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Research Article

Chronic juvenile recurrent parotitis: a retrospective study in a tertiary care hospital in South India

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ABSTRACT

Background: Chronic juvenile recurrent parotitis is a rare disease with unknown etiology. This study was aimed to analyze clinico-social parameters of chronic juvenile recurrent parotitis in children.
Methods: Case records of nine children with recurrent parotid swellings and radiological diagnosis of chronic juvenile recurrent parotitis were reviewed from September 2012 to March 2015. These were assessed for parameters like gender, age, laterality, number of recurrences, symptoms of presentation, associated conditions, radiology reports, treatment done, outcome and follow up. Results were analyzed by using percentage.
Results: Male to female ratio was 2:1. Mean age of presentation was 6.33 years. Five cases (62.5%) were left sided, three cases (33.3%) were right sided and one case had bilateral parotid involvement. Pain and swelling were present in all nine patients. Fever and whitish discharge from duct were present in seven and two patients respectively. Numbers of recurrences were in range of one to 12 times per year. Six (66.7%) children had associated active viral respiratory tract infection during or just before the attack. Sonography was diagnostic in all cases. Sialography showed punctate sialectasis. Two patients developed suppuration in their second recurrence and underwent drainage. Mean follow up period was 7.1 months (1- 24 months).Seven patients were asymptomatic till last follow up. Two patients had recurrence while on oral antibiotic treatment.
Conclusions: In chronic juvenile parotitis morbid recurrences are common after variable interval and even while on treatment. Acute viral infections contribute to exacerbation of symptoms. Sonography is useful in diagnosis. It may rarely suppurate and need drainage. However a larger prospective study is needed to establish these findings.

Keywords: CJRP, Sonography, Sialography, Sialoendoscopy

INTRODUCTION

Though bilateral inflammatory parotid swellings in children are common, chronic juvenile recurrent parotitis (CJRP) is a rare cause of parotid enlargement. It is an inflammatory disorder of parotid gland characterized by multiple episodes of unilateral or bilateral parotid swellings with or without pain.[1] Exact etiology of CJRP is not known.[2] Morbid recurrences are common after resolution of initial episode. Since exact etiology is not clear, treatment of this condition is also not standardized. Sonography is the primary investigation and it helps in providing anatomical details of ductal dilatation and obstruction. We reviewed our data on CJRP and analysed selective clinico-social parameters. We also discussed available literature on management of this arduous disease.

METHODS

The study design is retrospective record based. All cases below 12 years with radiological diagnosis of CJRP seen

in paediatric surgical outdoor were included in study. Time period was two and a half years from September 2012 to March 2015. Out of 11 patients of parotid swelling, nine cases meeting inclusion criteria were evaluated for parameters like gender, age, laterality, number of recurrences, symptoms of presentation, associated conditions, sonography and sialography details, treatment, outcome and follow up. Two patients with history of first episode and clinical diagnosis of mumps were excluded. Results were analysed using percentage.

RESULTS

Table 1: Distribution of study parameters.

Parameters n=9	Results	Percentage %
Male : female	2:1	-
Mean age at presentation	6.33 years	
Laterality	5 left side 3 right 1 bilateral	62.5 33.3 11.1
Symptoms at presentation	Pain 9 Swelling 9 Fever 7 Whitish discharge from duct 2	100 100 77.7 22.2
Mean duration of symptoms	7. 4 days (3 hours to 20 days)	-
Symptom free interval	10 days – 1 year	-
Associated respiratory infection during attack	6	66.7
Maximum recurrences per year	12	-
Recurrence during follow up	2	22.22

Male to female ratio was 2:1. Five cases (62.5%) were on left side, three (33.3%) on right and one case had bilateral parotid involvement. Youngest child was four years old and eldest was 12 year with mean age at presentation of 6.33 years. Pain and swelling were most common presenting symptoms seen in all cases (Figure 1). Fever in seven (77.7%), and whitish discharge from Stenson's duct in two (22.2%) cases were next common symptoms of presentation. Discharge was more conspicuous on pressing the gland. Six (66.7%) patients had associated active respiratory tract infection just before acute attack. One child also had associated vitamin D resistant rickets and renal tubular acidosis along with stage four chronic kidney diseases. One child had seizure disorder and he was on sodium valproate. Recurrences were in range of one to 24 with average of 6.2 times. Duration of symptoms ranged from three hours to 20 days with mean of 7.3 days. Symptom free interval ranged from one month to one year. One patient with 24 episodes had recurrence every month for two years (12 attacks per year) and one with 12 episodes had recurrences every two months (six attacks per year). Sonography was done in all cases as primary investigation (Figure 2 A, B, C). Reports showed multiple hypoechoic lesions with enlarged intraglandular lymph nodes and enlarged gland. Sialography was done in two cases. It revealed punctate and globular ductal ectasia with non-obstructed stenson's duct. (Figure 3A, B, C) Fine needle aspiration cytology (FNAC) in two patients was inconclusive. Culture for aerobic organisms from drained collection and ductal secretions was done in three patients and it showed no growth. All nine patients were managed with amoxicillin plus clavulanic acid and analgesics along with supportive treatment for viral respiratory tract infections. Two patients developed suppuration and underwent drainage. Mean follow up period was 7.1 months (1-24 months). Seven patients were asymptomatic till last follow up. Two patients had recurrence while on treatment (Table 1).

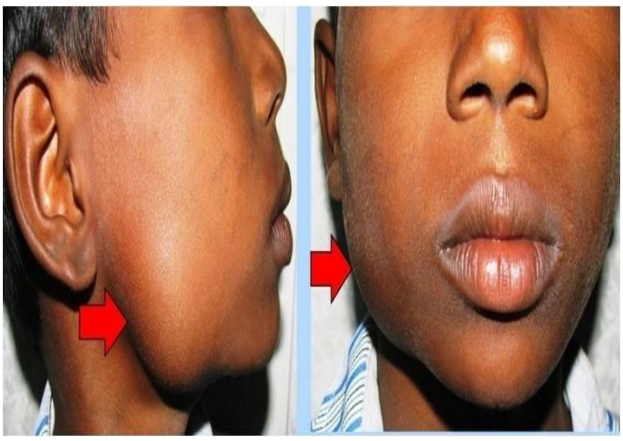

Figure 1: Clinical photograph of a 12 year old boy with right parotid swelling.

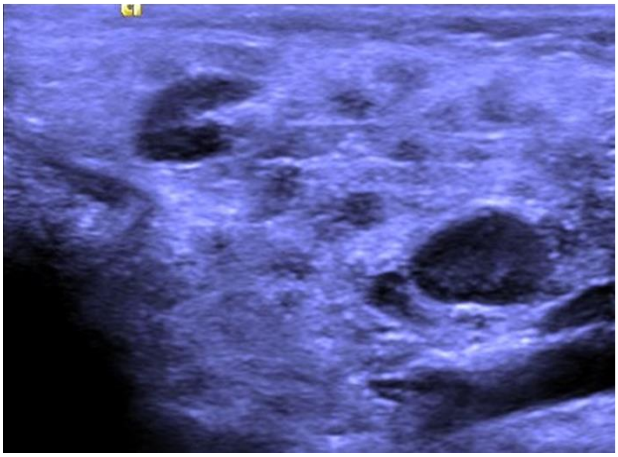

Figure 2 (A): Sonography of parotid gland showing intra parotid lymph nodes.

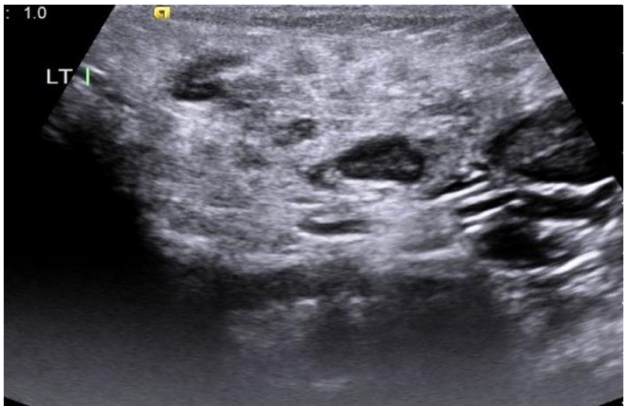

Figure 2 (B): Sonography of parotid gland showing multiple hypoechoic areas.

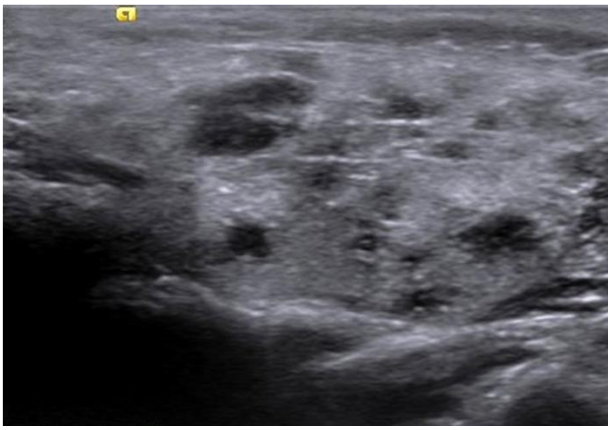

Figure 2 (C): Sonography of parotid gland showing multiple hypoechoic areas with enlarged gland.

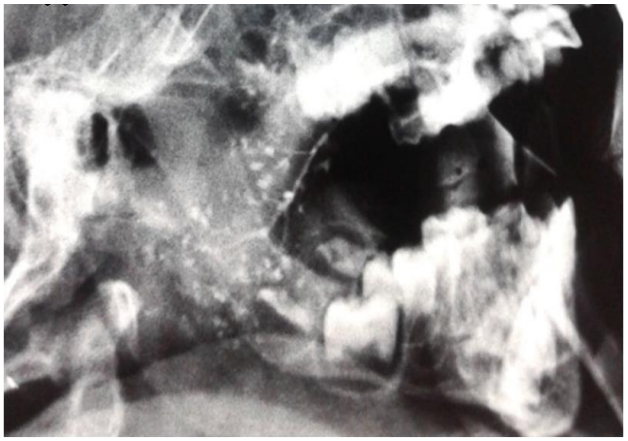

Figure 3 (A): Conventional Sialography showing ductal ectasia as "punctate lesion".

DISCUSSION

CJRP is often synonymously referred in literature as infantile chronic recurrent parotitis (ICRP) or juvenile recurrent parotitis (JRP).[3,4] Juvenile form of recurrent parotitis was first described by von Reuss.[5] Since then its exact etiology and treatment remain elusive. In his review of 5000 cases of chronic sialadenitis, Seifert reported 27% incidence of CJRP. He distinguished between juvenile and adult form. Juvenile form of parotitis is reportedly ten times less common than adult form.[6]

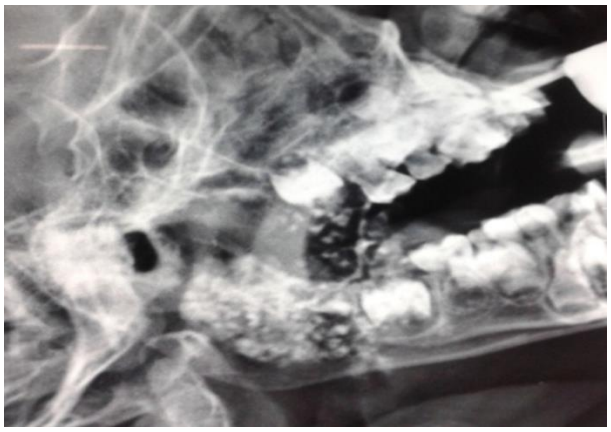

Figure 3 (B): Conventional Sialography showing ductal ectasia as "punctate lesion".

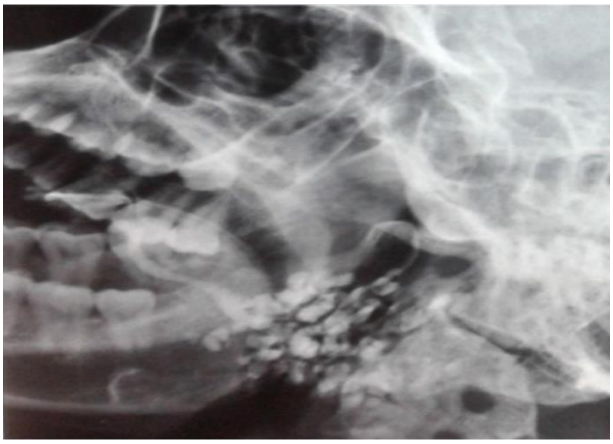

Figure 3 (C): Conventional Sialography showing "Globular pattern" in advanced case with non-dilated stenson's duct.

Most commonly CJRP affects age group of three to six years, but patients may range from three months to 13 years.[7,3] Some studies mention biphasic age distribution with first peak at two to five years and second at 10 years.[8]Boys are more commonly affected, but some studies also report identical incidence in both gender.[9,3] Usually unilateral involvement is more common than bilateral. Boys outnumbered girls in our study and the age of presentation correlated with literature.[10] In our series, eight cases were unilateral. Left side was more commonly affected than right. One bilateral case also had initial left side infection followed by right sided involvement after six months.

The aetiology of CJRP is reported as multifactorial. Genetic factors, familial cause with autosomal dominant inheritance, autoimmune causes like Sjogren's disease, viral etiology like mumps, allergy, and IgG3

immunodeficiency have all been implicated as possible causes of CJRP.[9] But there is no definite evidence in support of any one of these. Recently Morales –Bozo and Wu AJ, et al showed increased matrix metalloproteinase two and nine (MMP2 and MMP9) in CJRP and Sjogren disease. These high levels were linked to degree of gland damage.[11,12] Recent studies have also concluded that congenital malformation of ducts with bacterial super infection is most likely pathogenesis. Saliva examinations have shown higher levels of IgA, lactoferrin, kallikrein in affected cases.[13] In our study three members of one family were affected with similar disease. Mother and child were simultaneously treated and her sister was also affected with similar disease. Other six (66.7%) cases had onset of acute respiratory infections just prior to and during acute symptoms. Since these acute episodes were associated with acute respiratory tract infections, role of virus in recurrence or acute exacerbation appear significant. But small sample size and retrospective nature of our study is a limiting factor for establishing these findings. Our study is also limited by the fact that being retrospective, all details of required investigations for other causes like HIV, immunodeficiency or auto antibodies were not available.

Most common symptoms of recurrent parotitis include fever and painful swelling.[10]The swelling and pain are independent of meals and seasonal variations.[9] Swelling may persist for 24-48 hours or sometimes even for months or years.[9] Whitish viscous saliva from Stenson's duct may be rarely seen. Mean reported frequency is eight episodes per year. [7]Shkalim V et al has reported more than 20 attacks per year.[14] A retrospective study of 30 patients has reported 34 attacks per year.[10] Symptom free interval is variable and may range from months to years.[3] In our study symptom free interval was in range of one month to one year. Maximum number of recurrences per year in our study was.[12] These findings suggest that CJRP is randomly recurring and intervening normal period does not mean cure.

CJRP can be provisionally diagnosed by clinical symptoms and examination.[9] This may be aided by radiological investigations like ultrasonography, magnetic resonance imaging (MRI) and conventional sialography. Ultrasonography is a primary investigation and is sensitive in diagnosis of CJRP.[15] Ultrasonography features of CJRP includes multiple hypoechoic lesions with gland enlargement and lymph nodal enlargement.[1] Studies have suggested that hypoechoic areas and anechoic areas in parotid are consistent with ductal ectasia and lymph node enlargement.[16]Clinical examination and sonography as a combination are sensitive tools for diagnosis of CRP. MRI Sialography is recently been advocated as useful supplement to conventional sialography.[17] It is non-invasive and delineates parenchymal and ductal abnormalities better than conventional sialography. In our review reports were suggestive of similar findings. Two patients were subjected to conventional sialography. It revealed ductal ectasia with punctate pattern and normal stenson's duct in one and globular pattern in another. However role of conventional sialography is now debated because of its invasiveness and radiation exposure. None of our patients had been subjected to MRI Sialography.

Since its first description in 1909, as its aetiology remains obscure, the treatment of CJRP is also still elusive. Children report with recurrent acute swelling and pain and initial treatment is aimed at reducing the acute inflammation by analgesics. Antibiotics like amoxicillin with clavulanic acid or cephalosporin have been suggested to expedite this process.[9] Irrigation of gland with normal saline often leads to relief of symptoms. Use of sialoendoscopy along with irrigation with normal saline and cortisone has been done with good results. Shacham et al in their study of 65 patients did irrigation with saline and hydrocortisone after sialoendoscopy. 95% of patients were symptom free following one treatment in 6-36 months followup.[18] However, a study of 14 patients from Milan, reported recurrence rate of 36% with this treatment.[15] Although none of our patient underwent regular irrigations with saline or antibiotics, two patients in whom conventional sialography was performed reported relief from pain and the whitish mucoid discharge was diminished. All other patient initially responded to analgesics and antibiotics. Two of our patients had acute suppuration after previous episodes and required drainage. Though literature report CJRP as non suppurative, Stong et al treated at least four similar patients of CJRP with suppuration in five years.[19] As seen in our study, sometimes with acute exacerbation suppuration may occur.

This disease is often self-limiting and symptoms subside with increasing age.[3] It has been therefore suggested that for mild symptoms or after single episode, analgesics and antibiotics may be used. Ductal irrigation and sialoendoscopy may be used for severe symptoms or recurrent cases. Complete failure of these treatment and gland with total lymphatic transformation with immunological end stage may need total parotidectomy as last resort.[20] Our experience with these cases has revealed that this treatment does not seem to alter the course of disease. Two of our patients had recurrences while on treatment.

CONCLUSION

We conclude that CJRP is a difficult disease to treat as morbid recurrences are common after variable interval and even while on treatment. Acute viral infections contributed to exacerbation of symptoms in this study. Sonography with typical features is useful in diagnosis. Cases with mild to moderate severity can be managed with antibiotics and analgesics. However, they may rarely suppurate and need drainage. A larger prospective study is needed to establish these findings.

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