boerhaave's syndrome which most of the time require surgical intervention. UGIE of the index case was normal. Other differential diagnosis would be pneumothorax. Clinically it might be difficult identify a small pneumothorax but in an emergency setting chest Xray will exclude its possibility. Acute coronary syndrome, Aortic dissection, pulmonary embolism can present with chest pain and dyspnoea but those are not associated with subcutaneous emphysema.

Conclusion

Hamman's syndrome/ Macklin's syndrome is defined as subcutaneous emphysema with the presence of (SPM). It is usually benign and self-limiting thus management is conservative. But other sinister causes like Boerhaave's syndrome should be excluded first as this is a diagnosis of exclusion.

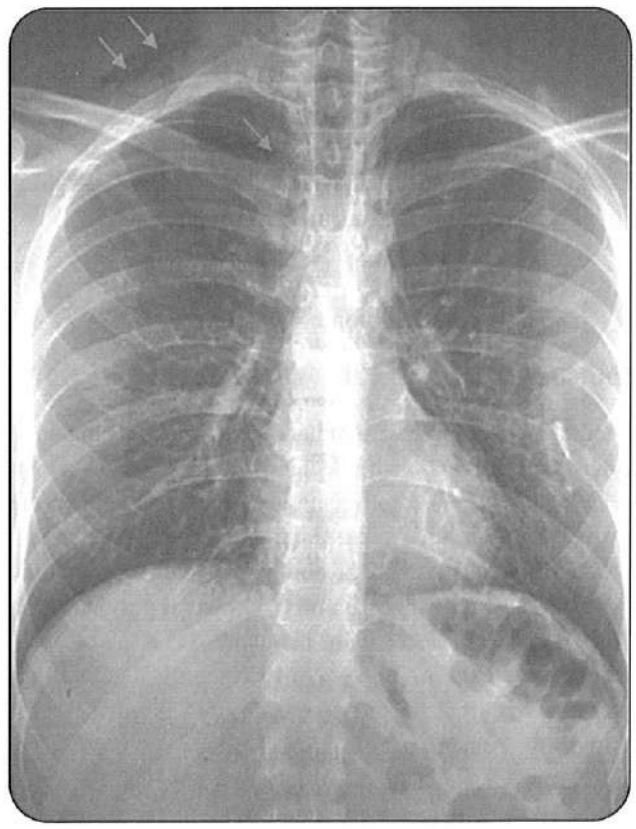

Figure 1 : Red arrows demonstrating subcutaneous emphysema and pneumomediastinum

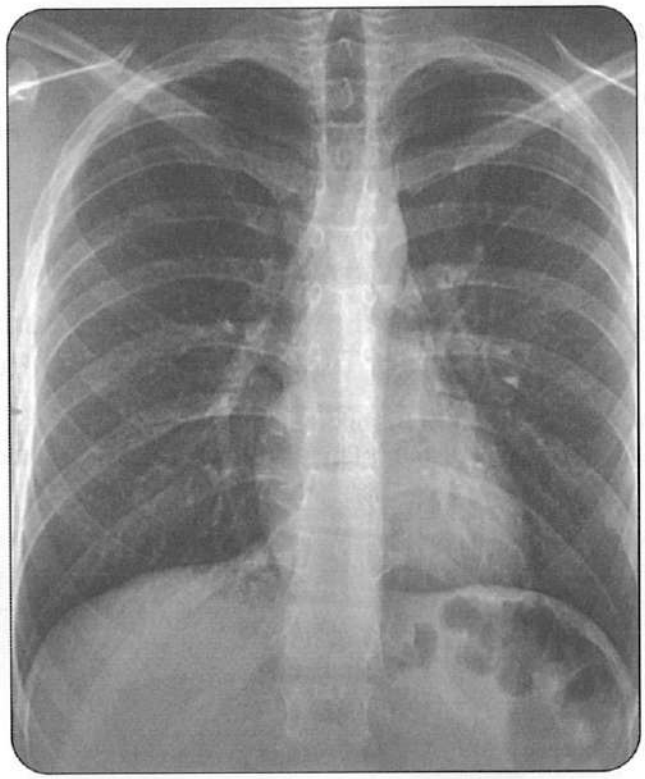

Figure 2 : Complete resolution of emphysema on discharge

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CASE REPORT

Hypothenar wasting as a first clinical manifestation of Superior Pulmonary sulcus (Pancoast) Tumor of the lung: A Case Report

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Abstract:

Lung malignancies are leading cause of the death in worldwide. Here we report a case of 60 year old gentleman presented with hypothenar muscle wasting and loss of weight for 3 months duration found to have Pancoast tumor of the lung.

Keywords: Pancoast tumor, Hypothenar wasting

Introduction

Superior pulmonary sulcus tumors have been applied to neoplasms located at the apical pleuropulmonary groove, adjacent to the subclavian vessels. [1,2] The actual pulmonary sulcus comprises the thoracic costovertebral gutter on either side of the vertebral column and is limited by the arch of the first rib superiorly and the diaphragmatic insertion inferiorly[1].

Due to the peripheral location of the tumor, pulmonary symptoms such as cough, hemoptysis, and dyspnea are uncommon until late in the disease[1]. Lesions in the superior sulcus may result in shoulder and arm pain (in the distribution of the C8, T1, and T2 dermatomes), Horner syndrome, and weakness and atrophy of the muscles of the hand, a constellation of symptoms referred to as Pancoast syndrome or Pancoast-Tobias syndrome [2]. Supraclavicular lymph node enlargement and prominent weight loss are each observed in approximately 25 to 35 percent of cases [2]. Superior sulcus tumors may produce a phrenic or recurrent laryngeal neuropathy or superior vena cava syndrome in 5 to 10 percent of patients[4].

Mainly diagnostic method biopsy and histology and the overwhelming majority of superior sulcus tumors are non-small cell lung cancers (NSCLCs), and in the past were mainly squamous cell carcinomas [2], although in subsequent series adenocarcinomas predominate [5].

Case presentation

62 years old gentleman presented with left hand numbness for three months duration mainly ulnar side little finger and movements not affected and loss of weight for last 3 months. No history of trauma or chronic cough or haemoptysis and no fever. He was known patient of hypertension and ischemic heart disease (Non ST Elevation myocardial Infraction) for eight year duration. He was a manual worker and smoker for last 10 years duration used cigarette 20 packs year.

On arrival he was 45kg BMI was 20kg/m2, clubbing and left side axillary deep lymph node palpable hard in nature nearly 2cmx1cm. on auscultation found left side upper lobe air entry completely nil and dull to purcuse. Upper limb examination found left hand hypothenar wasting (Figure 1 (arrow)) and sensory loss at the medial one third of the finger and C8, T1 dermatome patten and finger power and grip not affected. Other system examinations was normal.

Laboratory investigations reveled Full Blood Count (FBC) haemoglobin 12.4g/dl, WBC 6.7x103 Platelets 220x109, ESR 96mm in 1st hour, chest xray reveled Left side upper lobe opacity (Figure 2). He underwent lymphnode excision biopsy found to have adeno carcinoma (Figure 3), Contrast enhance computer tomography (CECT) of chest reveled 6cmx5cm dense lesion with 1st rib destruction at the left side upper lobe of the lung more favored of pancost tumor of the lung (Figure 4). Bronscopy not done due to definite cause was found with the biopsy and CECT findings.

After that he followed oncology clinic for further care and we reviewed this patient at clinical level after three months he developed sings and symptoms of Horner syndrome such as left side miosis, partial ptosis and hemi facial sweating.

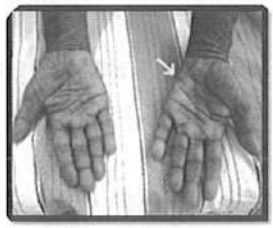

Figure 1

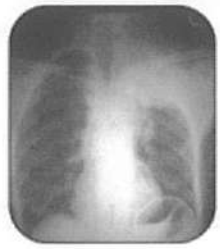

Figure 2

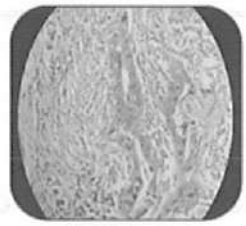

Figure 3

Figure 4

Discussion

Superior sulcus tumors are characterized by a distinct constellation of presenting signs and symptoms due to their location. The most common initial symptom of superior sulcus tumors is shoulder pain, present in 44 to 96 percent of patients [6], neurological complications includes extension of tumor to the C8 and T1 nerve roots results in upper extremity neurologic findings in approximately 8 to 22 percent of cases [4] and sympathetic chain involvement leads to Horner's syndrome. Our patient later the cause of the disease he developed Horner's syndrome.

Involvement of these nerve roots may result in weakness and atrophy of the intrinsic muscles of the hand, or pain and paresthesia of the fourth and fifth digits and the medial aspect of the arm and forearm. Abnormal sensation and pain in the T2 territory (the axilla and medial aspect of the upper arm) may also be an early finding, and the triceps reflex may be lost [4].

Our patient presented only left side hypothenar area numbness and found to have wasting of the hypothenar eminence.

Core needle biopsy is generally preferred to provide tissue for histology and molecular markers. The location of superior sulcus tumors allows the majority to be diagnosed by percutaneous needle biopsy.

Here we didn't undergo biopsy and bronchoscopy due to highly vascular place and tumor already inverted into deepen vascular and nerve plane, and CECT suggested upper lobe tumor inversed and suggested Pancoast tumor. Definite treatment option is surgical intervention when tumor is not invasive and early stages[6]. Here our patient TMN stage is almost T3M2N3 so benefited only chemotherapy.

Conclusion

Elderly patients presented with even muscular skeletal chest pain cannot be neglected and presenting complain of the lung malignancy not always chronic cough but also presented with neurological phenomenon also.

Conflict of interest

None

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Case Report

A Young Brain on Confusion Reported as Anti NMDAR Encephalitis

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Abstract:

Anti N-methyl-D-aspartate receptor (NMDAR) encephalitis is a potentially fatal autoimmune syndrome in which there is profound dysregulation of neurotransmission. It is increasingly being recognized as one of the common causes of encephalitis, but is frequently misdiagnosed. Since the early initiation of immunotherapy gives good recovery, the disorder should be kept as a differential diagnosis in patients presenting with acute onset of behavioral/psychiatric symptoms

Keywords:
Anti-NMDAR encephalitis, Auto immune encephalitis

Introduction

Encephalitis has several etiologies, but in resource-poor settings, most cases are thought to be infectious in etiology. Despite infections being a common cause, auto immune-mediated encephalitis and paraneoplastic encephalitis are increasingly being recognized.

Anti-N-methyl-D-aspartate receptor (anti-NMDAR) encephalitis is autoimmune encephalitis that is mediated by anti-NMDAR antibodies, which is an IgG antibodies directed against the NR1 subunit of the NMDA receptor. These NMDA receptor is located in the forebrain and hippocampus which is play a role in neuro psychiatric manifestation of the patient.

Here we reported a young female presented with acute behavioral chances with seizure episodes diagnosed and treated as anti-NMDAR encephalitis.

Case report

16years old previously healthy female was admitted with the history of abnormal behavior for three weeks duration. Following detail clinical assessment she had visual and auditory hallucination and social withdrawal behavior. She developed fever one day prior to the admission. . She didn't have history of substance abuse or positive family history of psychiatric illness. There were no features of alopecia, skin rash, oral ulcers or history of arthritis to suggest SLE or other connective tissue disease. She didn't have neck stiffness and her haemo dynamics were normal.

Subsequent day she developed GTC fits and her level of consciousness reduced. She underwent extensive blood and imaging studies.

Her initial investigations as follows

Name of test	Values	Name of test	Values
FBC	WBC 16000 N 85%, L 9%, Hb 13.2 platelet 326000	CSF Appearance Total count RBC Polymorphs Lymphocyte Sugar Protein	 Colour less clear 3 cell/ mm 0 % 0% 100% 84mg/dl (RBS 118) 22 mg/dl
ALT AST ALP Alb	75 15 39 35.8	Cuture CSF Blood Urine	No growth No growth No growth
Bu S.Creatinine	50 0.87	NCCT brain	Normal
CPK	154	MRI, MRA brain	Normal
UFR	Normal	CT Abd and Pelvis	Normal
S.Ca S.Mg	8.4mg/dl 1.19	USS Abd	Normal
Na K	144 3.8	EEG	B/L frontal Delta burst activity seen
Covid Ag	negative	NMDAR antibody	Positive

She was initially treated as meningitis with ceftriaxone, acyclovir and dexamethazone. Because of recurrent fits and low level consciousness she was electively intubated and managed in ICU.

With the subsequent investigations and positive NMDAR antibody she was diagnosed and managed as NMDAR encephalitis.

During ICU stay her coarse of illness complicated with Rhabdomyolysis and AKI and vascular catheter associated sepsis and managed accordingly..

Discussion:

This young female presented with three weeks history of abnormal behavior and fever for one dayduration. Based on the initial clinical scenario and presumptive diagnosis of meningo encephalitis made and treatment started accordingly. But the subsequent blood and CSF investigations not favored the diagnosis of meningo encephalitis and warrant to think of alternative diagnosis.

Her disease progressively worsens inspite of antibiotics and complicated with recurrent seizure episode. Her GCS progressively reduced and she was intubated and managed at MICU.

Because of recurrent seizure episode her disease course was complicated with rhabdomyolysis and acute kidney injury.

Imaging of head, chest, abdomen, and pelvis not reveled any abnormalities.

Initial EEG showed diffuse slow wave activity compatible with encephalopathy. Subsequent EEG showed delta burst activity predominantly frontal region.

Diagnostic criteria of anti-NMDA receptor encephalitis

probable anti-NMDA receptor encephalitis
All three criteria must be met: 1. Rapid onset (<3 months) of at least four of the six following major groups of symptoms:¶ • Abnormal (psychiatric) behavior or cognitive dysfunction

- Speech dysfunction
- Seizures
- Movement disorder, dyskinesias, or rigidity/ abnormal postures
- Decreased level of consciousness
- Autonomic dysfunction or central hypoventilation

2. At least one of the following laboratory results:
   - Abnormal EEG (focal or diffuse slow or disorganized activity, epileptic activity, or extreme delta brush)
   - CSF with pleocytosis or oligoclonal bands

3. Reasonable exclusion of other disorders

Definite anti-NMDA receptor encephalitis*

IgG anti-GluN1 antibodies in the presence of one or more of the six major groups of symptoms, after reasonable exclusion of other disorders

Based on the criteria with available investigations, probable diagnosis of anti-NMDA receptor encephalitis made. Patient was treated with IVIG, plasma paresis and prednisolone initially IV and subsequently oral.

Subsequently we received the positive result of NMDAR antibody in CSF from Medical Research Institute and confirmed our diagnosis.

Significant numbers of anti-NMDA receptor encephalitis are associated with ovarian teratoma. But CT abdomen and pelvis of our patient not revealed any tumor.

Treatment of anti-NMDA receptor encephalitis is primarily focused on immune suppression and tumor resection if teratoma is found.

Initially patient was treated with IV methyl prednisone, IVIG and plasma paresis. But patient not shown considerable improvement to this initial treatement. subsequently patient was started second line agent Rituximab.

In the treatment of anti-NMDA receptor encephalitis Rituximab and cyclophospamide are considered as second line agent.

After 10 weeks of inpatient treatment she improved clinically and biochemically. She was transferred to peripheral hospital with the plan of continuation of physiotherapy and oral immunosuppressant

Conclusion

Anti-NMDA receptor encephalitis should be considered in a patient who presented with acute onset of Abnormal (psychiatric) behavior / seizures/ movement disorders with the reasonable exclusion of other causes.

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Case Report

Primary CNS vasculitis; a rare not to miss diagnosis

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Abstract:

Vasculitis is an inflammation of the various sized blood vessels in the body. Among all the types of vasculitis, primary CNS vasculitis is a rare entity. Most frequent presentations are headache, transient ischemic attacks, strokes and cognitive impairments. Diagnosis is basically done by brain biopsy and treated with immunosuppressants. We report a case of 50-year-old male patient presented with a history suggestive of an ischemic stroke ultimately diagnosed to have primary CNS vasculitis. With the treatment of immunosuppressants, patient achieved a complete recovery

Keywords: Vasculitis

Introduction

The vasculitis is a heterogeneous group of disease, which has the potential to cause inflammation in the vessel wall without causing necrosis (1). Vasculitis is principally classified according to size of the vessel which is affected. The other classification is whether vasculitis is the primary disease or it is secondary to other systemic disease. Among all, primary CNS vasculitis is an uncommon entity (2). Even though the commonest presentation is the headache, some people present with strokes, transient ischemic attacks, seizures and cognitive impairments (3,4). Conventional angiography will help in diagnosis but the brain biopsy is considered to be the gold standard (5,6). So physicians should have a high suspicion on primary CNS vasculitis when patients present with multiple uncorrelated neurological symptoms.

Case report

A 50-year-old male patient who was diagnosed to have diabetes, dyslipidemia and ischemic heart disease presented to a local hospital with a complain of sudden onset left side arm, leg and facial weakness. He was managed as acute ischemic stroke. After few days he presented to our hospital with evolving neurological deficits which were severe headache, unsteadiness and visual field impairment. He was a nonalcoholic and a non-smoker. On examination he was conscious and rational. He was having right sided homonymous hemianopia with mild upper motor neuron type facial nerve palsy. Left side upper and lower limb tone was mildly impaired with power of 4/5 in both proximal and distal muscles. Left side all the deep tendon reflexes were diminished and extensor plantar response was present. Right upper limb and lower limb examination was normal. Left sided cerebellar signs were present. No significant sensory impairment was identified. Apart from mild bilateral ankle edema, cardiovascular, respiratory and abdominal examinations were unremarkable.

Full blood count showed mild thrombocytosis which was noted as reactive in blood picture. Serum electrolytes, UFR, renal profile, liver profile, lipid profile and fasting blood sugar were normal. In echo, ejection fraction of 50% and no intra cardiac thrombus noted. Chest radiograph and sinus xray were normal. He was having high HBA1C of 13.7% and ESR of 83. MRI brain showed acute ischemic stroke in the splenium and hemorrhagic sub-acute infarctions in right side temporal lobe, thalamus and left side cerebellar hemisphere. There were some sub-acute ischemic infarctions in frontal subcortical white matter and left occipital lobe. MRA showed diffuse irregularity of the cranial parts of bilateral internal carotid arteries and luminal narrowing in petrus cavenous segment of right internal carotid. Carotid duplex had soft plaque at left carotid bulb without flow limiting stenosis. P-ANCA came as positive and lumbar puncture showed significant elevation of protein with the presence of oligo clonal band. Even though all the investigations were highly suggestive of primary CNS vasculitis as the gold standard we needed to do brain biopsy

but patient refused. Patient was started on induction dose of prednisolone and gradually tailed off to maintenance dose. Then prednisolone was converted to azathioprine maintenance dose. With the beginning of treatment, patient exhibited dramatic improvement in all the neurologi cal impairments.

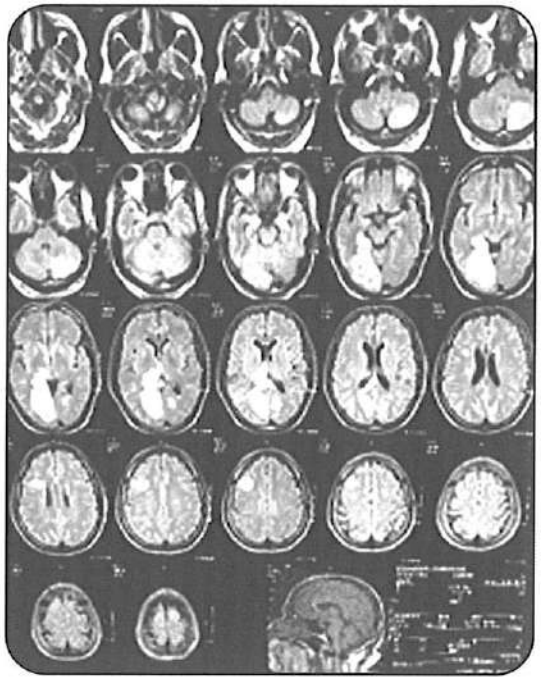

Figure 1. acute, subacute ischemic infarctions and hemorrhagic infarction.

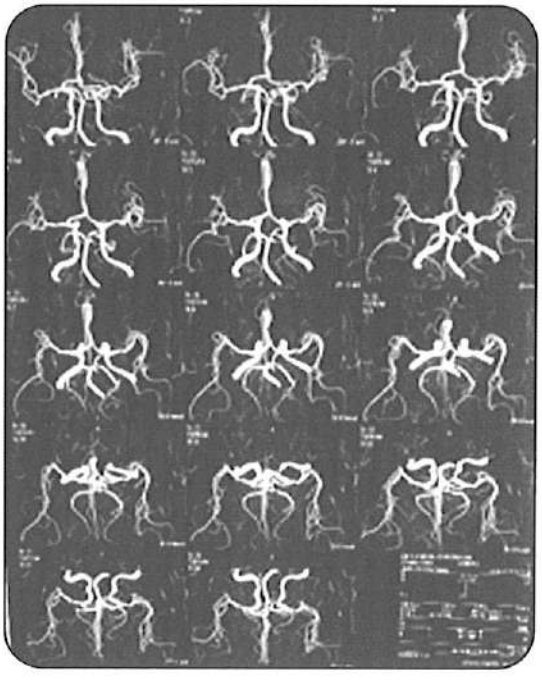

Figure 2. diffuse irregularity of cranial internal carotids and luminal narrowing of petrus cavernous segment of right internal carotid.

Discussion

The vasculitis is a heterogeneous group of disease, which has the potential to cause inflammation in the vessel wall without causing necrosis (1). It happens when one's immune system attacks self-blood vessels. There are various classifications for vasculitis. The commonest one is classification according to the size of the vessel. They are large vessel, medium size vessel and small vessel. The other one is classification as either primary or secondary. Malignancies, infections, medicines and other diseases can cause secondary type of vasculitis. In primary vasculitis, usually the cause is unknown.

Primary CNS vasculitis is an uncommon disorder which causes vessel inflammation in the brain or spinal cord. Patients usually presents in their forties (2). Clinical symptoms are often nonspecific. Commonest presentation is the headache. Other features are strokes, transient ischemic attacks, cognitive dysfunctions, seizures, blurring of vision and ataxia. Usually the patients exhibit combined symptoms (3,4). Commonest mimickers of primary CNS vasculitis are secondary vasculitis, reversible cerebral vasoconstriction syndrome, susac syndrome and neurosarcoidosis (5).

Even though ESR and ANCA not specific for primary CNS vasculitis, it may support the diagnosis of inflammation. In lumbar puncture studies, there will be an elevated protein levels without significant cells. Most often oligo clonal bands will be positive. Conventional angiograms and MRA support the diagnosis (6). Gold standard test is the brain biopsy but it is also not highly specific or sensitive (7). Steroid is the main induction agent in management. In sever disease, other immunosuppressants, mainly cyclophosphamide is combined with steroids. Azathioprine, methotrexate and mycophenolate mofetil are used in maintenance therapy (8,9). Functional assessment is followed up using modified rankin scale. There will be a good long term functional outcome when using combination of a immunosuppressant and a steroid as maintenance therapy (10).

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Case Report

Rhino-Orbito-Cerebral Mucormycosis (ROCM)

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Abstract:

Rhino-orbital-cerebral mucormycosis (ROCM) is a rare aggressive fungal infection with severe morbidity and high mortality caused by a group of saprophytic fungi of the class Phycomicetes. Management is challenging and requires rapid diagnosis, correction of underlying risk factors and aggressive antifungal treatment with surgical debridement.

Keywords:
Rhino-orbital-cerebral mucormycosis

Introduction

Rhino-orbital-cerebral mucormycosis (ROCM) is a rare aggressive fungal infection with severe morbidity and high mortality caused by a group of saprophytic fungi of the class Phycomicetes(1,2,3). This occurs particularly in the immunocompromised such as those with poorly controlled diabetics, hematological malignancies and chronic renal failure etc. Management is challenging and requires rapid diagnosis, correction of underlying risk factors and aggressive antifungal treatment with surgical debridement.

Case Report

A 52-year-old female presented with painful facial swelling to oral and maxillofacial unit Teaching Hospital Karapitiya. She was a diabetic for 18 years and complained of headache, left side infraorbital numbness, nasal congestion and blurred vision of the affected side.

On examination there was facial drooping on the left side, multiple discharging extra oral sinuses, discolored overlying facial skin, obliterated left side nasolabial fold and periorbital oedema. Examination of the left eye showed painful restricted eye movements, partial ptosis and proptosis. There was an intraoral discharging sinus on the palatal aspect of left maxillary first molar with necrosis of adjacent tissues. (Figure 1)

The occipitomental radiograph showed a completely opacified left maxillary sinus and Computer Tomograph showed the extension of involvement of left nasal, paranasal sinuses and orbital cavity. (Figure 2) The laboratory investigations showed 15.200 WBC/mm3 (77.8% neutrophiles, 12.3% lymphocytes, 7.6% monocytes), creatinine 0.4 mg/dl blood, urea 16 mg/dl, glucose level 320 mg /dl.

Clinical, imaging and incisional biopsy findings were in favour of a fungal infection. Initial plan was to surgically explore and debride. Prior to the surgery referrals were made for further ophthalmological assessment and glycaemic control.

Surgical debridement of involved paranasal sinuses was done through both intraoral (Cadwell Luc) and extraoral (Subcillary) approaches under general anaesthesia. (Figure 3) Deep tissue samples were sent for histopathological and fungal studies which confirmed as mucormycosis.

She was put on systemic antifungals; intravenous liposomal Amphotericin B 50mg daily for 45 days. Repeated surgical debridements were done until fungal culture reports became negative. Patient was discharged with oral amphotericin B 500mg for six hourly for 1week. Follow-up clinical examination showed no evidence of fungal disease for last 6 months.

Discussion

Fungal infections of the nasal or sinus mucosa are initiated by inhaling fungal spores. The infection could spread into neighbouring structures such as the orbits, extraocular muscles and optic nerve (1). This could extend to the brain if ethmoidal and orbital veins get involved. There could be necrosis of any involved soft tissue or hard tissue due to the characteristic angio-invasion [(2)].

Mucormycosis that involves the nasal cavity and the paranasal sinus usually arise with signs and symptoms of rhinosinusitis. Once it spreads to the orbit, it causes cellulitis, proptosis, ophthalmoplagia as seen with our patient and eventually may lead to blindness[4]. Headache, confusion, cranial nerve deficits and hemiparesis are indicative of cerebral involvement of the infection[2].

Clinical signs and symptoms are rational from an anatomic basis but the diagnosis of the infection remains a challenge as there are a number of other conditions mimicking the same clinical features. Therefore, it is important to combine the clinical picture with the relevant special investigations. We obtained a multiplaner CT scan views to determine the extension of the infection. Characteristic features that could be noted are mucosal thickening, changes in air-fluid levels and bone destruction in advanced cases. MRI provides a better description of the blood vessel involvement and intracranial extension of the infection [1]. The definitive diagnosis of RCOM can be confirmed by histopathological examination, or by culture on Sabroud's agar of the tissue samples [1].

Because of possible involvement of multiple vital structures in the vicinity the management is challenging. Hence, a multidisciplinary team approach should be undertaken [3]. As RCOM is quite rare in immune competent patients, it is always sensible to check on patients' immune status.

Prompt surgical intervention is often mandatory with debridement and extended resection of all the necrotic tissues as systemic liposomal Amphotericin B is not effective on necrotic tissues[1]. With the use of Amphotericin B it is good practice to monitor renal and liver functions. For refractory cases it has been reported in the literature to use posaconazole, deferasirox plus lipid polyenes, recombinant cytokines granulocyte colony-stimulating factor, granulocyte macrophage colony-stimulating factor, interferon-g and hyperbaric oxygen[5].

Following complete resolution of the infection, reconstructive surgical procedures would be beneficial for most of the cases. Prognosis of RCOM is very much dependent upon the degree of immunosuppression, site of involvement, extent of infection, timeliness of therapy, and form of treatment provided [2].

Conclusion

Key factors in managing RCOM are multidisciplinary approach, reversal of underlying predisposing conditions, surgical debridement followed by systemic antifungal treatment.

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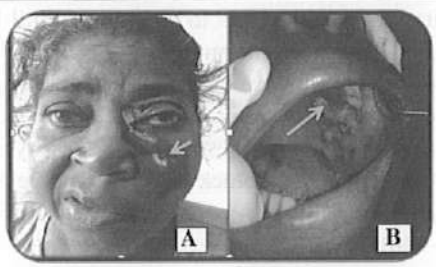

Figure 1; A-extraoral presentation
B-Intraoral presentation

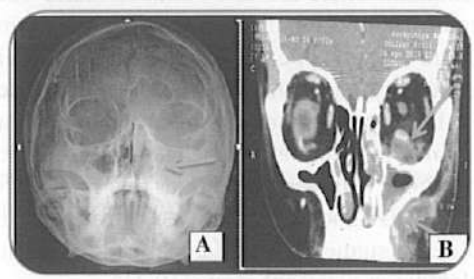

Figure 2; A OM view showing opacification of left maxillary sinus
B CT coronal view showing

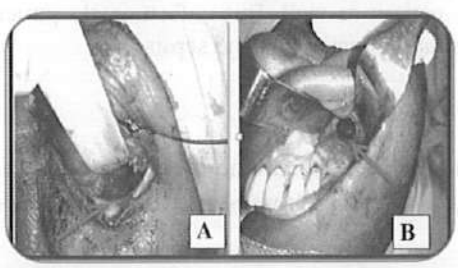

Figure 3; A-Subcilliary approach to left orbit
B-Caldwell Luc approach to left maxillary sinus

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Case Report

Astrazenica COVID vaccine induced Guillain barre syndrome

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Abstract:

Guillain barre syndrome is an immune mediated rapidly progressive polyneuropathy which manifests as ascending symmetrical motor weakness. We report a case of a 87 year old female patient, who is diagnosed to have diabetes presented with ascending symmetrical motor weakness following the Astazenica vaccine. The diagnosis of GBS was confirmed by NCS. After starting of IVIG treatment and plasmapheresis, she showed almost complete recovery from the GBS but unfortunately died from aspiration pneumonia and sepsis. This is probably the first case report in Sri Lanka.

Keywords:
Guillain barre syndrome

Introduction

Guillain barre syndrome (GBS) is an acute, rapidly progressive polyneuropathy which manifests commonly as ascending symmetrical motor weaknesses with diminished deep tendon reflexes. Apart from this common presentation, there are some aberrant clinical manifestations such as asymmetrical weaknesses, preserved deep tendon reflexes, pure sensory manifestations and autonomic instability as the first presentation. There are few variations have been described according to the pathogenesis, demyelinating type (AIDP) and axonal type. In axonal variants, there could be pure motor type (AMAN) or motor sensory mixed type (AMSN). Fisher syndrome is also an another variant of GBS (1-3). GBS is an immune mediated disease which is commonly occured following infections and rarely after vaccinations (4-7). Diagnosis is usually confirmed by the electrophysiological studies (8-9). Main stay of treatments are immunoglobulin or plasmapheresis (10-11).

Case report

A 87 year old female patient who was diagnosed to have diabetes presented to the ward with bilateral lower limb weakness and back pain for 3 days duration. She complained of difficulty in getting up from sitting position. She also complained of upper limb numbness but no obvious weakness. Following day, she developed difficulty in swallowing and breathing. As she went into an impending respiratory arrest, we had to electively intubate the patient and transferred to ICU. She denied any diarrheal or respiratory infections, but she had been given astrazenica vaccine 2 weeks prior to the incident. On examination, bilateral lower limbs were flaccid. Proximal muscle power was 3/5 and distal power was 4/5. Both ankle and knee reflexes were diminished with bilateral flexor planters. Sensory system, upper limb motor system, cranial nerves and cerebellar system were normal. Subsequently patient's upper limb power and neck power were reduced. After intubation patient had few coarse crepitation probably due to aspiration. Full blood count, ESR and CRP were normal initially. Patient had a small hypokalemia and corrected with intravenous potassium. Non contrast brain was normal and nerve conduction study showed abnormalities restricted to sensory and F waves with mild conduction block but preserved velocities. Nerve conduction study was consistent with GBS with AIDP type which was caused by the Astazenica vaccination. She was treated with intra venous immunoglobulin and several cycles of plasmapheresis. She showed dramatic improvement with treatment and we were able to extubate the patient as well. But unfortunately patient died from aspiration pneumonia and sepsis.

Discussion

Guillain barre syndrome is one of the most severe form of acute rapidly progressive paralytic neuropathies. Usual

presentation is symmetrical ascending motor weakness with diminished deep tendon reflexes but there are some atypical presentations as well. They are asymmetrical weaknesses, preserved deep tendon relaxes, pure sensory manifestations and prominent autonomic instabilities. About 20% of GBS patient can have respiratory muscle paralysis. Guillain barre syndrome is mainly divided into four sub types such as, acute inflammatory demyelination polyneuropathy (AIDP), acute motor axonal neuropathy (AMAN), acute motor sensory axonal neuropathy (AMSAN) and fisher syndrome (1-3). As GBS is an immune mediated disease, usually happened following an infection mostly after viral or bacterial. There are some published case reports regarding GBS following SARS COV2. The other main cause for GBS is vaccinations. Even though there are evidences for GBS following influenza vaccinations, there are currently no reported cases of Astrazenica vaccine (4-7).

Guillain barre is a clinical diagnosis and supported by some other investigations which are manly electrophysiological studies and lumbar puncture. In nerve conduction studies we can differentiate either sub types according to the electrophysiological patterns. Lumbar puncture in the second week of the disease shows elevated protein with normal cell counts. But normal CSF protein count doesn't rule out the possibility of GBS. We need to do antibody screening like anti-GM1, GM1b, GQ1b and GD1a ganglioside antibodies to identify the causes for GBS (8-9). If a patient develops respiratory failure, he is needed to be artificially ventilated until the respiratory muscles gain their strength. Definitive treatment consists of intra venous immunoglobulin and plasmapheresis (10-11). About 10% of the patients will deteriorate within 8 weeks of starting immunoglobulin treatment and it is called treatment related fluctuation which is needed another cause of IVIG. Out of all, 5% of patients go into chronic inflammatory demyelinating polyneuropathy in acute onset (A-CIDP). Some will not be able to walk alone even after 6 months of illness. About 3-10% will die from GBS related complications (12).

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Case Report

Dual Cystic Artery in Association with caterpillar Hump

Cheagar S, Piyarisi D L

Abstract:

Moynihan's hump with dual cystic artery is a rare occurrence and there have been single case report in a cadaveric dissection. We report the first case of dual cystic artery in association with caterpillar hump of right hepatic artery in a routine laparoscopic cholecystectomy

Keywords:
Moynihan's hump with dual cystic artery

Introduction:

Moynihan's hump with dual cystic artery is a rare occurrence and there have been single case report in a cadaveric dissection. Laparoscopic Chelecystectomy is a formidable surgery as it requires considerable number of cases over longer period of time to encounter most of its abnormal anatomy for being able to deal with most of the surprises it has to offer, confidently. Here the author want to discuss the dual cystic artery in association with moyniham hump in terms of its surgical anatomy, incidence, and its significance and care in dealing with it intra operatively

Case Report: A 45 year old male patient referred to surgical clinic with history of dyspeptic symptoms for six month duration. Investigations revealed gall stone with no evidence of previous acute cholecystitis. Liver functions were normal. Patient was planned for routine laparoscopic cholecystectomy. Intra operatively found to have extensive omental adhesion to gall bladder and to the region of callots triangle. Dissection was carried out with diathermy hook, Maryland dissector. A gauze swab inside was much useful in the blunt dissection as well as to achieve haemostasis from oozers in the region of callots triangle and to study the region carefully.

Initially it was found to have a large caliber artery within callots triangle. At this point it was believed as it is the cystic artery, as it was abutting the supero medial aspect of the infundibulum of the gall bladder. Fig (b). But the caliber of the artery stopped the operator from proceeding to clipping it. Further detailed dissection with Maryland with close up view was able to separate the part of the infundibulum from the artery and the hump course of the artery came into view. The transverse striations of the artery at close up view reminded the operator the "Caterpillar Hump Sign" of the right hepatic artery. Fig (C). On further dissection two tiny branches were able to be identified from its hump. At this point the finding of two branches from right hepatic artery made further hesitation to go ahead with clipping. The need of demonstrating the two said cystic arteries ending on gall bladder surface was felt and further assistance was sought from consultant.

Decision to stop further dissection and clipping of those tiny short coursed cystic arteries were appreciated as they can be easily avulsed. Subsequently the cystic arteries were clipped with L300 titanium clips and the cystic artery by L400 titanium clips. Rest of the dissection was uneventful.

Discussion: Laparoscopic Chelecystectomy is a formidable surgery as it requires considerable number of cases over longer period of time to encounter most of its abnormal anatomy for being able to deal with most of the surprises it has to offer, confidently. Here the author want to discuss the dual cystic artery in association with moyniham hump in terms of its surgical anatomy, incidence, and its significance and care in dealing with it intra operatively. Secondly want to revisit the attitudes that played in preventing inadvertent damage to the structures in the region of callots triangle with few suggestion of his own.

The usual anatomy is that, the right hepatic artery after its origin from hepatic artery proper crosses anterior to the portal vein and then passes behind the common hepatic duct to enter the Calot's triangle (bounded by cystic duct, common hepatic duct and lower edge of the liver). As it approaches the cystic duct, it gives off the cystic artery and

then turns upwards, behind (and between) the right hepatic and the cystic duct to the right lobe of the liver. The cystic artery normally arising from the right hepatic within the triangle, passes in the triangle towards the neck of gall bladder where it typically divides into two branches one of which runs on the attached surface of the gall bladder and the other on its peritoneal surface [1]. Cystic artery commonly arises from right hepatic artery in 63-92.5% of cases [2-5].

Dual cystic arteries typically represent separate origins of the superficial and deep branches of this artery from right hepatic artery [1]. Sometimes the deep cystic arises from right hepatic and superficial branch from some other source resulting in dual blood supply to gall bladder or both may arise from an anomalous source [1,3]. If the cystic artery that arises from the caterpillar hump is typically short, it may easily avulsed from hepatic artery, if excessive traction is applied to the gall bladder [12]. Here the operator has given it to his immediate supervisor to perform this task as it was proving to be crucial to avoid avulsion of the both cystic artery from Moynihan's hump and to prevent troublesome bleeding.

Tortuous right hepatic artery also called caterpillar hump or Moynihan's hump is rare but dangerous anomaly the incidence of which ranged between 1-12.9% [6-9].The caterpillar hump of right hepatic artery may be mistaken for cystic artery and inadvertently ligated. Thus it is important for the surgeons to appreciate tortuous right hepatic artery forming Moynihan's hump, to dissect it and locate the origin of cystic artery and prevent damage to the right hepatic artery. The presence of "caterpillar hump" should be suspected when an unusually large "cystic artery" is viewed through the laparoscope. [12] This is crucial to avoid excessive bleeding from the injured right hepatic artery during surgery and prevent post-operative complications [11]. Here in this particular case the caliber of the vessel within the callot's triangle made the operator hesitant to go ahead with clipping despite the advocates given by the nursing officers to do so, but also in close up view of a large caliber vessel, the transverse striations reminded the name "caterpillar" as it is almost akin to a caterpillar.

The operator want to revisit the attitudes he had which would have been possibly played part in dealing with decision making. Being able to listen to nursing officers doesn't mean one have to act on it. As well, being brave doesn't mean should not seek senior opinion.

Operator find it useful to have a dry swab to dissect the cHypereosinophilia, Hypereosinophilic Syndrome, Liver lesions, Toxocara, dissector.

Fig (a)

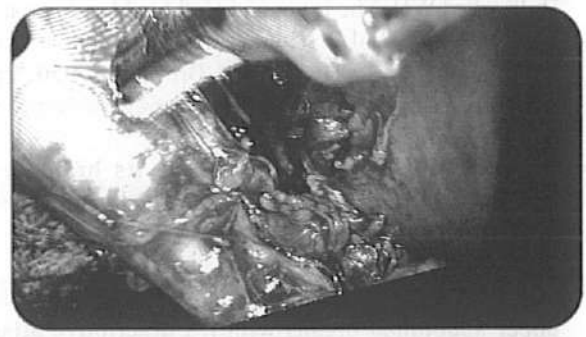


Fig (b)

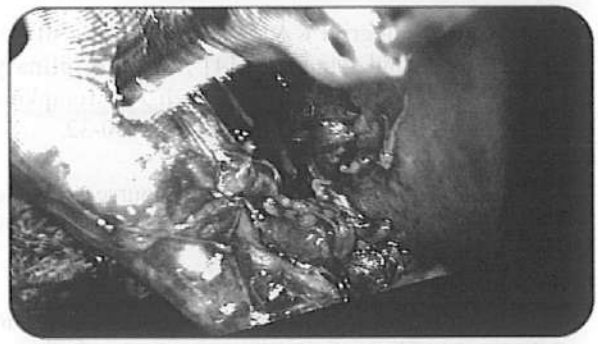


Fig (c)

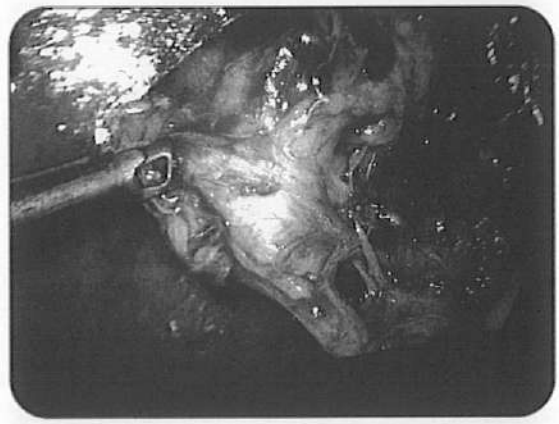
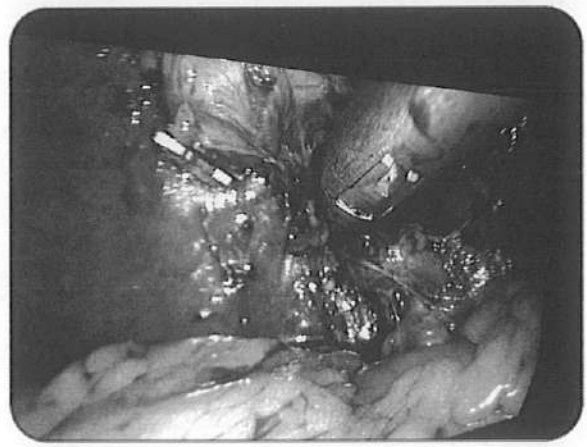


Fig (d)



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Case Report

A child with hypereosinophilia and hypoechoic liver lesions caused by toxocara infection

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Abstract:

Hypereosinophilic Syndrome (HES) is a rare disease that can be difficult to diagnose in children. It can involve the infiltration of various organs by a large number of eosinophils, particularly heart, skin, lung, liver, nervous system, and gastrointestinal tract. The parasitic infection is relatively common in tropical countries as the cause for hypereosinophilia. We report a three-year-old boy with the hypereosinophilia and hypoechoic multiple focal lesions in the liver caused by toxocara infection.

Keywords: Hypereosinophilia, Hypereosinophilic Syndrome, Liver lesions, Toxocara,

Introduction

Hypereosinophilic syndromes (HESs) are a group of rare disorders characterized by peripheral blood eosinophilia of 1.5 × 103/uL or higher and evidence of end organ manifestations in an otherwise healthy child and exclusion of secondary causes1. Severe blood eosinophilia is defined as AEC ≥1.5 × 103/uL and should prompt a thorough evaluation for an underlying cause1,2. The organs which are involved in hypereosinophilia (HE) are heart, skin, lung, liver, nervous system, and gastrointestinal tract. Liver involvement of the HES does occur in 43% patients and several reports have described the findings of hepatic involvement of HES in adults3,4. Sonography shows multiple poorly defined, small, round or oval scattered focal lesions involving the both lobes of the liver. HE due to the parasitic infection is relatively common in tropical countries. Although humans are not definitive hosts of dog roundworm Toxocara canis which transmissions to humans are possible from ingestion of embryonated contaminating soils, hands or fomites1,5. Herein we present a case of HE and multiple poorly defined, hypoechoic liver lesions caused by toxocara infection. As per our best knowledge there are no published data from Sri Lanka on this entity.

Case report

A three-year-old previously healthy boy presented with the history of fever for 3 weeks duration. He had intermittent high fever with chills without any systemic involvement. He frequently plays in ground where there are many street dogs. Otherwise the history revealed no significant information. On physical examination, he was an adequately grown child and had no hepatosplenomegaly or significant lymphadenopathy. The leukocyte count was 28.6 × 103 u/L, with 46% of eosinophils (AEC 19,952) haemoglobin 10.8 g/dL and platelets 424 × 103 u/L. Blood film showed severe eosinophilia without any abnormal cells and evidence of mild iron deficiency anemia. The liver function tests and liver enzymes were normal. The results of a stool test for ova and parasites were normal. The inflammatory markers were normal, urine and blood cultures were sterile. Toxocara IgG antibody was positive and anti-filarial and toxoplasma antibodies were not detected. Sonography of the abdomen revealed multiple ill-defined hypoechoic lesions in the liver. The echo cardiogram was normal. He was treated with oral albendazole 400mg twice a day for seven days to treat toxocara infection. The repeated FBC showed leukocyte count decreased to 424 × 103 with 29% of eosinophils (AEC 5,249) and the liver lesions were completely disappeared after one week of the treatment.

Discussion

HES is defined by the presence of peripheral and bone marrow eosinophilia and the infiltration of multiple organs by mature eosinophilic cells and exclusion of secondary eosinophilia. Clinical manifestations of eosinophilia differ very much between patients2. Secondary eosinophilia is caused by parasite infection, allergic, systemic vasculitis, drugs, and nonmyeloid malignancy. In our case the cause for the HE was the toxocara infection as there was a serological evidence. The organs that are more frequently affected in hypereosinophilic conditions are heart, skin, lung, liver, nervous system, and gastrointestinal tract and in 50-90% of cases there is hepatic involvement2. Abdominal ultrasound shows multiple hypoechoic areas in the liver3. The differential diagnosis to consider are multiple liver abscesses, paracitic infiltration, fungal infections and malignant lesions. In our patient, the liver was involved. Definitive laboratory diagnosis of toxocaral infection can be made by histological examination of various organ specimens, including the liver brain and lung4. Histological examination of the liver biopsy specimen revealed eosinophilic granulomatous inflammation and enzyme-linked immunosorbent assay using Toxocara canis excretory-secretory antigens can confirm the diagnosis5. The liver biopsy was not performed to our patient as the lesions were completely resolved in subsequent sonography after treatment. The best choice for serological diagnosis of the toxocariasis is by the initial use of TES-ELISA (Toxocara canis excretory – secretory antigen), after which any positive result should subsequently be tested by Western blotting (WB) which is not available in Sri Lanka currently[6].

Toxocara canis is a dog round worm and the transmissions to humans are possible from ingestion of embryonated contaminating soils, hands or fomites and ingestion of raw meat1. Epidemiologic studies have shown dog ownership to be a primary risk factor for childhood T. canis infection1(2)(2). Our patient has the close contact with street dogs as he used to play outside frequently. The treatment for toxocara infection is with oral albendazole 10 mg/kg b/w daily for 5 days, and the clinical improvement was found in 47% of patients with toxocariasis3. Our patient showed clinical response to the treatment as evidenced by the reduction in eosinophilic count and the complete resolution of the liver lesions.

Conclusion

Hypereosinophilic syndromes are rare disorder among children and the clinicians should aware of the differential diagnosis. Hepatic involvement is common due to eosinophilic infiltration. Toxocara is a known cause for hypereosinophilia which is treatable with albendazole therapy.

Conflict of Interests

The authors declare that they have no conflict of interests regarding this paper.

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