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Case Report

A case of juvenile recurrent parotitis with thrombocytopenia

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ABSTRACT

Juvenile recurrent parotitis is defined as recurrent inflammatory parotitis in children of unknown etiology. Juvenile recurrent parotitis is the second most common cause of parotitis in childhood, only after mumps. It is a rare disease characterised by recurrent parotid inflammation associated with non-obstructive sialectasis. Here we present a 10-year-old boy with 6 recurrent episodes. The child came with complaints of bilateral swelling below the ear, fever and malaise for 1 week. He had recurrent episodes of painful swelling below both the ears for the past 2 years. He previously had 5 episodes which was unilateral associated with pain and fever, lasting for 7–10 days and treated with analgesics and antibiotics. During these episodes he had no dryness of mouth and eyes, increased salivation or altered taste sensation, joint pains/swelling, weight loss, night sweats and skin rashes. There is no pain during eating and no family history. Lab findings reveal thrombocytopenia during each episode and resolved spontaneously. Relevant investigations ruled out Systemic Lupus Erythematous, Sjogren syndrome, Tuberculosis, Infectious mononucleosis and immunodeficiencies. FNAC reveals granulomatous sialadenitis. Bone marrow examination shows normocellular marrow with increase in megakaryocytes. Serum ACE and IgG4 levels were normal. IgG levels were elevated. Child was started on oral steroids and improved clinically. This case depicts a rare association of recurrent parotitis with thrombocytopenia.

Keywords: Recurrent, Parotitis, Thrombocytopenia

INTRODUCTION

Recurrent juvenile parotitis is an idiopathic condition which usually begins between 3 and 6 years of age with male predominance. It is a rare, recurrent non obstructive and non-suppurative parotid inflammation having a multifactorial etiology. Diagnosis should be made after ruling out Sjogren, lymphoma and immunodeficiencies. In most cases symptoms resolves spontaneously by puberty.

Juvenile recurrent parotitis is defined as recurrent inflammatory parotitis in children of unknown etiology.[1] Juvenile recurrent parotitis is the second most common cause of parotitis in childhood, only after mumps.[1] It is a rare disease characterized by recurrent parotid inflammation associated with non-obstructive sialectasis.[2] Here we present a 10-year-old boy with 6 recurrent episodes.

CASE REPORT

A 10-year-old male child came with complaints of bilateral swelling below the ear, fever and malaise for 1 week. He had recurrent episodes of painful swelling below the ears for the past 2 years. He previously had 5 episodes which was unilateral associated with pain and fever, lasting for 7–10 days and treated with analgesics and antibiotics. During these episodes he had no dryness of mouth and eyes, increased salivation or altered taste sensation, joint pains/ swelling, weight loss, night sweats and skin rashes. It is aggravated due to dehydration and

cold weather. There is no pain during eating and no family history of parotid swelling. He has mouth breathing while sleeping for 1 year. Physical examination reveals Bilateral tender firm parotid swelling with bilateral multiple non tender cervical lymphadenopathy. His systemic examination was unremarkable. Initial possibilities of sjogren syndrome, sarcoidosis was considered.

Laboratory investigation reveals concurrent thrombocytopenia (<0.9 lakhs/cu.mm) and resolves with episodes. USG shows multiple hypoechoic areas in the parotid. CT scan of neck shows diffuse enlargement of both parotid glands with multiple tiny hypoechoic areas scattered in both superficial and deep lobes and enlarged upper deep cervical lymph nodes bilaterally on upper (Level-II) and middle (Level-III) jugular group, suggestive of autoimmune or infective etiology. Autoimmune markers (ANA, Anti-SSA, Anti-SSB) and rheumatoid factor are negative.

Serology for Epstein-Barr virus is negative. Inflammatory markers (ESR, CRP), serum immunoglobulins (IgG, IgM, IgA) and complement levels (C3, C4) are in normal range. PCR for tuberculosis is negative. Fine Needle Aspiration Cytology (FNAC) of parotid gland revealed sheets of lymphocytes, histiocytes, and occasional epithelioid-like cells admixed with neutrophils, infiltrating and destroying glandular structures, suggestive of granulomatous sialadenitis. In view of recurrent thrombocytopenia, bone marrow was done and shows normocellular marrow with mild increase in megakaryocytes. Serum IgG levels were elevated. Serum ACE levels and IGG4 levels were normal. Child was started on oral steroids and on follow up.

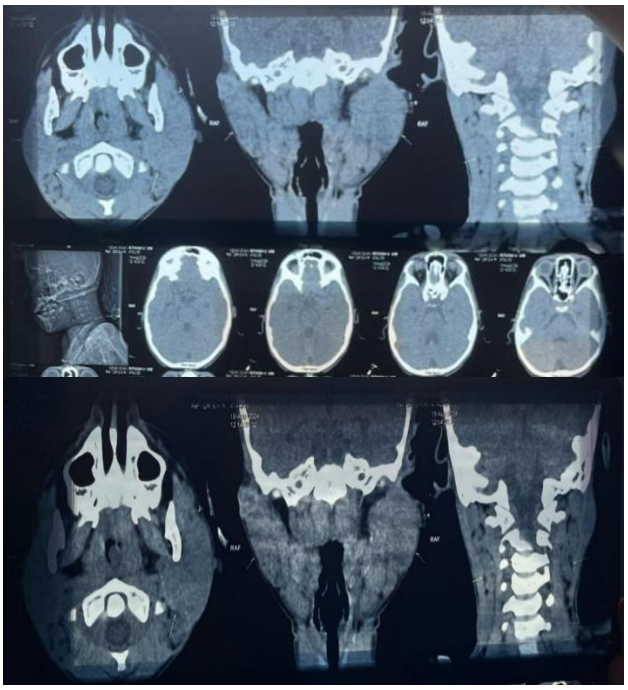

Figure 1: Adiographic pictures.

DISCUSSION

Recurrent juvenile parotitis is an idiopathic condition which usually begins between 3 and 6 years of age with male predominance.[1,2] The number of attacks changes from 1 to 5 years.[2] It is a rare, recurrent non obstructive and non-suppurative parotid inflammation having a multifactorial etiology. Many causative factors such as allergy, infection, local autoimmune factors and genetic inheritance have been suggested, but none have been proved.[3,4]

Clinical symptoms of juvenile recurrent parotitis include recurrent parotid swelling and pain associated with fever which usually lasts 2 to 7 days. The pathology is usually unilateral but can be bilateral with symptoms predominant on one side.[1] In 80-90% cases, symptoms resolve spontaneously by puberty.[5] The histological features of juvenile recurrent parotitis include sialectasis of peripheral ducts in the parotid gland with periductal lymphocytic infiltration.

Congenital ectasia of the ducts and ascending infection from the mouth have been postulated to explain the pathogenesis.[1] Sialography is commonly used for diagnosis, but ultrasound superseded this procedure.[6] Initial treatments is conservative since 90% has spontaneous remission. Analgesics, warmth, attention to oral hygiene and massage of the parotid gland are helpful.[5] Antibiotic usages is often proposed to prevent further parenchymal damage. More aggressive treatments like parotid duct ligation and parotidectomy are reserved for those with persistent symptoms.[7]

CONCLUSION

Though recurrent juvenile parotitis is a rare condition in childhood, morbid recurrences are common. Hence, parents should be counselled about the benign course of the disease and its resolution by puberty. Short course of steroids gives clinical response in some patients.

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