

# Management of Recurrent Juvenile Parotitis Complicated with Parotid Abscess and Fistula: A Case Series



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## Abstract

**Background:** Juvenile recurrent parotitis (JRP) cause huge impact in children as it led to poor feeding during attacks and reduce quality of life. Children will experience painful parotid swelling and fever. It is known to be self-limiting, however may progress to chronic parotitis. It usually resolves after puberty without significant sequelae. Average recurrence rate in affected children is about 7 times per year. Sequelae of parotitis in children infrequently progress to suppurative phase and extremely rare to develop parotid fistula following parotid abscess in this group of patients. We report 8 cases of JRP complicated with parotid abscess and fistula which eventually resolved with conservative measures. A case series was reviewed in a tertiary academic institution from September 2019 until December 2022. All children with recurrent parotid infection managed during that period of time were recruited. They were treated by the same paediatric otorhinolaryngology (ORL) consultant.

**Results:** There were 8 children in this series in which 3 of them developed parotid fistulas after undergoing incision and drainage. All of the fistulas resolved within four months with conservative treatment. Two patients required ultrasound surveillance to monitor the progression of parotid swelling, which later resolved without the need for further intervention. All children were successfully discharged after one year of follow-up when there was no sign and symptoms of recurrence.

**Conclusion:** Juvenile recurrent parotitis can be successfully treated by conservative measures such as emphasizing adequate hydration and oral hygiene. Incision and drainage if needed may cause fistula but eventually resolved without further surgical intervention.

**Keywords:** Parotitis; Case series; Abscess; Fistula; Children

**Abbreviations:** JRP: Juvenile Recurrent Parotitis; ORL: Otorhinolaryngology; TB: Tuberculosis; M: Male; F: Female; I&D: Incision and Drainage; HPE: Histopathological Examination; C&S: Culture and Sensitivity; WCC: White Cell Count; AFB: Acid Fast Bacilli; PAS: Periodic Acid-Schiff; ENA: Extractable Nuclear Antibody; C3: Complement 3; C4: complement 4; RF: Rheumatoid Factor; ANA: Antinuclear Antibody; NA: Not Available; USG: Ultrasonography Neck; CT: Computed Tomographic; MRI: Magnetic Resonance Imaging; CECT: Contrast-Enhanced Computed Tomography; mm: millimeter; cm: centimeter

## Introduction

Recurrent parotitis in children is a rare scenario. Early detection of Juvenile recurrent parotitis (JRP) will improve patient's prognosis as it could be sentinel sign of immunological or autoimmune diseases [1]. JRP affects mostly boys between 3 and 6 years old with a predilection seen among males with the male to female ratio of 2:1 [2]. Reduction in salivary flow and associated with ascending bacterial invasion the ductal system may contribute to one of the factor recurrent inflammation [3]. The exact aetiology remains unknown and is deemed multifactorial. These children develop recurrent painful parotid swellings associated with fever. Among these rare cases, it was predominantly unilateral parotid

involvement, but bilateral parotid involvement has been reported by Sawant et al. [4] and, Gardner et al. [5]. It usually resolves after puberty without significant sequelae unless systemic morbidity such as immunodeficiency, lymphoma or Sjogren syndrome is present [6]. Parotid abscess rarely occurs in children. Parotid fistula is a known complication of parotid abscess but in children, it is extremely rare [7]. We report management outcomes of eight cases of JRP from our medical Center.

## Methods

The study design consisted of a retrospective case series at a single tertiary academic institution. Patients who presented

with history of recurrent parotid infections to our centre either new cases or from another center for further management were included. Informed consents were obtained from legal guardians. Data were collected from September 2019 until December

2022 including demographic, clinical features and management outcomes. Incisional and drainage (I&D) that was performed in patients were a small incision on the most fluctuant area rather than a Modified Blair incision.

Results

Clinical Presentations

Table 1: Summary of patients' data.

Case	Age (Years) and gender	Lesion	Duration of illness (months)	Treatment		Complication	Outcome at 1 year of follow up
				Antibiotic	Surgical management		
1	3, M	Unilateral	6	Yes	I&D	Fistula	Resolved
2	4, F	Unilateral	6	Yes	I&D	Fistula	Resolved
3	10months, F	Unilateral	3	Yes	I&D	No	Resolved
4	4, M	Unilateral	8	Yes	I&D	No	Resolved
5	2, F	Unilateral	12	Yes	I&D	No	Resolved
6	5, F	Unilateral	3	Yes	I&D	No	Resolved
7	4, M	Bilateral	24	Yes	I&D	No	Resolved
8	6, F	Bilateral	36	Yes	I&D	Fistula	Resolved

Abbreviations: M: Male, F: Female, I&D: Incision and drainage

Juvenile recurrent parotitis in our case series comprised of eight children, ranged from 10 months old to 6 years old. All children completed their childhood vaccinations. All of them had no comorbidities. Most of them presented with recurrent unilateral cheek or neck swelling. However, we identified two cases with recurrent bilateral parotid swellings. Besides, three out of eight children were complicated by a fistula following the surgical procedure, which resolved completely in less than four months. All eight children's findings were summarized in Table 1.

In our series, the youngest patient was a 10-month-old female with a background of recurrent left parotid abscess, which required I&D. She presented with persistent left cheek swelling that was painful, erythematous, and caused fever. Clinically, the cheek swelling fluctuated and was tender on palpation. She was treated for a recurrent parotid abscess in three months with a series of imaging procedures and completed multiple courses of antibiotics. More than 50% of cases presented with painful swelling; however, a 4-year-old male in Case 4 presented with painless, persistent right pre- and post-auricular swelling with no history of Tuberculosis (TB) contact. The histopathological examination (HPE) and all TB investigations were not suggestive of TB infection. He was follow-up until the wound healed well. In Case 7, the complaint of bilateral parotid swelling preceded by the right side and then the left side, which were persistent and painful, but there was no fever. He was treated as TB parotitis and completed TB therapy for six months at another center prior to the referral to our center. When he was seen at our centre, the parotid

swelling worsen and he underwent I&D, and on subsequent follow-up, the wound improved and subsequently healed with no further recurrence.

In this series, the longest duration of illness took about 3 years from the onset of the first presentations. She experienced recurrent bilateral parotid swelling due to sialolithiasis, which often resolved with antibiotics and sialagogue. She also had multiple histories of left parotid infection, which required I&D and was complicated by a fistula. She experienced persistent discharge of the left parotid region for about one week. She had a superimposed infection, and on examination, a fistula was seen on the left parotid. She was referred to the dental team for oral hygiene and treated conservatively. The fistula gradually resolved over a few months.

Investigations

Blood investigations, TB workups, and tissue HPE were routinely sent from our centre. All of the patients had a raised white cell count (WCC). Two patients underwent autoimmune investigations to rule out other concomitant pathologies or infections by paediatricians that reviewed the patients prior to referral to ORL, as detailed in Table 2. They reported the results as non-reactive. Acid-fast bacilli (AFB) were not detected in three out of four cases of chronic granulomatous disease, while another case was treated for TB parotitis at another centre. All cases of chronic granulomatous disease had a raised the erythrocyte sedimentation rate (ESR).

**Table 2:** Investigations performed on patients.

Case	Investigations						
	WCC	Erythrocytes sedimentation rate	Pus culture and sensitivity	AFB	Mycobacterium tuberculosis rapid polymerase chain reaction	Autoimmune screening	Histopathology examination
1	↑	NA	No growth	NA	NA	NA	NA
2	↑	NA	No growth	NA	NA	NA	NA
3	↑	NA	Group C Streptococcus	NA	Not detected	NA	No malignancy
4	↑	↑	Staphylococcus aureus	Not detected	Not detected	NA	Chronic granulomatous disease
5	↑	↑	No growth	Not detected	Not detected	NA	Chronic granulomatous disease
6	↑	NA	No growth	NA	NA	NA	NA
7	↑	↑	No growth	NA	Not detected	Non-reactive for ENA, C3, C4, RF	Chronic granulomatous disease
8	↑	↑	No growth	Not detected	NA	Non-reactive for ANA	Chronic granulomatous disease

**Abbreviations:** WCC: White Cell Count, AFB: Acid Fast Bacilli, ENA: Extractable Nuclear Antibody, C3: Complement 3, C4: Complement 4, RF: Rheumatoid Factor, ANA: Antinuclear Antibody, NA: Not Available.

In our series, Case 3 and Case 4 had positive pus culture and sensitivity (C&S), which were Group C Streptococcus and Staphylococcus aureus, respectively. A 10-month-old female in Case 3 had 2 surgeries (I&D). The C&S revealed Group C Streptococcus and raised WCC. She had no evidence of a fungal infection or Mycobacterium tuberculosis. Histological report showed no evidence of malignancy in the left parotid. Case 4 had history of persistent painless right pre- and post-auricular

swelling for 8 months, underwent investigation for preauricular cold abscess. Tissue C&S of post- and pre-auricular swelling revealed positive Staphylococcus aureus and raised ESR. However, TB workup such as Ziehl-Neelsen and acid-fast bacilli were negative, and the Mycobacterium tuberculosis rapid polymerase chain reaction was not detected. The periodic acid-Schiff (PAS) stain was negative for fungal elements Table 3.

**Table 3:** Imagings performed on patients.

Case	Ultrasonography Neck (USG)	Computed Tomographic (CT) Neck	Magnetic Resonance Imaging (MRI) Neck
1	Performed	No	Performed
2	Performed	Performed	No
3	Performed	Performed	No
4	Performed	Performed	No
5	Performed	No	No
6	Performed	Performed	No
7	Performed	Performed	No
8	Performed	Performed	No

**Imagings**

In this series, ultrasonography (USG) of the neck was performed as the first line of investigation. Majority of the patients (75%) underwent computed tomographic (CT) neck as they had recurrent swelling or pain and to look for extension of the swelling or abscess collection. Only one patient had MRI in

another centre prior to the referral, as he had a recurrent episode of neck abscess that required multiple I&Ds and was complicated by a fistula. After being treated, Case 1 was when referred to our centre, the symptoms recurred. She had an episode of painful, recurring swelling on the right side of her face. Within a month of the first symptoms, she had 2 I&Ds because of a parotid abscess recurred. The MRI revealed multiple abscesses in the superficial

lobe of the right parotid gland, with the largest measuring 18 mm x 22 mm x 17 mm. She was symptom-free for a 5-month interval, then developed fever and left parotid swelling. The USG neck showed changes in parotitis without abscess collection. However, she responded well to intravenous antibiotic.

Our oldest patient, Case 8, a 6-year-old child was treated for juvenile parotitis with sialolithiasis. She underwent multiple incisions and drainages, eventually complicated by a fistula prior to the referral to our centre. She was closely follow-up with surveillance USG neck and parotid every three months interval for a year. USG neck was reported as bilateral sialolithiasis with bilateral gland sialectasis with dilated ducts on the left 2.3 mm and right 1.9 mm, respectively. She was symptom free for six months after the last follow up. The latest USG parotid reported the bilateral parotid was not enlarged, and there was no dilatation of the ducts.

We also reported a case of persistent painless right pre- and post-auricular swelling in a 4-year-old male in Case 4. He had no history of TB contact. TB investigations were all negative except for pus C&S. USG neck surveillance was performed. Subsequently, the neck swelling was noted to be worsening, as evidenced by multiple right cervical abscess formations, whereby post auricular 1.5 cm x 1.0 cm and another collection in the deeper layer measuring 0.5 cm x 0.7 cm. There was another preauricular collection in the right preauricular, measuring 0.9 cm by 0.7 cm. The parotid gland was normal. She was subjected to right post-auricular wound debridement, right preauricular I&D, and biopsy under general anaesthesia. Histological examination reported as a chronic granulomatous disease. Subsequent follow-up revealed a well-healed scar and no more swelling.

Case 5 and Case 6, who were 2-year-old, and 5-year-old females respectively had histories of recurrent infected parotid swelling and completed multiple courses of antibiotics. Serial USG necks were performed and reviewed during the ORL clinic appointment. The USG neck of Case 5 had multiple heterogenous hypoechoic locules within the left parotid gland, which were likely to represent recurrent or residual intraparotid abscesses or suppurative intraparotid adenopathy. The swelling persisted upon regular follow-up, and HPE was reported as a chronic granulomatous disease. However, the swelling gradually reduced in size and resolved within few months and subsequently resolved at one year of follow-up.

While the USG neck of a 5-year-old female had features of chronic juvenile parotitis, which was commonly seen in autoimmune diseases such as Sjogren's syndrome, however pediatrician assessments showed no evidence of autoimmune disease. The contrast-enhanced computed tomography (CECT) neck showed the right parotid gland was more enlarged and bulkier than the left one. The parotid duct showed no hyperdense focus, suggesting calculus. Both cases underwent multiple I&Ds

and were monitored with serial USG necks.

## Treatment

All of the patients received either oral or intravenous antibiotics as part of their medical treatments. Our case series showed that all JRP complicated with parotid abscess underwent I&D, and one of them also had experienced ultrasound guided aspiration. Four cases reported of chronic granulomatous disease resolved without complication however one of them had complicated with fistula and required follow-up which subsequently resolved with conservative management. Case 5 and Case 6 required serial USG necks as the lesion persists, however successfully discharged from ORL Clinic less than one year follow up after being treated conservatively by referring to dental team for oral hygiene emphasizing and keep hydrated.

## Outcome

In this series, 3 out of 8 cases developed parotid fistulas following I&D procedures. However, all of the fistulas gradually resolved in less than 4 months. There was no sign or symptoms to suggest recurrent after 1 year of follow up.

## Discussion

Juvenile recurrent parotitis (JRP) manifests between the age of 3 and 6 years old with an average of eight episodes of acute flare per year [8]. To date, Brodie et al. [9] stated that the recurrence 2 episodes of parotid sialadenitis per year is adequate to diagnose JRP and the frequency of illness will determine its severity. Garavello et al. [10] proposed few inclusions and exclusions criteria to diagnose JRP such as age <16 years, recurrent unilateral or bilateral swelling and at least two episodes during the last six months, otherwise, must exclude obstructive lesions, dental malocclusion, Sjogren syndrome or congenital IgA immunodeficiency. Aetiopathogenesis of JRP remains multifactorial encompassing hereditary, congenital ductal malformations, infections and autoimmune disorders. Reduced or obstructed salivary flow predisposes for parotitis. Inadequate hydration results in exacerbation of parotitis but can be conservatively managed with hydration and sialagogic agents such as sour foods. Xie et al. [3] found out in his study that children usually had attack either night or early in the morning due to low saliva flow as lack of stimulation during that period.

Poor oral hygiene predisposes for ascending parotid ductal infection and progression of this pathology due to non-intervention will result in suppurative parotitis and eventually parotid abscess [7]. However, JRP rarely suppurate and require drainage [2]. Only few reports of the existence of parotid abscess in children. Parotid abscess among paediatric group is rare and life-threatening event. Thus, abscess should be evacuated either by aspiration or incision and drainage [11]. However, in our cases, all cases progress to parotid abscess either in cases of partially treated with antibiotic or recurrence of infection on different occasions. Streptococcus

pneumonia and Haemophilus influenza are the commonest pathogens isolated in a series of 10 children with parotid abscess [7]. In our study, Case 3 had recurrent parotid abscesses, and the pus culture yielded Group C Streptococcal while Case 4 had tissue culture positive for Staphylococcus aureus. Drainage method was either spontaneous rupture, needle aspiration or incision followed by intravenous Metronidazole combined with Penicillin or Cefuroxime [7]. Clindamycin was proposed by Benaim et al. [12] study for complicated case with fever, purulent discharge or failure to resolve within 48 hours of conservative treatment. While Amoxicillin-Clavulanic Acid still the first line antibiotics for acute JRP flare-up management [12]. In our cases, all of cases underwent I&D whenever the symptoms persist or did not respond to medical treatment such as antibiotic. We performed I&D at the most fluctuant area as the imaging showed the abscess was superficial.

Persistent JRP will contribute to chronic salivary dysfunction and more complicated complication. Fistula as a sequelae to parotid abscess is not common especially in paediatrics but the presence of a congenital first branchial cleft fistula predisposes a child for recurrent parotid infection [7]. Secondary parotid fistula resolves spontaneously while congenital fistula anomaly required surgical excision to prevent recurrent parotid infection [7]. According to Ijaduola et al. [13], infection is the most common causative agent for parotid fistula. In this series, three cases complicated with parotid fistula resolved gradually by times. Parotid sialolithiasis is rare in paediatrics age group. Underlying Sjogren's disease may be the cause of JRP. Manifestation includes dry mouth, burning oral mucosa, halitosis and often associated with significant dental carries. JRP and Sjogren's disease share similarities in terms of clinical symptoms and presence of sialectasis. Saliva in Sjogren's disease may appears thick and sticky while in JRP may not be so.

Serologic study facilitates the diagnosis. In acute flare-up cases, amylase and C-reactive protein should be routinely monitor [12]. It includes elevated levels of anti-RO (SSA), anti-La (SSB), rheumatoid factor (RF), antinuclear antibodies (ANA), erythrocyte sedimentation rate (ESR) and hypergammaglobulinemia. Sjogren's disease may co-exist with other autoimmune disorders such as systemic lupus erythematosus (SLE) and rheumatoid arthritis. In our Case 7 and Case 8, had the longest duration of illness compared to other cases and few autoimmune screenings test were taken but negative as shown in table 2.

Juvenile recurrent parotitis achieves complete spontaneous resolution is approximately 90 % of the cases. Adequate hydration, analgesia, sialagogues, warm compression and parotid gland massage are among the conservative measures practiced together with systemic antibiotic. First line pain control such as Acetaminophen and Ibuprofen should be optimized and avoid opioid [12]. Anatomical factors such as ductal dilatation, strictures and stenosis predisposes for JRP [14]. Ultrasound is

a non-invasive diagnostic modality compared to sialography and safe on children. Inflamed gland can be described by hyperechoic, heterogenous, or diffusely hypoechoic parenchyma. Besides, it demonstrates hyperechoic nodule bigger than 2 mm with a posterior acoustic shadow or known as sialolith. Sialectasia or lymphocytic infiltration will be referred to areas of hypoechogenicity of 2-4 mm [15]. Parotid ultrasound is the first line diagnostic tool. Besides, we routinely use US neck and parotid in disease surveillances and monitor progression of diseases during follow up. Those anatomical factors are often dealt with ductal probing, dilatation, normal saline and steroid irrigation by means of sialendoscopy resulting in lower recurrence rate reported by Gardner et al. [5] and, Tomar et al. [14]. Up to 95% of patients in a study were symptom free after one sialendoscopic treatment at 6-36 months follow up [16].

Another study mentioned a 36% recurrence rate after sialendoscopy [17]. Acute flares of JRP which response poorly to the conservative measure will require sialendoscopy procedures [2]. Studies done by Schneider et al. [18] revealed good prognosis of the usage of salivary gland endoscope during acute phase as well as prophylactic treatment during the nonacute phase. However, the similar study found that the similar outcome between the usage of antibiotic and sialendoscopy [18]. By performing sialendoscopy not without complications even though it is infrequent, it can cause iatrogenic ductal injury thus lead to perforation, stenosis, or strictures [9]. However, JRP demands higher cost of sialendoscopy compared to those with sialolithiasis even though both need support from post anaesthesia care unit [19]. Magnetic Resonance sialography is another recent technologically advanced and less invasive option compared to conventional sialendoscopy. JRP is self-limiting and incidence rate is inversely proportionate with age. Approximately 80 - 90% of JRP cases resolve by puberty. Past treatment options such as duct ligation, tympanic nerve section, sclerosing substance injection, radiotherapy and parotidectomy have been abandoned as treatment options with less morbidity now exists.

### Conclusion

Juvenile recurrent parotitis is a rare but is a treatable disease. Systemic causes may need to be ruled out in recurrence cases if clinically indicated. Any abscess should be drained besides optimize first line antibiotic but may be complicated with fistula that eventually healed with conservative management. Ultrasound is a good tool to be used as first line of investigation and useful for surveillance to guide the management plan.

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### Conflict of Interest

The authors declare that they have no competing interests.

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