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REGULAR ARTICLE

Is recurrent parotitis in childhood still an enigma? A pilot experience

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Abstract

Aim: To test the hypothesis that dental malocclusion with mandibular misplacement may be a causative factor for recurrent parotitis (RP) through unbalancing of masticatory muscles.

Methods: Thirteen patients (age 4–14 years) who were referred to a dental clinic for RP and malocclusion were treated by oral appliance positioning for a 6-month period. Monthly visits were scheduled regularly.

Results: Symptoms were clearly improved in nine children. No effect was obtained in three patients. One patient was lost at follow-up.

Conclusion: Occlusal intervention is effective in patients with RP and associated malocclusion. It should be considered an important option for the treatment of such intriguing disorder.

. . . the aetiology of recurrent parotitis remains an enigma (1).

Recurrent parotitis (RP) is a rare, intriguing inflammatory condition of unknown aetiology. It is characterized by recurrent episodes of swelling and/or pain of the parotid gland, usually accompanied by fever and malaise (2). School-age children are mainly affected, but symptoms disappear at puberty in the majority of cases (3). Management of RP is conservative, and aggressive surgical treatment should be reserved only for adults with severe persistent problems (4).

Dental occlusal disorders, which can increase muscle tension, are recognized to play an important role in the pathogenesis of several diseases, and interocclusal appliance, which may induce muscle relaxation, has been successfully employed for treatment of many conditions (5).

So far, the prevalence of malocclusion among patients with RP has not been established. In our personal experience, children with RP usually manifest mandibular misplacement. Therefore, our primary hypothesis was that RP is the consequence of a dysfunctional disorder caused by dental malocclusion. This hypothesis was tested by evaluating the effect of the occlusal intervention on the recurrence of symptoms of parotitis in patients with RP and concomitant malocclusion.

PATIENTS AND METHODS

Thirteen children (aged 4–14 years; seven males and six females) with a history of RP and suspected malocclusion were referred to the dental clinic (EB) by paediatricians who had attended national educational courses on craniomandibular disorders over the previous 3 years.

The duration of the symptoms ranged from 2 months to 8 years (mean: 2.1 years), and the frequency of attacks varied from twice a month to four times a year. One patient had a history of persistent swelling of the parotid gland. Three children also presented symptoms of temporomandibular disorders.

Ultrasonography was obtained in 10 children, which revealed hypoechoic areas of the parotid gland in all of them. Computerized tomography and magnetic resonance imaging were performed in two subjects; they showed increased density and inhomogeneous, patchy changes of parotid parenchyma, respectively. All patients had previously received various medical treatments with no success. Due to frequent relapses of purulent infection, surgical incision of the parotid gland had been obtained in two cases.

On recruitment, patients underwent orthodontic assessment and clinical inspection of mandibular posture on the three spatial planes (sagittal, horizontal and frontal) to detect possible jaw deviation from normal occlusion: deep bite, retruded mandible and cross-bite. To evaluate the contact relationship of the occlusal surfaces of the upper and lower teeth and to obtain the information to prepare a personalized oral appliance, an alginate impression was taken of the dental arches, as it has been previously described (6). Dental stone models were made from impressions of the teeth and were transferred in an articulator in a relationship individually determined for each of the three spatial shifts connected with the mandible repositioning. The relationship was chosen by the orthodontists (VB, EB) of our team through the registration of a wax check-bite directly in the patient's mouth. The interocclusal relationship for each of the three spatial shifts connected with the mandible repositioning was determined. The vertical shift aimed to obtaining 1-mm overbite between the antagonist central incisors; the sagittal shift aimed to obtaining 1- to 2-mm overject and the lateral shift aimed to lining up the upper and lower

Figure 1 First phase: mandibular positioning by application of the oral device.

Figure 2 Second phase: the device was modified to allow orthognatodontic movements, while maintaining mandible positioning.

median labial fraenulum.Any possible dental misplacement was considered, when determining the above-mentioned parameters.

An individualized acrylic oral splint was realized and applied to each patient (Fig. 1). The device was endowed with orthodontic clamps, which fixed it to the teeth of the lower dental arch and could be easily removed by the patient. It consists of an occlusal bite plane set between the two antagonist dental arcades, which modifies the vertical dimension, and a repositioning wall, which works over the vestibular surfaces of the upper incisors and canines. This repositioning wall modifies both the sagittal and the lateral relationship between the maxilla and the mandible. Patients were required to wear the appliance continuously, except at mealtimes.

For the purpose of the study, a 6-month treatment was established for each patient, with monthly assessment by the orthodontist to monitor functioning of the device. The total duration of splint positioning depended on the severity of the disease, the benefits obtained and the patient's acceptability. During monthly assessment, tolerance was evaluated by the orthodontic specialist who questioned the patient about difficulty in using the oral appliance as prescribed.

If symptoms had disappeared after 6-month positioning of the device, orthodontic treatment was started according to individual need. The resolution of symptoms of RP during the device treatment phase justifies anticipating the orthodontic treatment during the deciduous dentition. In case of no resolution of symptoms, timing of orthodontic treatment is postponed to the dentition period.

During the orthodontic treatment – which was out of purposes of the present study – visits were scheduled on regular basis until the end of the treatment. The oral device was modified at each visit to allow orthodontic movements of the teeth (Fig. 2), and it was definitively removed when the mandible–maxilla position was considered to be stable, that is (i) a solid intercuspation of teeth was obtained without mandibular dislocation, deviation or shift during closure; (ii) there was no interference during mandibular centrifugal movements; and (iii) noxious habits disappeared.

The length and type of this second phase are related to the individual patient and the response to treatment. The defined goal is that of every interceptive orthodontic treatment (i.e. the resolution of the most important occlusal problems such as the increased overbite, the class twoincisors relationship and the lateral cross-bite). The third phase of orthodontic treatment, the final one, will be postponed to the ideal 12–14 years of age.

The small number of cases of juvenile RP precluded the possibility of dividing them into a study group and a control group, as it would be preferable.

RESULTS

Demographic and clinical characteristics of the patients are shown in Table 1. Different types of malocclusion were present: increased overbite, retruded mandible and jaw deviation were found in three, six and seven patients, respectively.

One patient (no. 11) was lost during the treatment phase. During the 6-month treatment period, eight patients did not report symptoms suggestive for parotitis; one patient (no. 4) presented acute symptoms some weeks after starting treatment, but no further episodes occurred subsequently; no significant improvement was obtained in three patients (no. 1, 6, 10). Interestingly, two of them had received surgery for frequent relapses of purulent parotitis.

The oral appliance was tolerated by all subjects. During the initial phase of treatment, some children complained of excessive salivation which completely disappeared after few days. No temporomandibular joint discomfort was reported.

DISCUSSION

Despite several pathogenetic hypotheses have been suggested and new insights have been proposed over the years, the cause of RP remains an enigma (7). RP is generally associated with reduced salivary flow and nonobstructive sialectasis; however, it is not clear if sialectasis is the cause or rather the effect of the infection ascending from the mouth (8).

Management of RP is traditionally restricted to treatment of acute attacks, usually with antibiotics and steroids. Unfortunately, these drugs do not change the natural course of the disease (3). Although a recent study demonstrated that a combined endoscopic approach composed of lavage, ductal dilatation and hydrocortisone injection is effective in children with RP (9), there are no defined indications for

Table 1 Demographic and clinical characteristics

Pt no.	Age (years)/ Sex	Duration of symptoms	Frequency of episodes	Side of parotitis	Occlusal problem	Evolution	Follow-up
1	11/F	8 years	Monthly	Bilateral	Retruded mandible	Failure	
2	6/M	5 years	Monthly	Right	Right deviation	Symptoms disappeared	49 months
3	8/M	2 years	Every 1–2 months	Bilateral, right prevalence	Retruded mandible	Symptoms disappeared	34 months
4	4/M	12 months	Every 3–4 months	Right	Right deviation	Single relapse	25 months
5	10/F	3 years	Every 2–3 months	Right	Deep bite, right deviation	Symptoms disappeared	36 months
6	4/F	6 months	Every 2 weeks	Bilateral, right prevalence	Mild deep bite	Failure	
7	14/F	2 years	Every 1–2 months	Right	Right deviation	Symptoms disappeared	18 months
8	6/F	18 months	Every 1–2 months	Right	Right deviation	Symptoms disappeared	22 months
9	5/M	14 months	Every 1–2 months	Bilateral	Retruded mandible	Symptoms disappeared	24 months
10	9/F	12 months	Every 1–2 months	Right	Deep bite, retruded mandible, right deviation	Failure	
11	4/M	12 months	Every 1–2 months	Left	Left deviation	Lost at the follow-up	
12	7/M	12 months	Monthly	Bilateral	Retruded mandible	Symptoms disappeared	24 months
13	5/M	2 months	Persistent parotid swelling	Bilateral	Retruded mandible	Symptoms disappeared	12 months

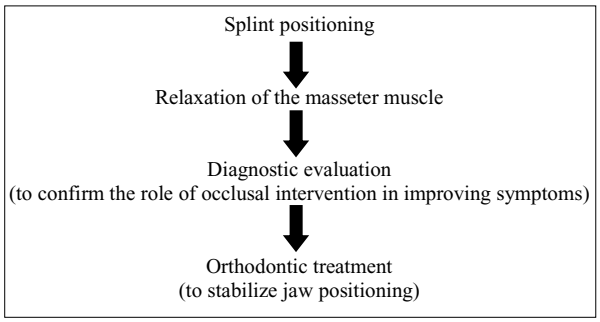

Figure 3 Diagnostic and therapeutic algorhythm in recurrent parotitis.

the prevention of relapses. Controversy also exists on timing and type of surgical intervention, which may be required for the most severe cases in adulthood (10).

The rationale of our study was based on the hypothesis that a dysfunctional disorder caused by malocclusion is responsible for RP. Under optimal anatomic and physiologic circumstances, there is a functional balance between occlusion, muscles of mastication and joint structures (functional bite relationship). Mandible displacement due to malocclusion may induce an abnormal tone of the masseter muscle, which maintains a close anatomic connection with the parotid gland. In particular conditions (i.e. during a meal), the increased activity of the masseter muscle may provoke intermittent obstruction of the Stenson's duct, and reduction of the salivary flow may therefore occur. In children with cross-bite, the reduced salivary flow may also be the consequence of the decreased swallowing activity, which has been demonstrated in such condition (11).

Our study was the first to evaluate the effect of an oral device in children with RP and concomitant malocclusion. The recognition of malocclusion is considered crucial for early diagnosis of temporomandibular joint disorders. If imbalance of the functional bite relationship is revealed, a diagnostic orthotic is an essential tool to help determine a better functional relationship in order to avoid system compromise and to produce muscle relaxation (12). Therefore, the benefit of a splint appliance is the ability to establish a reversible diagnostic relationship, to evaluate the tissue and system response over time and to determine if the new position is better for the patient than the habitual bite association (12) (Fig. 3).

In the last 10 years, different devices have been used for the treatment of conditions such as headache, temporomandibular joint pain and obstructive sleep apnoea syndrome (5,6). Despite the claimed effectiveness of such approach, comparing the results is difficult due to differences between methods and the subjective evaluation of the outcome (5). Most of our patients benefited from the occlusal intervention. How did it work? We suggest that the interocclusal appliance may interact with the pathogenetic mechanisms of RP by blocking the cascade of events due to mandible displacement. It may improve the masseter muscle relaxation, thus avoiding the transitory parotid duct obstruction and then restoring the normal salivary flow (Fig. 4).

Indeed, decreased myoelectrical activity of masticatory muscles has been found after positioning of an occlusal splint, either in patients with bruxism (13) or in subjects with masticatory dysfunction (14). As occlusal splints are able to stimulate salivary secretion, particularly during chewing-like movement in both bruxism and normal subjects (15), increased salivary secretion per se should not however be overlooked as potential or concomitant therapeutic mechanism.

The high prevalence of unilateral symptoms in patients with RP (3,16) has been confirmed in our study. Unilateral gland involvement was found in seven patients, and a side prevalence of symptoms was evident in two of six children with bilateral involvement. It should be emphasized that in cross-bite circumstances, the masseter muscle tone is increased only on the side of mandible deviation. This finding may well justify the unilateral involvement of the

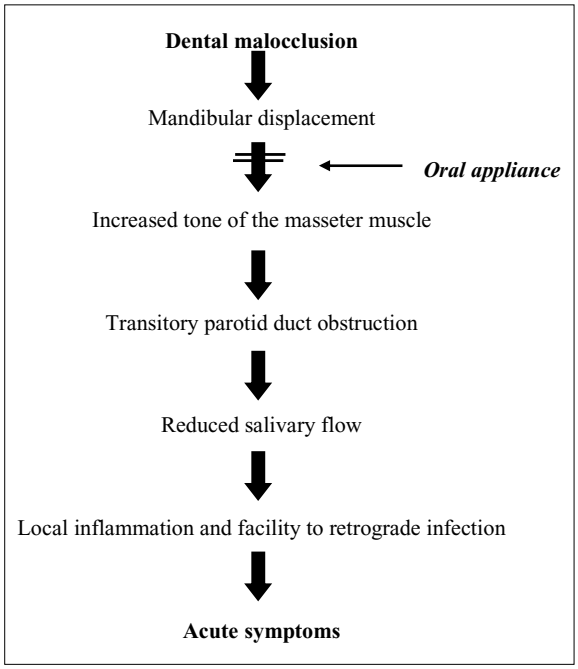

Figure 4 Pathogenetic mechanisms for recurrent parotitis and the effect of oral appliance.

parotid gland. On the other hand, coexistence of deep bite or retruded mandible with cross-bite might explain the prevalence of unilateral symptoms in subjects with involvement of both glands (17). Higher rate of secretion and relatively richer mucus composition with antiseptic properties of the submandibular gland in comparison with the parotid gland have been claimed to explain the lack of submandibular involvement in RP (2,8).

Several authors reported that symptoms of RP usually subside after puberty (3,8,17), but the reason is unknown. This epidemiological aspect is difficult to reconcile with our primary hypothesis, since malocclusion is prevalent to a higher degree in permanent dentition. However, we infer that physiological growth of the jaw ramus may pull away the masseter muscle insertions on the mandible and zygomatic arch, thus reducing the effect of muscular contraction on the salivary duct in patients with mandibular displacement. Occasionally, symptoms of RP persist after puberty (3,17,18) and even occur in adulthood (1,3). The lack or incorrect alignment of natural teeth and/or inappropriate prosthodontic treatment might modify the mandibular–maxillary relationship, thus reproducing a condition which is functionally similar to malocclusion in childhood.

From a scientific point of view, the major bias of our study is that it was not controlled. We are aware that it would be preferable to evaluate also a control group; however, we thought it was unethical to perform such study, because symptoms had persisted over long time despite common treatments. Descriptive studies often represent the first scientific toe in the water in new areas of inquiry (19), and in our study we made every effort to respect the fundamental elements of descriptive reporting. We nonetheless hope that our pilot experience may act as a stimulus for future well-designed, controlled studies.

In conclusion, the benefits obtained with a personalized oral jaw-positioning appliance in patients with RP and malocclusion seem to be striking in our experience. If this effect is due to decreased myoelectrical activity of masticatory muscles or due to increased salivary secretion after positioning of an occlusion splint remains debatable. We therefore suggest that the orthodontic evaluation should be an integral part of the clinical assessment in patients with RP. If malocclusion is present, occlusal intervention should be properly considered. Further studies are necessary to evaluate the long-term effect of the definitive orthodontic treatment to obtain occlusal stability in such patients.

References

1. Chitre VV, Premchandra DJ. Recurrent parotitis. *Arch Dis Child* 1997; 77: 359–63.
2. Leerdam CM, Martin HC, Isaacs D. Recurrent parotitis of childhood. *J Paediatr Child Health* 2005; 41: 631–4.
3. Geterud A, Lindvall AM, Nylen O. Follow-up study of recurrent parotitis in children. *Ann Otol Rhinol Laryngol* 1988; 97: 341–6.
4. Motamed M, Laugharne D, Bradley PJ. Management of chronic parotitis: a review. *J Laryngol Otol* 2003; 117: 521–6.
5. Major PW, Nebbe B. Use and effectiveness of splint appliance therapy: review of literature. *Cranio* 1997; 15: 159–66.
6. Villa MP, Bernkopf E, Pagani J, Broia V, Montesano M, Paggi B, et al. Randomized controlled study of an oral jaw positioning appliance for the treatment of obstructive sleep apnea in children with malocclusion. *Am J Respir Crit Care Med* 2002; 165: 123–7.
7. Kolho KL, Saarinen R, Paju A, Stenman J, Stenman UH, Pitkaranta A. New insights into juvenile parotitis. *Acta Pediatr* 2005; 94: 1566–70.
8. Ericson S, Zetterlund B, Ohman J. Recurrent parotitis and sialectasis in childhood. Clinical, radiologic, immunologic, bacteriologic, and histologic study. *Ann Otol Rhinol Laryngol* 1991; 100: 527–35.
9. Nahlieli O, Shacham R, Shlesinger M, Eliav E. Juvenile recurrent parotitis: a new method of diagnosis and treatment. *Pediatrics* 2004; 114: 9–12.
10. Nahlieli O, Bar T, Shacham R, Eliav E, Hecht-Nackar L. Management of chronic recurrent parotitis: current therapy. *J Oral Maxillofac Surg* 2004; 62: 1150–5.
11. Ingervall B, Thilander B. Activity of temporal and masseter muscles in children with a lateral forced bite. *Angle Orthod* 1975; 45: 249–58.
12. Keller DC. Diagnostic orthotics to establish the functional mandibular-maxillary relationship for orthodontic corrections. *J Gen Orthod* 2001; 12: 21–5.
13. Harada T, Ichiki R, Tsukiyama Y, Koyano K. The effect of oral splint devices on sleep bruxism: a 6-week observation with an ambulatory electromyographic recording device. *J Oral Rehabil* 2006; 33: 482–8.
14. Freeland TD. Muscle function during treatment with the functional regulator. *Angle Orthod* 1979; 49: 247–58.
15. Miyawaki S, Katayama A, Tanimoto Y, Araki Y, Fujii A, Yashiro K, et al. Salivary flow rates during relaxing, clenching and chewing-like movement with maxillary occlusal splints. *Am J Orthod Dentofacial Orthop* 2004; 126: 367–70.

16. Sitheeque M, Sivachandran Y, Varathan V, Ariyawardana A, Ranasinghe A. Juvenile recurrent parotitis: clinic, sialographic and ultrasonographic features. *Int J Paediatr Dent* 2007; 17: 98–104.
17. Galili D, Marmary Y. Juvenile recurrent parotitis: clinical-radiologic follow-up study and beneficial effect of sialography. *Oral Surg Oral Med Oral Pathol* 1986; 61: 550–6.
18. Mandel L, Kaynar A. Recurrent parotitis in children. *N Y State Dent J* 1995; 61: 22–5.
19. Grimes DA, Schulz KF. Descriptive studies: what they can and cannot do. *Lancet* 2002; 359: 145–9.