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Bilateral Parotitis: Unusual initial manifestation of acute lymphoblastic leukemia in otherwise healthy 33 months old child: Case report

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Parotitis or parotid gland enlargement in childhood is commonly due to infections and inflammatory conditions. It is a very rare presentation as a primary neoplasm in paediatric age group. However it is one of the sites of relapses in secondary malignancies. In this report, we present a 33-month old child who presented with bilateral parotitis as a manifestation of B-cell lymphoblastic leukaemia.

Introduction: Parotitis commonly presents in the paediatric age group with infection and inflammatory causes. Parotitis as a presentation of acute leukemia is unusual and very rarely reported. However, low percentage of such presentation is lack of awareness of this unusual presentation as a common paediatric malignancy which should be taken into consideration.

Case presentation: A 33 months old child, with no previous comorbidities, presented to the Emergency department with a 3-month history of bilateral parotid swelling. The swelling was painless and progressively increasing in size

(Figure 1)

Figure 1. Parotid swelling on presentation.

Previously he received multiple courses of antibiotics with no improvement. Fever was present three days prior to his presentation. There was no history of difficulty in swallowing, respiratory distress, ear discharges, eye redness, weight loss, skin rash, gum bleeds, joint pains nor any previous history of similar swellings in the past. In addition, his birth history was unremarkable. He is immunized for his age. On examination, he was sick looking, pale and febrile. There is significant swelling in bilateral infra auricular region, largest is right sided measuring 5*4 cm, firm with mild tenderness,

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lifting ear lobe, no skin changes, with positive findings of bilateral posterior auricular and inguinal lymph nodes. There is neither restriction of neck movement nor difficulty in opening his mouth and mastication. Throat examination revealed mild congestion. No hepatosplenomegaly was noted. His investigations showed: total white blood cell count 7.3*109 cells/L, neutrophils 0.0*109 cells/L, lymphocytes 6.3*109 cells/L, Haemoglobin 5.5 g/dl, platelet count 450*109 cells/L and Creactive protein 27.3 mg/L. The respiratory viral panel including COVID-19 PCR came negative. Chest X-ray: normal, neck ultrasonography reported both parotid glands to appear bulky and heterogeneous in appearance with multiple sub centimetric intra parotid lymph nodes and increased vascularity with cervical lymphadenopathy (Figure 2). Child was admitted with initial impression of right sided parotitis and febrile neutropenia with anemia. On 1st Day of admission, as per febrile neutropenia protocol, intravenous Tazocin was started empirically and transfusion with packed red blood cells commenced. Blood and urine culture reported no growth, repeat full blood count post transfusion showed Hb 9.5 g/dL, WBC: 3.9*109 cells/L. Blood film features were suggestive of reactive changes to infection or inflammation with marked neutropenia 0.2*109 cells/L and anemia, repeated twice, no blasts seen. Haemolytic screen and rest of the anemia workup was normal. He was admitted for total of 7 days. Paediatric haematologist was continuously updated. Upon completion of Tazocin for total of 7 days, parotid swelling significantly improved. He was then discharged with Cefdinir for 7 days with a total 14 days of antibiotics. Child was brought by parents after 1 week to paediatric clinic for follow up. One day prior to the visit, mother noticed the swelling recur which progressively increased in size, no redness noticed, no pain and child remained afebrile with good activity and fair oral intake. Repeat full blood count showed Hb of 8.5 g/dL, neutrophils 0.1*109 cells/L. Viral work up: Mumps IgG +ve, -ve IgM CMV IgG +ve, IgM intermediate. EBV, HIV and hepatitis screening were negative. A Figure 1 Parotid swelling on presentation additional 7 days of Cefdinir was advised. Red flags explained and parents were advised to report immediately to emergency in case of worsening symptoms. Child was seen again after 2 days in our clinic with pallor. Repeat investigations showed Hb of 6.5 g/dL, neutrophils 0.0*109 cells/L. Examination also revealed recurrence of swelling in the right infra-auricular region around 3*3 cm with multiple posterior cervical lymph nodes with no tenderness and skin changes. He was admitted to the ward for blood transfusion. Meanwhile bed was arranged in haematology oncology center for further investigations and shifted the next day. Investigation post transfusion done in oncology centre showed Hemoglobin 8.7 g/dl, platelet 363*109 cells/L, white blood cells 2.9*109 cells/L, neutrophils of 0.0*109 cells/L, lymphocytes 2.8*109 cells/L. Blood film confirmed mild microcytic hypochromic with severe neutropenia, few activated lymphocytes seen. Ultrasound abdomen showed: Hepatomegaly with coarse echo texture. Neck ultrasound: both parotid glands markedly enlarged with heterogeneous echogenicity and multiple

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hypoechoic foci and increased vascularity in colour doppler in keeping with acute parotitis. Both submandibular glands are enlarged with multiple enlarged cervical lymph nodes mostly in all level, the largest lymph node seen in submandibular regions, in right side measures 3.8*1.8 cm and in left side measures 2.3*1.4 cm no collection seen. He underwent bone marrow aspiration which reported the trilineage haematopoiesis as markedly reduced and replaced by blasts at 80% of the total nucleated cells. Immuno-phenotyping of the bone marrow is consistent with B acute lymphoblastic leukaemia. Fine needle aspiration cytology of parotid glands showed infiltration by lymphoblasts. The child was started on the Children's Oncology Group ALL protocol and parotid swelling subsided within 72 h of starting the steroids. The bone marrow study for minimal residual disease done at the end of induction period was negative. The child is currently in consolidation phase of chemotherapy and doing well.

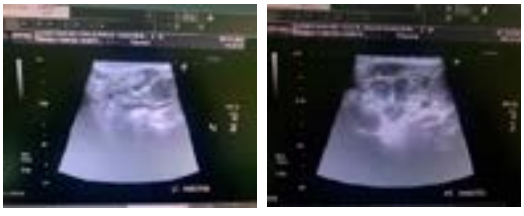

Figure 2. Left parotid gland, right parotid gland.

Discussion: The parotid gland is the largest salivary gland located in the retromandibular fossa, just anterior to the ear and sternocleidomastoid muscle. The commonest causes of bilateral parotitis in paediatric age group are infective aetiology, then inflammatory causes. However, mumps is the most common, followed by *Staphylococcus aureus, Cytomegalovirus, Epstein–Barr virus, coxsackievirus*, tuberculosis and then human immunodeficiency virus. In children with HIV, salivary gland involvement recognized as firm, non-tender and usually asymptomatic. Parotid neoplasm is rarely found in paediatric age group. Bilateral parotidomegaly is rarely reported as a presenting manifestation of acute leukaemia. However, there are a few reported cases of bilateral parotid gland enlargement as a presenting manifestation of relapsed pediatric acute myeloid leukemia as well as L-asparaginase induced acute parotitis during intensification therapy of acute lymphoblastic leukemia. Akanksha Garg, reported a 10-year-old girl of acute myelomonocytic leukaemia with normal cytogenetics, after completion of first consolidation of the BFM 2004 AML protocol, presented with bilateral parotid gland enlargement. Bone marrow examination was suggestive of a relapse. Fine needle aspiration of the parotid gland showed presence of myeloblasts. Kathwate J, reported a 7-year-old boy with ALL who presented with fever and bilateral painful parotid enlargement during intensification phase therapy. Ultrasonography of parotids revealed enlarged, hypoechoic and hyperaemic glands with few enlarged lymph nodes, after discounting L-ASP parotitis resolved within a week 3. Review of literature showed no other drugs except L-ASP can cause parotitis. In Addition,

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a fifteen-month old child who presented with fever and bilateral parotid gland enlargement of 10 days duration, examination revealed generalized lymphadenopathy, hepatosplenomegaly, Investigations confirmed CD 10+ B-cell Acute Lymphoblastic Leukaemia (ALL). Fine needle aspiration cytology of parotid glands showed infiltration by lymphoblasts, reported by Vikas Agarwal which is the same findings of our case. Four cases reported by Kulkarni, with parotid enlargement as initial presentation of acute leukaemia; one of them had bilateral parotidomegaly others were unilateral; However, all the four children had hepatosplenomegaly and lymphadenopathy with no CNS involvement. Reported one child had a combined bone marrow and CNS relapse after 18 months of treatment. It has been noticed a delay in diagnosis of acute leukaemia for more than one month once the child presented with parotid swelling as the initial manifestation. Naithani and Mahapatra and Saha et al. reported a similar case, biopsy of the parotid gland done to confirm tumour infiltration same what has been done in our case although peripheral smear showed no blasts and bone marrow aspirate was conclusive of ALL. In addition, Naithani and Mahapatra also reported involvement of facial nerve with parotid enlargement as an initial manifestation of acute leukemia all the reported cases till date who presented with parotidomegaly showed complete resolution of the parotid enlargement within 72 hours of starting the steroids and the same thing occurred with our case upon follow up. In Oman, there is no similar case reported till now, however only one case report of unilateral proptosis as a rare presentation of AML in a six-year-old girl.

Conclusion:

Parotitis is one of the common presentations seen in paediatric age group with infectious and inflammatory aetiology. In Addition, Parotitis as a presentation of acute leukaemia is unusual and very rarely reported. However low percentage of such presentation should be taken into consideration and physicians should be aware of this unusual presentation of a common paediatric malignancy.

Biography

Anood Al Rawahi is a pediatrics physician at Department of Pediatrics at Armed Forces Hospital.

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An audit to assess quality of neonatal chest radiographs in a district general hospital compared to European commission guidelines

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Background:

Neonatal chest radiography is a common diagnostic examination, especially in preterm neonates where anatomical as well as biochemical immaturity impacts the synthesis and secretion of surfactant. Quality of neonatal chest radiography varies significantly, with concerns regarding increased radiation dose due to failure to appreciate neonatal anatomical proportions. High mitotic capacity in neonates increase the risk of developing cancer by 2 to 3 times when exposed to radiation compared to adults. Hence, the main aim of this completed audit cycle was to assess the baseline of adherence to the standards of neonatal chest radiograph imaging and thereby carrying out an intervention to implement changes that would improve the quality of the radiographs to minimize unnecessary radiation to this age group.

Methods:

A pre-intervention retrospective audit was conducted over 1-year period in 2020 selecting 100 neonatal chest radiographs, in AP view and supine position, in a neonatal ICU in Medway NHS Foundation Trust. Six parameters from the European Commission Guidelines (1996) were used to assess the adequacy of chest radiographs, with an aim in reducing unnecessary radiation dosage. The parameters used were inspiration, rotation, craniocaudal collimation boundary, transverse collimation boundary, head position and arm position. An intervention was made via departmental presentation and neonatal grand-round presentation with recommended changes published in the trust intranet that was available to view for all the staffs in the hospital. After 6 months a reaudit was performed including neonatal chest radiographs over 2 months that were assessed using the same parameters.

Results:

Significant improvements were noted with reduction in upper thorax rotation by 42.9% (from 49% to 28%) and reduction in lower thorax rotation by 48.9% (from 47% to 24%). Head position was straight in 74% compared to pre-intervention 57%. Cranial collimation boundary improved by 20% and caudal collimation boundary improved by 47.8%. Improvements were required in inspiration, transverse collimation boundary and arm position.

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Conclusion:

Implementation of European Guidelines has shown to improve the quality of neonatal chest radiographs. Significant improvements in the craniocaudal collimation and head position along with reduced rotation led to reduced exposure to radiation in neonates in the hospital. Further need of intervention has been acknowledged to reduce radiation dose by defining surface marking for collimation boundaries due to the anatomical differences. The hospital is planning to incorporate this in the departmental Standard Operating Procedure.

Biography

Nitish Raj has completed his MBBS from Tribhuvan University from Nepal. He has been academically active since his medical school showing outstanding performance. He has been an integral part of organising Basic Science Olympiad in Nepalese Army Institute of Health Sciences from 2012 to 2019 as Student Ambassador for Elsevier. After registration with the General Medical Council of the United Kingdom, he has served the prestigious NHS with clinical experience in Trauma and Orthoapedics as well as General Surgical Department. He has a special interest to develop a career in Trauma radiology and Intervention Radiology.

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An infant with persistent high-grade fever: Having a low threshold for suspecting and treating atypical Kawasaki disease

Roshan Bharani*, Shaju Edavana, and Priyavarshini Ramesh
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Summary:

We report a previously healthy 8-month-old infant who presented with a 6-day history of high-grade fever. Initial clinical examination revealed no fever, but two classical features of Kawasaki disease were observed in bilateral conjunctivitis and polymorphous rash. Initial investigations revealed anaemia, and high CRP, white cells, platelets and neutrophil count. As chest x-ray showed right mid-zone consolidation, he was treated for a chest infection. However, he was still pyrexial with high inflammatory markers after a 5-day course of intravenous antibiotics. While he did not fulfill the criteria for classic Kawasaki disease, a diagnosis of atypical Kawasaki disease was considered. After discussion with Rheumatology, he was treated as atypical Kawasaki disease with IV immunoglobulin and high-dose aspirin. The case identifies how an important diagnosis can manifest differently and needs a high index of suspicion to make this diagnosis in a timely fashion to prevent serious sequelae.

Background:

Patients with atypical or incomplete Kawasaki Disease (KD) do not fulfill all of the diagnostic criteria of classic KD. For a diagnosis of atypical KD, the patient must have:

• Fever for 5 or more days.

• Two of five clinical features for typical KD (which comprises bilateral non-purulent conjunctivitis, changes in upper respiratory tract mucous membranes such as strawberry tongue, erythema/oedema/desquamation of hands and feet, polymorphous erythematous rash on trunk and limbs and cervical lymphadenopathy>15 mm).

• C-reactive protein (CRP) greater than 3.0 mg/L and/or Erythrocyte Sedimentation Rate (ESR) greater than 40 mm/h.

• Compatible laboratory findings (at least 3 of the following: albumin, ≤ 3.0 g/dL; anaemia for age; elevation of alanine aminotransferase; platelets ≥ 450,000 µL after seventh day of illness; white blood cell count ≥ 15,000 µL; urine ≥ 10 white blood cells/high-powered field).

• Positive echocardiography findings for coronary artery aneurysms/dilatation.

• No other reasonable explanation for illness.

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Since there is no singular sensitive test to confirm KD, it is challenging to establish a definitive diagnosis quickly, and failure to treat early often lead to coronary artery complications occurring in children with KD. In the developed world, KD is the biggest cause of acquired heart disease in children. The mainstay of treatment of KD remains Intravenous Immunoglobulin (IVIG) infusion within 10 days of onset of the disease. IVIG can decrease the risk of coronary artery abnormalities from 25% to 5%. A detailed history and physical examination are required to reveal the cause of other features of KD, but atypical KD should still be considered when all classical features of KD are not present.

Case Presentation:

An 8-month-old White Eastern European male was referred to Paediatrics with a 6-day history of persistent high-grade fever (39-40˚C) and 2-day history of bilateral non-purulent conjunctivitis and widespread, maculopapular, non-urticarial blanching rash. There was no cough or coryzal symptoms, or history of recent illness in the household. Two days before admission, he was started on oral amoxicillin in primary care.

The patient was born by normal vaginal delivery at 41+2 weeks gestation with a birth weight of 3.4 kg. He had remained fit and well in the past and did not take any regular medications. He was not known to have any allergies. His immunizations were up to date.

On examination, the patient was alert but irritable. He showed no symptoms or signs of respiratory pathology. His temperature was settled at 37.8˚C with Paracetamol. He had tachycardia (heart rate 150 beats per minute) and good peripheral perfusion. Anterior fontanelle was noted to be soft. He had a widespread maculopapular blanching rash on torso, arms and legs. He did not have reactivation of his BCG scar. His throat was mildly congested with pus spots on tonsils. He did not have cervical lymphadenopathy.

Investigations:

Initial Full Blood Count (FBC) revealed a haemoglobin concentration of 90 g/dL, a Mean Corpuscular Volume (MCV) of 74.3 fL and a White Cell Count (WBC) of 12.7*109 /L with 6.2*109 neutrophils, and 4.4*109 lymphocytes. The platelet count was initially 458*109/L and CRP was elevated at 101 mg/L. Liver function tests revealed a total bilirubin of 2.9 mg/dL, aspartate aminotransferase of 17 U/L, alkaline phosphatase of 148 U/L, total protein of 68 g/dL, albumin of 39 g/dL and globulin of 29 g/dL. Urinary microscopy showed normal white cell count. Urine and blood cultures did not grow pathogens. Chest x-ray showed a right mid-zone consolidation.

Echocardiography showed normal appearance of aortic arch and proximal coronary arteries with no evidence of atrioventricular valve regurgitation or pericardial effusion. Biventricular function was normal and right and left ventricular outflow tracts had unobstructed flow.

The CRP increased further during admission to 121 mg/L, while WBC count increased to 16.5*109, platelets count rose to 774*109 and ESR was 37. Fibrinogen was elevated at 7.10 g/dL, and troponin was elevated at 5.8 ug/L.

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Differential Diagnosis:

As this patient presented during the COVID-19 pandemic, Paediatric Inflammatory Multisystem Syndrome Temporally associated with SARS-CoV-2 (PIMS-TS) was ruled out based on clinical and biochemical criteria. He was covered for potential Sepsis till urine and blood cultures were reported to be negative. Further to the chest x-ray demonstrating a right mid-zone consolidation, causes of atypical pneumonia were also considered. Respiratory viral PCR sample and nasopharyngeal PCR sample were both negative for respiratory viruses. Cytomegalovirus and Epstein-Barr virus screen was negative. Antistreptolysin-O (ASOT) was below 200 ruling out recent group A streptococcus infection. Therefore, in the absence of any other plausible cause of illness, the patient was treated for Atypical KD.

Treatment:

The patient was given Intravenous Immunoglobulin Infusion (IVIG) in a dose of 2 g/kg and high-dose oral aspirin 12.5 mg/kg on his fifth day of admission and eleventh day of illness. He also completed a course of intravenous antibiotics and oral antibiotics during inpatient admission.

Outcome and Follow-up:

Following IVIG administration, the patient improved both clinically and biochemically and was not noted to be irritable. He went on to develop desquamation of his toes and fingers. He was discharged after 9 days of admission when his CRP had decreased to 22 and he had remained afebrile for 24 hours. His parents were advised to contact Paediatric Assessment Unit if he spikes fever within 48 hours of discharge. A local clinic follow up was arranged and a formal referral to tertiary Cardiology was made for urgent outpatient review.

Discussion:

While the exact cause of KD remains unclear, it is characterized by largely self-limiting systemic inflammation of medium-sized arteries. However, it constitutes a diagnostic conundrum as its principal presentation can mimic several other diseases, including measles, scarlet fever, and juvenile idiopathic arthritis. Current NICE guidelines recommend having a low threshold for considering KD especially in infants younger than 1 year, who often do not fulfill the criteria for KD at presentation and hence have delayed diagnosis and treatment, putting them at higher risk of experiencing coronary artery complications.6 Furthermore, principle features of KD, as shown in this case, may appear and disappear at different points in the clinical course of the disease and maybe subject to factors such as parental recall and clinician subjectivity. As such, it is recommended that diagnosis of KD should be considered in all cases of fever lasting 5 days or more; the presence of cardinal KD features make the diagnosis more likely but their absence should not rule out KD.

Learning points/take home messages:

- Consider KD in infants with prolonged fever, irritability and unexplained or culture-negative shock/meningitis/lymphadenitis, unresponsive to antibiotics.
- Suspicion of KD should involve early discussion with Rheumatology and Cardiology colleagues to initiate IV immunoglobulin and high-dose aspirin.
- IV immunoglobulin is most effective when given within first 10 days of illness, and reduces risk of coronary artery complications from 25% to 5%.

Biography

Roshan Bharani is working as a ST4 Paediatric trainee at Ipswich NHS Foundation Trust (NHS East Suffolk and North Essex).

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Prevalence of vitamin D deficiency in pregnant women and their babies in Bhaktapur, Nepal

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Oxford University Clinical Research Unit, Nepal

Background:

Vitamin D deficiency has been observed worldwide in pregnant women and their newborns. Maternal vitamin D deficiency can lead to deficiency in their newborn baby and has been linked with various complications during pregnancy and delivery. There is risk of premature delivery and it is associated with high neonatal mortality.

Methods:

Seventy-nine pregnant women who were admitted to the Siddhi Memorial Hospital for delivery and their newborn babies were enrolled in the study. Maternal blood samples were taken before delivery while umbilical cord blood samples of their babies were taken after delivery. Serum vitamin D level and calcium level were assessed by fluorescence immunoassay using Ichromax vitamin D kit and endpoint method, respectively in the Siddhi Memorial Hospital laboratory.

Results:

Mean +/− SD serum vitamin D and calcium levels in pregnant mother before delivery were 14.6 +/− 8.5 ng/ml and 8.0 +/− 0.5 mg/dl, respectively, and in the cord blood were 25.7 +/− 11.2 ng/ml and 8.6 +/− 0.9 mg/dl, respectively. Eighty-one percent of the mothers and 35.8% of their babies were found to have vitamin D deficiency. Although 97.5% of the pregnant women were taking calcium supplementation, serum calcium was found lower than the normal reference value in 67% of the pregnant women and 64.2% of their babies. There were a linear relationship between the maternal and baby's serum vitamin D ($P<0.001$) and calcium ($P<0.001$) levels.

Conclusion:

There is high prevalence of vitamin D and calcium deficiency in pregnant mothers and newborn babies in Bhaktapur, Nepal. Pregnant women need to be supplemented with adequate amounts of these nutrients.

Biography

Saraswati Budhathoki has done her Bachelor Nursing and working at Oxford University clinical research Unit in Nepal.

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Seed bezoars as a rare cause of fecal impaction in pediatric patients: A report of two cases from Pakistan

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Large Bowel obstruction can occur due to numerous etiologies, with seed bezoars being an infrequent one. We herein present two cases of fecal impaction by rectal seed bezoars in pediatric patients for the first time from Pakistan, while also reporting areca nut (betel nut) fecal impaction for the first time in the literature. Our first patient was a 6 year old boy who, having consumed a bag of sunflower seeds, presented to the emergency department with the complaints of constipation and abdominal pain. Under sedation, the seeds were manually evacuated. Following that he continued to pass stool mixed with seeds over next 24 hours. His symptoms resolved and was discharged home next day. Our second patient was a 10 year old boy who presented to the emergency department with the complaints of pain in anal region and constipation after ingestion of areca nuts. Under general anesthesia, manual dis-impaction was done to evacuate the nuts. A mucosal tear along with small amount of bleeding was also observed, which resolved with anal packing for 6 hours. In both cases, counselling of parents was done to prevent such an incident in the future.

These cases highlight severe sequelae of ingestion of seeds/nuts by children, which is the formation of seed bezoars and intestinal obstruction, possibly leading to constipation and other possibly dangerous complications. Although manual dis-Impaction under general anesthesia almost always treats these patients, it is equally crucial to create awareness regarding the dangers of consumption of these items, especially in developing countries.

Biography

Shajie Ur Rehman Usmani is a 5th year MBBS student from Pakistan with an immense passion for Pediatric Cardiac Surgery, which he plans to pursue as a career. He is an active social worker and a Global Surgery enthusiast with a keen interest in promoting Universal Health Coverage for surgery in particular. Moreover, he is an avid public speaker and a leader-in-making who believes in the power of advocacy, especially for pediatric health care.

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