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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.

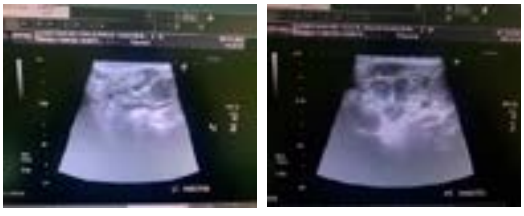

Figure 2. Left parotid gland, right parotid gland.

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.