

Bilateral Juvenile Parotitis: A rare bilateral location

Caner Sahin¹

¹Sakarya Akyazi State Hospital, ENT Clinic, Sakarya, Turkey.

Corresponding author: Caner Sahin, MD;

Address: Sakarya Akyazi State Hospital ENT Clinic, Sakarya, Turkey;

Fax: +902644187835; E-mail: drcaner2001@gmail.com

Abstract

Juvenile recurrent parotitis is an inflammation of the parotid gland of unknown cause in children. This disease is mostly unilateral although rarely can be traced bilaterally.

Herein we present a 5-year old boy with bilateral juvenile recurrent parotitis hospitalized at Sakarya Akyazi State Hospital, Turkey.

Keywords: juvenile, parotitis, Sjogren's disease.

Introduction

Juvenile recurrent parotitis (JRP) is an inflammation of the parotid gland of unknown cause in children (1). Unlike suppurative parotitis, purulent drainage patients are not monitored (2). This is a rare disease characterized by swelling of the parotid gland several times within a year. The disease is mostly unilateral although rarely can be traced bilaterally. In most cases, symptoms disappear or reduce in frequency in the period of puberty. The disease rarely continues into adulthood (3).

Symptoms usually begin around the age of three years and last for 3-7 days. During the attack, the parotid gland is swollen, redness occurs and the diagnosis can be mixed with mumps (4).

Herein we present a 5-year old boy with bilateral JRP in light of the respective scientific literature.

Case report

A 5-year old boy was admitted with complaints of developing bilateral parotid gland tenderness and swelling attacks to our clinic (Sakarya Akyazi State Hospital, Turkey) for the last two years. He had seven attacks in the first year and five attacks in the second year. In each attack, the patient consulted a doctor within 7-10 days and symptoms declined after medical treatment. He had no symptoms of dry mouth and dry eye. The patient had no skin-related complaints, no history of

allergies, swelling of joints, or blurred vision problems. Also, in the family there was no one person with similar symptoms. Between attacks, the patient stated that the general condition was completely normal.

Upon physical examination during the attack, there was evidence of swelling in both sides of the parotid gland of the patient. The lesion was highly sensitive upon palpation. There was no purulent secretion with pressure in the parotid gland region. The external examination of the patient did not reveal any pathology.

The patient's complete blood was drawn for routine biochemical analysis including erythrocyte sedimentation rate, immunoglobulin values, amylase, high specific rf, Sjogren's syndrome, anti-Ro and anti-La antibodies. An increase in the number of white blood cells was observed in blood tests (Table 1). Other biochemical values were within normal limits.

The patient underwent bilateral parotid ultrasonography. Ultrasonography revealed bilateral parotid gland swelling; there was no solid mass or Stone. Ibuprofen, ampicillin-sulbactam treatment and fluid intake were performed to the patient. The attack ended after seven days of treatment. The patient had two more attacks in the following six months. These attacks also resolved within a week. The patient was referred for sialography.

Table 1. Laboratory parameters of the patient

Laboratory Values	
Hemogram	12.5 g/Dl (11-16 g/Dl)
Leukocyte	12.3 x10 mm ³ (4.6-10.2 x 10 mm ³)
Lymphocyte	6.1 x10 mm ³ (0.6-3.4 x 10 mm ³)
Neutrophil	8.2 x10 mm ³ (2-6.92 x 10 mm ³)
Sedimentation	39 mm/hour (0.1-20 mm/hour)
Amylase	89 u/L (28-100 u/L)
Anti-Ro	Negative
Anti-La	Negative

Discussion

Juvenile parotitis is one of the commonly encountered diseases in childhood. Symptoms of the disease usually start at the age of 3-6 years. This condition may be confused with diseases such as otitis media and mumps. The disease is usually monitored more frequently in men. It is usually unilateral and rarely encountered as bilateral. Frequency of attacks can vary between patients from 3-15 times per year. Swelling of parotid region is common during attacks of this disease. A decrease in symptoms and attacks is usually observed until puberty. Possibility of a decrease in saliva at older ages can be observed due to the destruction of the ducts during the attacks in 50%-70% of the cases (3). Most of the cases are idiopathic, but in rare cases Sjogren's syndrome or immune deficiency is the underlying cause of the disease.

The pathophysiology of the disease is multifactorial. Congenital ductal malformations, autoimmune disorders, viral diseases, and genetic factors are blamed for development of the disease. Decrease in saliva, stasis in duct system and emerging infectious are consequences of this disease. Dilation of peripheral ducts of gland, referred to as sialectasis, can be observed upon histological examination (3). It has been reported that the disease is both congenital and a combined condition as a result of a decrease in saliva production.

Diagnosis of the disease is mainly based on the recurrence of attacks, ultrasonography, sialography and ruling out underlying diseases (4). Sialoendoscopy is being increasingly used both for

diagnosis and treatment (5).

The treatment of the disease differs from conservative treatment up to surgical approaches. Conservative approach is recommended in cases where the number of attacks is not too high. During attacks, analgesics, fluids, antibiotics and massage treatments to the parotid region can be used. Usage of antibiotics may prevent infections and damages of ductus of the parotid region (5).

Usage of low-dose antibiotics is recommended in children with immunoglobulin deficiency (6). Sialoendoscopy can be used both for diagnosis and treatment. It is recommended for cases with more than five attacks in the scientific literature (7-9). Sialoendoscopy has disadvantages of being expensive, requires expensive equipment and there are few centers performing this procedure. Surgeons can observe strictures, narrow and wide sides of ductus. Lavage of ductus, dilatation, intraductal oil injection and intraductal application of corticosteroids can be made during the process (10). Yet, the number of cases reported in the literature is limited (11,12). Developing Sjogren's disease in later life is not uncommon in patients with JRP (13).

In conclusion, JRP is a rare disease reported in the literature. Underlying immunological diseases and Sjogren's disease should be assessed in patients with JRP (14). Diagnosis and treatment of patients with JRP is performed with sialoendoscopy. However, sialoendoscopy has disadvantages of being expensive, requires expensive equipment and there are few centers performing this procedure. Finally, patients with JRP should be monitored at regular intervals in collaboration with pediatricians.

Conflicts of interest: None declared.

References

1. Kolho KL, Saarinen R, Paju A, Stenman J, Stenman UH, Pitkäranta A. New insights into juvenile parotitis. *Acta Paediatr* 2005;94:1566-70.
2. Leerdam CM, Martin HC, Isaacs D. Recurrent parotitis of childhood. *J Paediatr Child Health* 2005;41:631-4.

3. Nahlieli O, Shacham R, Shlesinger M, Eliav E. Juvenile recurrent parotitis: A new method of diagnosis and treatment. *Pediatrics* 2004;114:9-12.
4. Ericson S, Zetterlund B, Ohman J. Recurrent parotitis and sialiectasis in childhood: Clinical, radiologic, immunologic, bacteriologic, and histologic study. *Ann Otol Rhinol Laryngol* 1991;100:527-35.
5. Gadodia A, Seith A, Sharma R, Thakar A. MRI and MR sialography of juvenile recurrent parotitis. *Pediatr Radiol* 2010;40:1405-10.
6. Ramakrishna J, Strychowsky J, Gupta M, Sommer DD. Sialendoscopy for the management of juvenile recurrent parotitis: a systematic review and meta-analysis. *Laryngoscope* 2015;125:1472-9.
7. Akar HH, Patýroglu T, Duman L. A selective IgA deficiency in a boy who presented recurrent parotitis. *Eur J Microbiol Immunol (Bp)* 2014;4:144-6.
8. Semensohn R, Spektor Z, Kay DJ, Archilla AS, Mandell DL. Pediatric sialendoscopy: initial experience in a pediatric otolaryngology group practice. *Laryngoscope* 2015;125:480-4.
9. Mikolajczak S, Meyer MF, Beutner D, Luers JC. Treatment of chronic recurrent juvenile parotitis using sialendoscopy. *Acta Otolaryngol* 2014;134:531-5.
10. Ardekian L, Klein H, Al Abri R, Marchal F. Sialendoscopy for the diagnosis and treatment of juvenile recurrent parotitis. *Rev Stomatol Chir Maxillofac Chir Orale* 2014;115:17-21.
11. Schneider H, Koch M, Künzel J, Gillespie MB, Grundtner P, Iro H, et al. Juvenile recurrent parotitis: a retrospective comparison of sialendoscopy versus conservative therapy. *Laryngoscope* 2014;124:451-5.
12. Katz P, Hartl DM, Guerre A. Treatment of juvenile recurrent parotitis. *Otolaryngol Clin North Am* 2009;42:1087-91.
13. Flaitz CM. Parotitis as the initial sign of juvenile Sjogren's syndrome. *Pediatr Dent* 2001;23:140-2.
14. Miziara ID, Campelo VE. Infantile recurrent parotitis: Follow-up study of five cases and literature review. *Braz J Otorhinolaryngol* 2005;71:570-5.