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SAT0500

JUVENILE RECURRENT PAROTITIS: A RARE MANIFESTATION OF RHEUMATIC IMMUNE-MEDIATED DISEASES

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Background: Juvenile recurrent parotitis (JRP) is the second most common childhood disease of the salivary glands after mumps and mainly affects children between the ages of 3 and 6. There is a male predilection, often with spontaneous resolution at puberty. However, in some cases, it can be the first manifestation of a rheumatic immune-mediated disease.

Objectives: To analyze the clinical, laboratory and imaging profile of children with JRP and investigate the prevalence of rheumatic immune-mediated diseases in these patients.

Methods: Retrospective study from 2008 to 2018 including all cases of recurrent parotitis with juvenile onset at our center. Parameters evaluated included gender, age, laterality, number of recurrences, symptoms of presentation, associated conditions, imaging details, blood tests, treatment, outcome and follow-up.

Results: 40 patients were identified over a 10-year period, with a female to male ratio of 5:3; 62.5% females. The youngest child was nine months old and the eldest was 16 years old with median age at presentation of 5 years. The median (min-max) follow-up period was 3 (0.04-33) years. 29 children (72.5%) only ever reported unilateral symptoms, while the rest had both glands affected, although not usually at the same time. Pain and swelling were the most common presenting symptoms, seen in all cases. Fever in 15 (37.5%) and whitish discharge from Stenson's duct in one (2.5%). 14 (35%) had palpable cervical lymphadenopathy and 4 (10%) had dental caries. The average reported frequency was 1.25 episodes per year, with a range of 0.43-5 episodes. Numbers of recurrences were in range of one to 15 with average of 3 times. Younger age at first episode didn't increase the likelihood of recurrences in our sample ($p=0.123$). Sonography was performed in 34 cases as primary investigation. Reports showed multiple hypoechoic lesions with enlarged intraglandular lymph nodes and enlarged gland. MRI was performed in 10, CT in 4, scintigraphy in 2 and biopsy of minor salivary glands in 1. The median level of C-reactive protein and erythrocyte sedimentation rate was 4 (range 0.2-55.5 mg/l, $n=30$) and 7 (0.5-76 mm in 1st hour, $n=20$), respectively. The median blood leukocyte count $8.13 \times 10^9/L$ (range 3.73-25.4 $\times 10^9/L$, $n = 36$). 20 (50%) patients had serum immunoglobulins measured, 6 of them having hypergammaglobulinaemia. One patient had a marginally low serum Immunoglobulin A (50; normal variation 55-210). Antibody screening was performed in 18 patients (45%), with 10 with positivity for antinuclear antibodies, 3 anti-SSA, 2 anti-SSB, 2 anti-RNP and 2 rheumatoid factor. One of the children had sialolithiasis in the parotid gland and other had an abscess with microaerophilic streptococcus isolated in culture of pus. Among all patients, 5 (12.5%) had a final diagnosis of a rheumatic immune-mediated disease: 3 with primary Sjögren Syndrome (SS) and 2 with mixed connective tissue disease. An idiopathic etiology was determined in 33 (82.5%). These were treated with analgesics and nonsteroidal antiinflammatory drugs with complete resolution of symptoms and the previous ones with immunosuppressants/immunomodulators.

Conclusion: The clinical, laboratory and imaging profile of our sample is in accord with the literature, except for sex predilection. Although recurrent juvenile parotitis is more common, primary juvenile SS should be considered. A high suspicion is crucial mainly because of a lower incidence of xerostomy and xerophthalmia in this population.

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SAT0501

THE IMPACT OF MORNING STIFFNESS ON THE DEFINITION OF INACTIVE DISEASE IN JUVENILE IDIOPATHIC ARTHRITIS

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Background: Juvenile idiopathic arthritis (JIA) is the most common chronic rheumatic disease in childhood. Morning stiffness is a major symptom of JIA, and is usually associated with active disease. The 2004 preliminary criteria for inactive disease (ID) in JIA did not include the assessment of morning stiffness, whereas the 2011 revision of the criteria has allowed the presence of morning stiffness (MS) lasting ≤ 15 minutes. MS was included in 2011 revision based on the consideration that MS of a short duration (i.e., ≤ 15 minutes) can represent residua of previously active disease without current active disease. However, it is unclear whether the disease status of children with ID who have or do not have morning stiffness is comparable.

Objectives: To compare the disease status of children with JIA who met the 2004 and 2011 revised criteria for ID in relation to the presence or absence morning stiffness.

Methods: A database of 1208 Italian children included in 2 multicenter studies (1,2) who underwent a total of 3380 visits was examined to identify all visits in which the patients fulfilled the 2004 or 2011 criteria for ID. In case a patient met the ID criteria in more than 1 visit, only the first visit was retained. For each visit with ID, the duration of morning stiffness was categorized as ≤ 15 min or > 15 min. Clinical assessments included demographic features and parent-reported outcomes

Results: A total of 668 visits in which patients met the criteria for ID were identified. Absence of morning stiffness was reported in 564 (84.4%) visits, whereas in 104 visits (15.5%) there was morning stiffness. Among the visits with morning stiffness, in 55 (8.2%) duration was ≤ 15 min, and in 49 (7.3%) duration was > 15 min. The table shows the comparison of disease duration and parent-reported outcomes between patients with absence or presence of morning stiffness.

	Patients meeting 2004 ID criteria			p-value
	No MS N = 564	MS ≤ 15 min N = 55	MS > 15 min N = 49	
Median [IQR] disease duration	2.2 [0.6, 6.3]	2.0 [0.8, 4.0]	3.4 [0.4, 7.3]	0.859
JAFS total score > 0	107 (19.0)	24 (43.6)	38 (77.6)	< 0.001
PRQL PhS > 1 SD*	61 (10.9)	22 (40.7)	38 (79.2)	< 0.001
PRQL PsS > 1 SD*	32 (5.8)	9 (17.0)	16 (33.3)	< 0.001
VAS DA > 0	170 (30.1)	44 (80.0)	43 (87.8)	< 0.001
VAS well-being > 0	211 (37.4)	42 (76.4)	44 (89.8)	< 0.001
VAS pain > 0	140 (24.8) 484	39 (70.9) 31	35 (71.4) 20	< 0.001
Remission	(87.5)	(59.6)	(41.7)	< 0.001

MS: morning stiffness; IQR: interquartile range, *above the mean of healthy children (2)

Conclusion: Among patients who met the 2011 criteria for ID, those with morning stiffness ≤ 15 min had worse parent-reported outcomes than those without morning stiffness. This finding suggests that parents may not perceive their child's disease state as true remission when lower degrees of morning stiffness are present. Notably, a sizeable proportion (7.3%) of children meeting the 2004 ID criteria had morning stiffness lasting > 15 min. The removal of the criterion "Duration of morning stiffness of ≤ 15 minutes" to "Absence of morning stiffness" in the definition for ID should be considered.

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