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Case Report

Juvenile recurrent parotitis

Lt Col R.P.S. Tomar^{a,*}, Col R. Vasudevan^b, Lt Col Manoj Kumar^c,
Maj D.K. Gupta^d

^a Classified Specialist (Pediatrics & Neonatology), MH Jalandhar Cantt, Punjab 144005, India^b Senior Advisor (Pediatrics), MH Jalandhar Cantt, Punjab, India^c Classified Specialist (ENT), MH Jalandhar Cantt, Jalandhar, Punjab, India^d Graded Specialist (ENT), MH Jalandhar Cantt, Jalandhar, Punjab, India

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Introduction

Juvenile recurrent parotitis (JRP) is defined as recurrent inflammatory parotitis in children of unknown etiology.¹ It is a rare condition and 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 immunodeficiency including human immunodeficiency virus. We present a 9-year-old boy who had 11 episodes of recurrent parotid swelling.

Case report

A 9-year-old boy presented with recurrent episodes of bilateral painful swelling below the ears for the past 4 years. He had

altogether 11 such episodes, each time the swelling lasted for 8–10 days. The last episode persisted for 6 days. During these episodes he did not have dryness of the mouth, increased salivation or altered taste. There was no history of dryness of eyes, joint pains or swelling and skin rashes suggestive of autoimmune disorder. There was no family history of recurrent parotid swelling. On examination the child was afebrile with parotid swellings which were smooth, soft to firm in consistency and tender. Pressure over the gland did not cause any purulent discharge from the parotid duct. There were no signs of xerophthalmia or xerostomia. His blood counts, erythrocyte sedimentation rate and serum immunoglobulin levels were normal. Human immunodeficiency virus (HIV) serology, rheumatoid factor, antibodies including anti-Ro and anti-La for Sjogren's syndrome were negative. Ultrasound of both parotid glands demonstrated heterogenous internal echogenicity and nodular hypoechoic areas suggesting sialectasis. A diagnosis of recurrent parotitis was made and the child was asked to take high fluid intake and analgesics during such episodes. He has remained asymptomatic on follow-up of 18 months.

Discussion

Juvenile recurrent parotitis is the second most common cause of parotitis in childhood, only after mumps.^{1,2} It commonly begins between 3 and 6 years of age and is often not diagnosed, goes unnoticed, or is mistaken for the mumps, otitis, or pharyngitis. The disease more frequently affects males but higher distribution in females was found when symptoms appeared later.³ Clinical symptoms of JRP include recurrent parotid inflammation with swelling and pain associated with fever

* Corresponding author. Tel.: +5533 (o), +91 9876671871 (mobile).

E-mail address: tomar15@rediffmail.com (R.P.S. Tomar).

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which usually lasts 2–7 days. This pathology is usually unilateral, but can occur bilaterally with symptoms usually more prominent on one side. The natural history of this disease is its recurrence with average number of attacks per year range from 1 to 20.¹ In 90% of the patients, the symptoms resolve spontaneously by puberty. In few severe cases there is progression leading to the destruction of the glandular parenchyma with a diminution of its functionality by 50%–80%.⁴ Although most of the cases are idiopathic, juvenile recurrent parotitis might be the first presenting symptom of an underlying variable immunodeficiency, HIV infection and Sjogren's syndrome.

The pathogenesis of JRP remains unclear and the present consensus favors a multifactorial origin. Various factors that have been suggested for the development of JRP include congenital ductal malformations, hereditary genetic factors, viral or bacterial infections, allergy, and local manifestation of an autoimmune disease.^{1–6} However, the main cause postulated to explain pathogenesis of JRP is decreased salivary production with an insufficient salivary outflow through the ductal system which favors ascending salivary gland infections via the oral cavity.³ Partial obstruction due to retention is gradually followed by duct dilatation, which further facilitates infection. Pathological studies reveal several structural changes in the affected parotid gland, while there is still a debate whether these are the primary abnormalities or the result of multiple episodes of parotitis. Histologically there are intraductal cystic dilatations of peripheral ducts with periductal lymphocytic infiltration, called as sialectasis.¹ The ectatic ducts are usually 1–2 mm in diameter and typical have a white appearance of the ductal layer without the healthy blood vessel coverage, when compared with a normal gland.⁷ This aspect is believed to be characteristic of JRP. Sialectasis was previously diagnosed by sialography, but ultrasound has superseded this technique.⁴ Although MR and MR sialography cannot substitute ultrasound, they can accurately depict findings such as sialectasis and signal intensity changes in the parotid gland depending upon the phase of the disease (acute vs. chronic inflammation).⁸ Quenin and colleagues found sialendoscopy more sensitive than ultrasonography.⁵

Treatment options range from conservative to invasive surgical procedures. Initially conservative treatment is indicated because the natural history of JRP includes spontaneous resolution in 90% cases. Analgesics, attention to good oral hygiene, massage of the parotid gland, warmth, use of chewing gum and sialogogic agents are helpful. Antibiotic treatment during attack is often proposed to prevent additional damage to the glandular parenchyma while low-dose prophylactic antibiotics have been recommended when an immunoglobulin A deficiency is observed.⁶ However, recurrent swelling and over a long period can affect patient's social life and school activities. Moreover, few patients can develop sequelae such as recurrent pain, chronic swelling and decrease in glands function. Such patients are candidates for

more interventional treatment which can further be substantiated with sialographic evidence of multiple strictures and a need to dilate them along with washing of multiple plaques. Nahlieli et al performed duct probing with lavage, dilation and hydrocortisone injection via sialendoscopy with resolution of symptoms and a very low recurrence rate on follow-up.⁷ Philippe Katz and colleagues used a less invasive treatment method by performing sialography and installing iodinated oil where recurrences were lesser in such treated cases.³ More aggressive treatment is justified only for those patients with persistent problems and includes parotid duct ligation, parotidectomy or tympanic neurectomy but all have unsatisfactory results.⁹ There are reports that some JRP with sialectasis are young individuals with Sjogren's syndrome or in whom SS will develop in adult life.¹⁰ Hence regular follow-up and early management are required for pediatric patients with bilateral parotid swelling and paediatricians should be aware of this disease and treatment options.

Conflicts of interest

All authors have none to declare.

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