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CASE REPORT

Clinical and laboratorial aspects related to juvenile recurrent parotitis

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Abstract:

Juvenile Recurrent Parotitis (JRP) is an inflammatory condition characterized by recurrent parotid gland inflammation episodes of non-suppurative and non-obstructive nature. This is the second most common salivary disorder in childhood. We present a case of a 5-year-old girl with a recurrent facial swelling on the left side. Panoramic x-ray image excluded any dental or osseous involvement, as well as the presence of sialoliths. Ultrasonography showed hyperechoic focal areas with cavitation of the parotid gland corresponding to sialectasis. Sorological tests for infections like HIV, cytomegalovirus, mumps and other chronic diseases were negative. Based on the clinical and laboratorial features, the diagnosis was compatible with JRP. After two months of follow-up, the patient returned without any swelling or symptomatology. Seven months later, the patient returned with a new facial swelling episode, and this time on the right side. Clinicians should be aware of this condition and include in the differential diagnosis of salivary gland diseases of pediatric patients.

Keywords: parotitis; salivary gland diseases; pediatric dentistry; diagnosis differential

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INTRODUCTION

Juvenile recurrent parotitis (JRP) is a recurrent parotid inflammation with no suppuration usually associated with a painful, intermittent, reddish swelling, accompanied by fever. The involvement of parotid glands can occur unilaterally or bilaterally, affects children between 3-6 years old and is dominant in male sex. Each episode may occur every 3-4 months and last a few days to weeks for complete resolution. After mumps (infection caused by the paramixovirus), JRP is the second most common lesion involving the salivary glands. In pediatric patients, swelling of the salivary glands may occur due to inflammation or microbial agents. In fact, one must consider the differential diagnosis of these lesions: siloliths, mumps, benign lymphepithelial lesion, acute suppurative parotitis, HIV infection, Mikulicz disease, Sjögren syndrome and Heerfordt's syndrome[1-5].

The diagnosis of JRP is made after the first occurrence (the first occurrence usually ends up being ignored), requiring a detailed medical history, evaluation of clinical and imaging aspects. Here, we present a detailed case of a 5-year-old girl with a recurrent facial swelling, emphasizing their diagnostic procedures and differential diagnosis.

CASE REPORT

A 5-year-old girl was referred to the Pediatric Clinical for evaluation of facial swelling on the left side. Her medical history revealed 3 previous episodes, being the first episode on the right facial side, followed months later by the swelling of the left side and then another recurrence on the right side, all within one year. Complete physical examination revealed a painful parotid enlargement on the left side accompanied by fever without lymphadenopathy (Fig. 1). On intraoral examination, there was no evidence of any swelling or lesion and the parotid had normal salivary function and no sign of suppuration. She showed no trismus or alterations in the oropharynx. After two months of follow-up, the patient returned without any swelling or symptomatology. Seven months later, the patient returned with a new facial swelling episode, and this time on the right side. No other different clinical characteristics from the previous episode could be noted.

Diagnostic imagining was indicated in our patient. The panoramic x-ray image excluded any dental or osseous involvement, as well as the presence of sialoliths (Fig. 2a). Ultrasonography showed hyperechoic focal areas with cavitation of the parotid gland corresponding to sialectasis (Fig. 2b). Fine-needle aspiration was not indicated due to the recurrence historic in this case.

Blood count, anti-cytomegalovirus, salivary amylase level, immunological tests (IgG, IgA, IgM immunoglobulins, IgG subclasses, anti-mumps antibodies), as well as autoantibody profile including antinuclear antibodies, anti-double-stranded DNA antibodies, anti-Ro (SS-A), anti-La (SS-B) antibodies, anti-HIV-1/2, rheumatoid factor, total proteins, protein C Reactive, VHS, and FAN were recorded. All tests revealed values within the normal range, except for amylase activity that was upregulated and total leucocyte count, with a left shift. HIV serological survey was negative. The patient was up to date with all her immunizations. All these features lead us to the diagnosis of Juvenile Recurrent Parotitis (JRP).

The established conduct was to manage the symptoms using only nonsteroidal anti-inflammatory and monitoring. There was a complete resolution within 20 days later without any other treatment and no other recurrence was registered in 18 months of follow-up.

DISCUSSION

The JRP is characterized by recurrent parotid gland inflammation episodes of non-suppurative and non-obstructive nature.[1] This inflammatory condition of salivary glands is the second most common in childhood and symptoms like parotid gland swelling, pain, fever, and malaise are present.[2] JRP is most often unilateral,

Figure 1. (a and b) Clinical aspect showing unilateral parotid enlargement in the left side during the active phase of JRP in the clinical evaluation. Lymphadenopathy was not present.

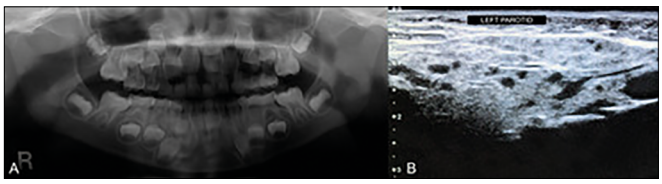

Figure 2. (a) Panoramic x-ray image showing no dental or osseous involvement. (b) Ultrasonography image of the left parotid glands with heterogeneous echogenicity and multiple hypoechogenic focal areas.

affects children between 3-6 years old and is dominant in male sex. Each episode may occur every 3-4 months and last a few days to weeks for complete resolution. Usually, JRP episodes vanish after puberty; however, this condition affects significantly children's quality of life.[1] Although the etiology of this affection remains unclear, a multifactorial cause (genetic, immune, infection, dehydration, allergy and ductal abnormalities) has been suggested.[1-3] Diagnosis is made at least after two attacks by medical history and clinical examination and confirmed by imaging.[1, 2] Image exams like ultrasonography or sialography may show sialectasis and ductal.[2] Despite being less frequent, the differential diagnosis should include other conditions such as siloliths, mumps, benign lymphepithelial, acute suppurative parotitis, HIV infection, Mikulicz disease, Sjögren syndrome and Heerfordt's syndrome.

The JRP is probably misdiagnosed and it is also known as recurrent parotitis or recurrent sialectatic parotitis. JRP is defined as a recurrent painful swelling of one or both glands with non-suppurative and non-obstructive parotid inflammation.[1] It has a male predominance and the first episode may occur between the first and second years. The peak age of onset is between 3 and 6 years and it is distinguished from suppurative parotitis by the lack of pus from the parotid duct.[1, 2] Redness and fever may be usually present. The patients may experience different frequency of episodes, usually each episode may happen every 3-4 months. When active, usually it may last a few days up to a couple of weeks.

The etiology and pathogenesis of JRP have not been clarified yet. However, some cases reported until today were believed to be associated with recurrent infection, allergy, congenital/structural defects and genetic factors.[1, 4-6] Also, Sjogren's disease, immune deficiency, hypogammaglobulinemia, isolated IgG3 deficiency, and IgA deficiency may be found in association with JRP.[2, 7, 8] Up to 60% of patients affected presents swelling of the parotid as the first sign. An autosomal dominant inheritance with incomplete penetrance was suggested based on a case report of a family with four generations of affected children by JRP.[5, 6] In our case a leukocytosis and high levels of amylase were present. These features were associated with JRP and were already described in the literature.[9]

There are two main hypotheses that try to explain the mechanism involved. One of them, suggests that decreased salivary flow leads to stasis and, over time, to damage to the ductules. The second main hypothesis is that the ductules are already abnormal, due to congenital/structural anomalies, and therefore susceptible to low-grade inflammation/infection, and this is supported by studies that evaluated bilateral sialograms considering the context of unilateral disease.[1, 4-6] Many asymptomatic glands had similar though often milder lesions. Furthermore, the histopathological study of these lesions supported degeneration due to latent chronic inflammation.[1, 4]

The diagnosis of JRP is made on a clinical basis and should be done by exclusion of other conditions. and may often be confirmed by ultrasonography, showing the classical features of sialectasis. Through this exam, it is possible to find multiple hypoechoic areas that correspond to the punctate pools seen on sialogram.[4] It typically shows widespread, multiple, sharply demarcated, round areas (1–3 mm) of contrast, which persist in delayed films.[4] Histologically, periductal lymphocytic infiltration is seen.[4] Computed tomography, magnetic resonance imaging (MRI) scans and MR sialography may also be used. Ultrasonography has a further benefit of excluding stones, abscesses and mass lesions, making other more invasive procedures unnecessary.[1]

In most of cases, JRP resolve itself spontaneously after puberty although some cases persist into adulthood.[6] The treatment basically is to control the symptoms and to prevent further damage. In a recent systematic review, the use of sialendoscopy has also been described as a successful and safe alternative treatment to JRP, allowing the irrigation and washout within the gland under direct observation addressing the inflammation without the use of anti-inflammatory drugs.[4] The successful effect is attributed to the removal of mucus plugs and dilation of structures. The major implications for the patients about the recurrences, besides the influence in the quality of life, is that it may also cause a progressive gland parenchyma destruction and consequently lead to superficial or total parotidectomy.[5, 6] In summary, clinicians should be aware of this condition and include in the differential diagnosis of salivary gland diseases in pediatric patients.

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