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Case Report

Juvenile Reccurent Parotitis - Case Report

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A B S T R A C T

Title: Juvenile recurrent parotitis (JRP) is one of the rare causes of chronic parotitis in children. Its physiopathology is debatable, with the implication of genetic, local, and systemic causes that combined cause repeated spells of parotitis. JRP could be suspected clinically and affirmed by ultrasound and MRI but should not be retained before eliminating other evident causes like mumps or Sjogren's disease.

Case presentation: We present the case of 11 years 11-year-old girl suffering from recurrent parotitis from the age of 5, the clinical examination and the US and MRI suggested JRP as a probable diagnosis. The patient benefited from sialendoscopy with irrigation of antibiotics and steroids.

Discussion: The diagnosis of JRP is retained after eliminating another obvious diagnosis like mumps and Sjogren disease. The US and MRI can be used to search for signs like communicating intraglandular formations corresponding to sialectasis. The acute phase is usually treated with analgesics, chewing gum, gentle parotid massage, sialendoscopy to explore the intraglandular duct and irrigation with saline solution. Often, the disappearance of the symptoms spontaneously marks puberty, but treatment could be used to minimize the number of recurrent spells and lessen the quality of life.

Conclusion: Sialendoscopy with irrigation is a promising therapeutic option with good initial results, according to the few studies that discussed this issue.

Keywords: Juvenile parotitis; Ductal ectasis; Sialendoscopy; Puberty

1. Introduction

JRP is one of the many causes of chronic parotitis in childhood[1] it is physiopathology remains uncertain. There is no consensus in the management of JRP, and because it is a rare disease, few cases have been reported in the literature to shed light on this entity. Yearly spells can vary widely between patients, and understanding why they resolve in puberty is unknown.

2. Case Presentation

We present the case of an 11-year-old girl with a medical history of recurrent swelling of the right parotid gland for the last four years admitted to our emergency department for another episode of painful swelling of the right parotid with a fever at 39 Celsius and alteration of the general state. The physical examination exhibited a painful swelling of the right parotid with reddish overlying skin with no associated facial palsy nor cervical lymphadenopathies; the buccal examination revealed a regular aspect of the saliva expressed for the Stenson's duct and normal dental status. The initial management consisted of white cell count, which was elevated and elevated (**Figure 1**).

C-reactive protein, a negative mumps serology, a negative autoimmune panel, and a US exam showed multiple hypoechoic lesions and heterogeneous echogenicity. We completed it later

with an MRI showing multiple communicating intraglandular formations with a liquid signal corresponding to diffuse sialectasis with a peripheral enhancement of a few of these ductal ectasias. Due to its recurrent nature, age, negative serologies, and the aspect of both the US and MRI, the diagnosis of Juvenile Recurrent Parotitis was retained. Initially, the management was conservative, and the patient was put on oral analgesics and anti-inflammatory drugs to ease the pain and an initial antibiotherapy consisted of 6 days of amoxicillin clavulanic acid. The patient was reviewed one week later with significant clinical improvement; the patient was notified of the recurrent nature of the disease, the possible occurrence of other acute episodes of painful swelling, and the importance of close follow-up to explore other therapeutical methods if needed (**Figure 2**).

In the latter six months, the patient exhibited four spells of acute painful swelling with a severe alteration of the QoL, with parents noting the onset of a depressive manner in the child with a tendency to isolation, which motivated us to discuss other therapeutical choices. One month later, our patient benefited from sialendoscopy with intraductal injection of steroids and antibiotics, showing more arguments in favor of JRP as ductal scaring the presence of multiple mucus plugs. The close follow-up at three months was normal. The patient exhibited no other acute spells of swelling and was advised to hydrate well, chew gum, and massage the parotid gland occasionally (**Figure 3**).

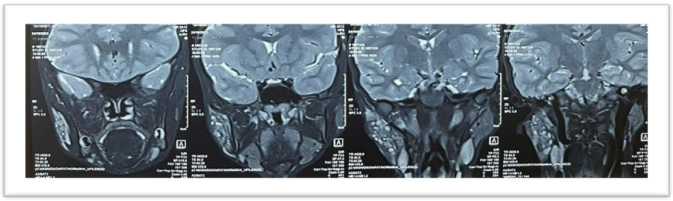

Figure 1: T2 coronal sequence showing an enlargement of the right parotid with multiple formations with a liquid signal corresponding to acinar dilatation.

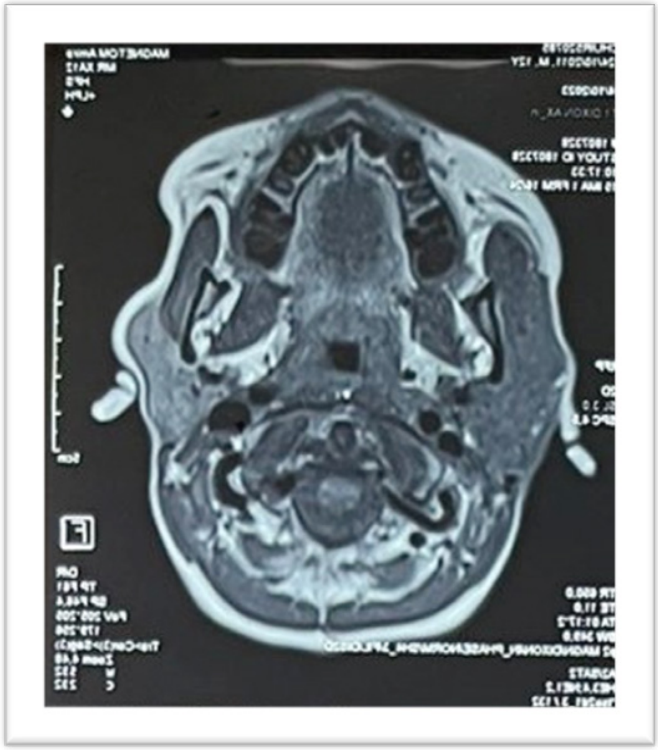

Figure 2: T1 sequence showing right enlarged parotid with a discrete hyper signal comparatively to the contralateral gland.

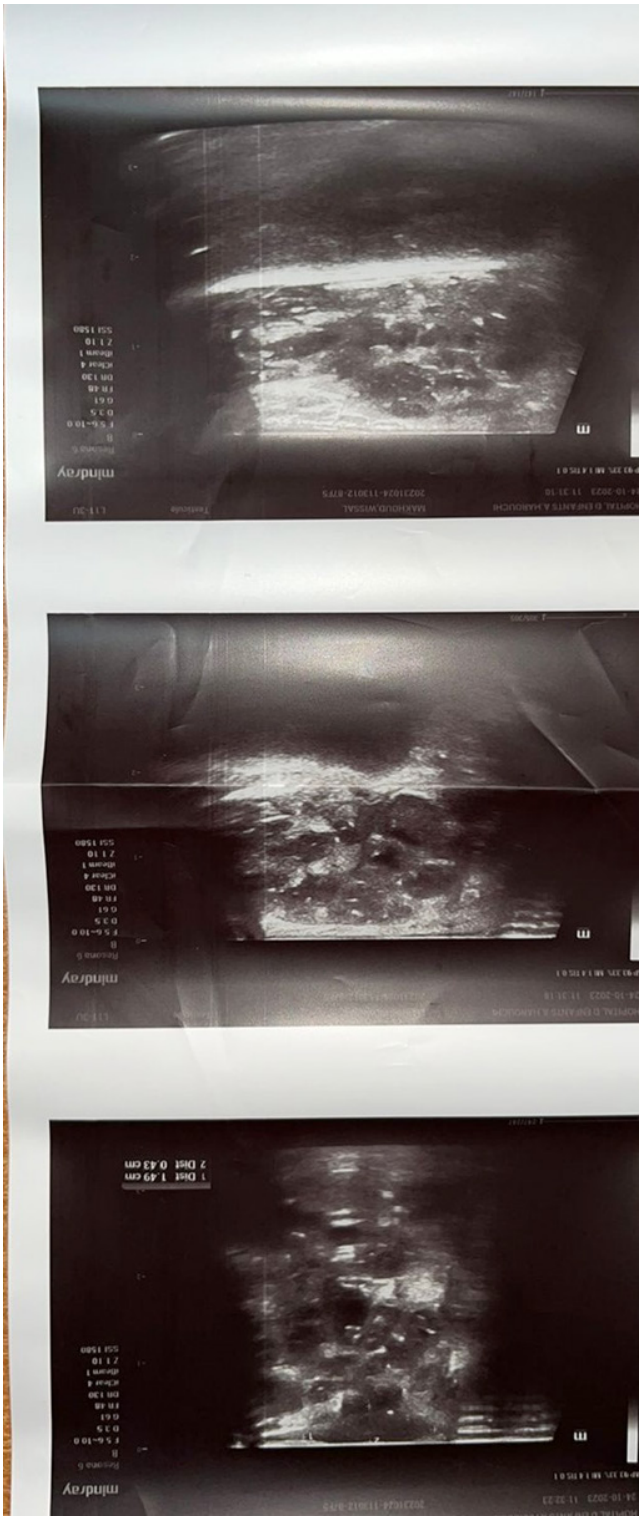

Figure 3: Ultrasound showing multiple hypoechoic formations with heterogeneous echogenicity.

3. Discussion

JRP is a rare form of parotid inflammation in children[2]. Its main characteristic is recurrent episodes of painful parotid swelling, often associated with fever and malaise[2]. The first episode typically occurs between the ages of 3 and 6 years (six months to sixteen years), more often in males[3]. The etiopathology of juvenile parotitis remains obscure[3]. The most accepted theories attribute this affection to the reducing salivary flow that favors abnormalities in the structure of distal ducts and predisposes to recurrent inflammation[5]. From the microscopic perspective, there are intraductal cystic dilatations of peripheral ducts called sialectasis. The ectatic ducts are usually 1 to 2 mm in diameter and have a white appearance of the ductal layer

without healthy blood vessel coverage, which is believed to be characteristic of JRP[6].

The diagnosis is based on the clinical aspect and can be confirmed by an ultrasonographic (US) study[4]. At this diagnostic procedure, typical findings are distal small roundish hypoechoic areas in the glandular parenchyma, corresponding to ductal dilatation, duct lymphocytic peripheral infiltration, or enlarged intraparenchymal lymph nodes[4]. Two MRI patterns have been identified as typical of juvenile recurrent parotitis: a T1-weighted hypointense and T2-weighted hyperintense signals, with contrast enhancement, in the acute inflammatory stage, and a T1-weighted and T2-weighted isointense signal, without contrast enhancement, in comparison with the other major salivary glands, in the chronic inflammatory[3,7].

Singh et al. considered sialendoscopy as their main mean of diagnosis in their series[8] due to generalized ductal stenosis, which was noted in the endoscopy procedure[8]. The most recognized sialendoscopy finding was represented by a pale, avascular, and stenotic stenson duct[9]. Nahieli et al. demonstrated the diagnostic value of sialendoscopy by visualizing strictures, hypovascularization, and white intraductal debris, which are characteristic of this disease[10]. They also showed the therapeutic efficacy of concomitant intraductal hydrocortisone lavage, with a 1-year recurrence-free rate of 89%.

Grande-Moreillo et al. concluded in their study that there was a significant difference in the number of inflammation episodes before and after sialendoscopy irrigation in their 9 cases of JRP[11].

4. Conclusion

Juvenile recurrent parotitis is a rare entity that is often seen in children before the age of puberty. Its cause is not quite elicited, and the diagnosis is retained in both clinical and specific findings on both MRI and ultrasound.

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