

Case Report

Juvenile Recurrent Parotitis: A Case Report

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Abstract: Juvenile recurrent parotitis (JRP) is a rare recurrent nonobstructive, nonsuppurative parotid gland inflammation in young children with a multifactorial etiology characterized by multiple episodes of parotid swelling and/or pain associated with fever or malaise over a period of years. In most cases the symptoms resolve spontaneously after puberty but all children should be screened to exclude Sjogren's syndrome, lymphoma and immuno deficiency including Human immuno deficiency virus. Our case is a nine year old girl who had 10 episodes of recurrent right parotid swelling in the past 2 years.

Keywords: recurrent, parotitis, juvenile.

INTRODUCTION

Juvenile recurrent parotitis (JRP), an inflammatory disease of unknown etiology of the salivary glands characterized by recurrent unilateral or bilateral swelling of the parotid gland with pain, redness, occasional fever and hypo secretion by the affected gland can at times be symptom free with very viscous or flocculent saliva. The age of onset is between 4 months and 15 years and is usually self-limiting with puberty and with the frequency of episodes decreasing and subsiding by adulthood. Boys are affected more often than girls. The classic pathological change in JRP is duct ectasia of the intraglandular duct [1]. Various imaging techniques often demonstrate less pronounced duct pathology on the asymptomatic side [2, 3]. In histological examinations, JRP presents itself in three stages: stage I with peri ductal lymphocytic infiltration; stage II showing the formation of lymphatic follicles; stage III, which is seldom seen, with lymphatic transformation of the gland [4]. Though very rare, it is still one of the most common salivary gland diseases of childhood after mumps [5].

The etiology and pathogenesis of this disease is believed to be multifactorial ranging from congenital malformation of the parotid glands leading to retrograde infection, allergy and association with autoimmune diseases. The diagnosis is mainly based on history and physical examination which is confirmed by imaging such as sialography and ultrasonography of the parotid

glands. Management of recurrent parotitis is also controversial. Treatment is conservative with antibiotics and analgesics with aggressive therapies for the more persistent symptoms [6, 7, 8].

CASE REPORT

A 9 year old female child presented with repeated episodes of painful swelling below the right ear (Fig.1). She had 10 such episodes lasting 5-7 days each in the last 2 years. The present episode was there for last 2 days. There was no history of fever or dryness of mouth and eyes, joint pains and swellings and skin rashes suggestive of autoimmune disorders. On examination a right parotid swelling was noticed which was roughly oval in shape measuring 3 x 4 cm in diameter, and the skin over the right cheek was stretched. On palpation it was firm in consistency, tender and febrile. There were no palpable lymph nodes in the neck. There was no erythema around the parotid duct opening and there was serous discharge on pressing the gland. The laboratory investigations revealed hemoglobin of 11.1 g/dl and total leukocyte count of 5,600 cells/mm³ with 63% neutrophils, 32% lymphocytes and 3% eosinophils. ESR was 10mm/hr. Investigations for autoimmune markers were not done. Human immunodeficiency virus (HIV) serology was negative. Ultra sonogram of the right parotid gland revealed multiple hypo echoic areas and heterogeneous distribution of internal echoes suggestive of inflammatory changes and dilation of acini in both

glands (Fig.2). Fine needle aspiration cytology findings were also favoring the diagnosis of sialoadenosis. Based on the history given by the patient, clinical picture, laboratory investigations, ultra sonogram, FNAC, a provisional diagnosis of Juvenile Recurrent Parotitis (JRP) was made. She was treated by i.v cefotaxime 500 mg bd for 3 days followed by oral taxim 100mg bd for

next 4 days along with a combination of Diclofenac 50 mg and Paracetamol 500 mg tid. Swelling as well as symptoms reduced significantly in three days (Fig.3). The child was asked to take plenty of fluids during such episodes and the parents were reassured that the symptoms might disappear as the child grew.



Fig-1: Swelling in the right parotid region below the ear

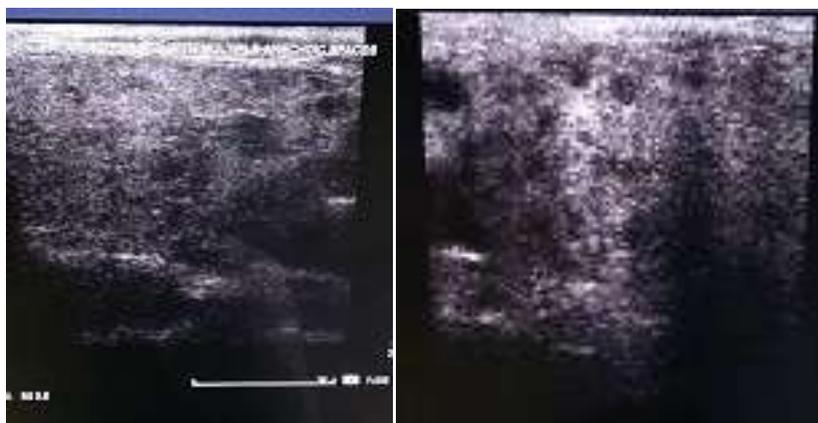


Fig-2: USG of right parotid gland showing multiple anechoic and hypo echoic spaces



Fig-3: Reduction of swelling in the right parotid region after treatment with antibiotics and analgesics

DISCUSSION

Juvenile recurrent parotitis characterized by recurrent episodes of swelling and pain in parotid gland [9] and is usually misdiagnosed as mumps but in contrast, the swelling is recurrent and affects the parotid gland both unilaterally and bilaterally though one is affected less. The onset of disease is early in life with a peak during 3-5years of age. Leerdam et al have shown a biphasic age distribution with peaks at 2-5 and 10 years [10]. In the present case the child had the first episode at the age of 8 years. The etiologies ranges from

immunodeficiency, allergy, upper respiratory infections, mumps etc but none has been conclusive [8]. Friiset a proposed an autoimmune origin, but failure to detect auto antibodies makes this unlikely [11]. Upper respiratory tract infections may set off attacks of sialadenitis by causing dehydration in a child with sialactasis [2,8]. Recurrent parotitis may be the first manifestation of HIV infection or an immune deficiency disease. Shkalim *et al.*; suggested lack of IgA may be involved in the pathogenesis of recurrent parotitis [12].

It is usually accompanied by pain, fever and malaise and the frequency of exacerbations can be quite variable, though the disease disappears completely in adult life [8]. In a study done by Leerdam *et al.*; in 53 children with recurrent parotitis the commonest symptoms were swelling (100%), pain (92.5%) and fever (41.5%)[8]. Surprisingly in our case, there was no associated fever during episodes.

Although conventional parotid sialogram is a hallmark in the diagnosis of juvenile recurrent parotitis the invasive nature and the difficulties encountered led to the advent of newer diagnostic modalities such as ultrasound, CT and MR sialography. MR Sialography does not require any contrast medium. It primarily images liquid structures and the flow more so after stimulation with ascorbic acid. These special features of MR Sialography allow its use also during acute episode of sialadenitis [12]. USG using 7.5 MHz high frequency transducer is superior and reveals enlarged parotid gland in majority of patients with multiple small hypo echoic areas measuring 2-3mm in diameter [13, 14]. The hypo echoic areas represent both sialectasis of peripheral ducts and surrounding lymphocytic infiltration. Sialoendoscopy gives better diagnosis with an endoscopic white appearance of the ductal layer without the healthy blood vessel coverage [5].

The recurrent attacks are treated conservatively with oral antibiotics and analgesics which is believed to prevent additional damage to glandular parenchyma [6, 8]. However, no prophylactic therapy is available. Good oral hygiene, massage of the parotid gland, warmth, use of chewing gum and sialogogic agents may be helpful in reducing the attack frequency [6]. More aggressive treatment is justified only for those patients with persistent problems and includes parotid duct ligation, parotidectomy or tympanic neurectomy depending upon preference and experience of the treating surgeon [6].

CONCLUSION

In general, most children need reassurance, analgesia and little intervention. The risk of over investigation and treatment should be borne in mind. We recommend a more conservative management for recurrent parotitis in children as most symptoms resolve after puberty. Antibiotics should be given before suppurative parotitis is excluded. Ultrasonography is sensitive than sialogram and should be done to confirm the diagnosis Other appropriate investigations such as blood tests for white cell count, amylase and autoimmune markers should be done to exclude other differential diagnoses such as Sjogren's syndrome and immunodeficiency.

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