



Review Article

Juvenile recurrent parotitis: A systematic review of treatment studies[☆]Werner Garavello^{a,*}, Monica Redaelli^b, Francesca Galluzzi^b, Lorenzo Pignataro^c^a Department of Otorhinolaryngology, School of Medicine and Surgery, University of Milano-Bicocca, Milan, Italy^b Department of Otorhinolaryngology, San Gerardo Hospital, Monza, Italy^c Department of Otorhinolaryngology, Department of Clinical Sciences and Community Health, University of Milan, Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico, Milan, Italy

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ABSTRACT

Juvenile recurrent parotitis is a rare recurrent inflammation of the parotid glands occurring in children. The etiology remains obscure and the treatment is still debated. In the present study, we perform a systematic review of the literature with the purpose of identifying and discussing the treatment options emerged over the last 28 years in order to prevent recurrent episodes of parotitis.

We ultimately included 24 studies. The definitions used for juvenile recurrent parotitis varied widely and none of the selected studies referred exactly to the same definition. Only one was a randomized controlled trial and it showed marked benefits with the use of Bear Bile and Huangqi, two traditional Chinese medicines. Two additional study on sialendoscopy included a control group but was not randomized. All the remaining contributions were case series or case reports. The vast majority (n = 19) of the selected studies reported on sialendoscopy. They all documented improvement of the condition following this intervention. An analysis grouping all these studies (corresponding to 336 children) showed that only 25.8% (95% Confidence Interval: 21.5–30.8) of the treated children had further recurrences. However, the only two controlled study on sialendoscopy showed a similar improvement in controls. The remaining four studies were on sialography (n = 2), on oral appliance in the specific group of children with concomitant dental malocclusion (n = 1) on ductal hydrocortisone infusion through catheter inserted in the parotid duct (n = 1). Improvements were documented in all four contributions.

This systematic review of the literature did not consent us to draw definite conclusions on the most suitable treatment for juvenile recurrent parotitis. The available evidence is indeed weak and difficult to interpret because of the scarcity of randomized controlled trials, the heterogeneity of the definitions used and the high rate of spontaneous resolution. Future large and well-designed randomized controlled trials that will include children fulfilling a shared definition of the condition are warranted.

1. Introduction

Juvenile recurrent parotitis (JRP) is a rare recurrent inflammation of the parotid glands occurring in children. It is the second most common inflammatory gland disease of childhood, after mumps [1,2]. JRP is characterized by recurrent episodes of painful parotid swelling, often associated with fever and malaise [3]. JRP is mostly unilateral even if, in less common cases, it can occur bilaterally, usually with a more predominant side. The usually age of onset is 3–6 years, and there is a tendency for spontaneous remission of the disease near the onset of puberty or late adolescence [4,5]. In a minority of cases, JRP is asso-

ciated with local favoring conditions such as dental malocclusion and sialolithiasis [6] or with systemic diseases such as the Sjögren syndrome and selective IgA deficiency [7–9]. JRP is a well-recognized condition but the etiology remains obscure (at least in idiopathic cases) and the treatment is still debated [10]. The treatment of the acute episodes aims to relieve symptoms and prevent damage to gland parenchyma. Most commonly, the condition is treated with a symptomatic approach of the relapses and using painkillers, corticosteroids and antibiotics with various regimes [11]. Considering these latter their use is controversy, indeed there is no evidence that antibiotic agents affect the duration of the acute episodes and long-term prophylactic dose could prevent re-

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* Corresponding author. Department of Otorhinolaryngology, Ospedale San Gerardo, Via Pergolesi, 33, 20052, Monza (MI), Italy.
E-mail address: werner.garavello@unimib.it (W. Garavello).

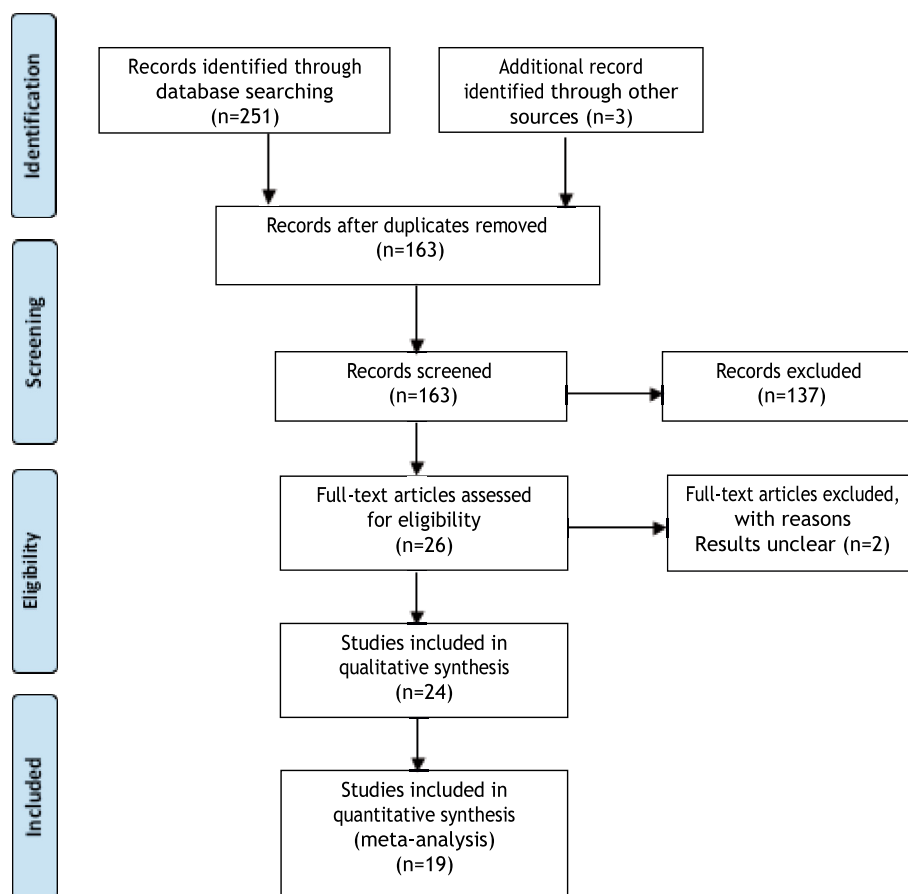


Fig. 1. Flow diagram of the identified studies.

currences. Some authors have proposed other conservative therapies such as massages of the gland, encouragement of fluid intake, hot compresses, mouth rinses, sialogogues or the use of chewing gum [6,10]. However, given the relevant impact on the quality of life of the affected children and their parents, there is the pressing need for more effective treatments, in particular for treatments aimed at eradicating the condition and preventing recurrences. In the present study, we perform a systematic review of the literature with the purpose of identifying and discussing the treatment options emerged over the last 28 years in order to prevent recurrent episodes of parotitis.

2. Methods

We searched PUBMED for articles published in the English language between January 1990 and April 2018 using the following MeSH search terms: “recurrent”, “parotitis” and “child OR children OR childhood OR pediatric OR juvenile” with restriction to the human species. Data was extracted independently by two investigators (W.G. and M.R.) who also performed an initial screening of the titles and abstracts of all articles to exclude citations deemed irrelevant to both observers. Reference lists of the selected articles and from other reviews on JRP were also evaluated. A binomial distribution model was used to calculate the 95% Confidence Interval (CI) of proportions.

3. Results

The PRISMA flow diagram of the identified studies is shown in Fig. 1 [12]. We ultimately included 24 studies (Table 1). They were all published during the last ten years [11,13–35]. Only one of the selected studies was a randomized controlled trial (RCT) [23]. This study investigated the benefits of the use of Bear Bile and Huangqi, two traditional Chinese medicines. Two studies included a control group but were not randomized [22,29]. All the remaining contributions were case series or case reports [11,13–21,24–28,30–35]. The vast majority reported on sialendoscopy [11,13,15,17–22,24–26,28–31,33–35] (19 out of 24, corresponding to 79%), two studies were on sialography [16,27], one Bear Bile and Huangqi, one on oral appliance in the specific group of children with concomitant dental malocclusion and one on ductal hydrocortisone infusion through catheter inserted in the parotid duct [14,23,32]. The definitions used varied widely among studies and none referred exactly to the same definition (Table 1). Table 2 shows the frequency of the different criteria used.

The characteristics of the populations studied and the main findings are summarized in Table 3. The number of included children varied widely, from 1 to 840. All studies report a beneficial effect of the studied intervention with the exception of Schneider et al. [22] and Rosbe et al. [29] who observed improvement following interventional sialendoscopy but reported similar benefits in the control group treated

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