Introduction: The purpose of this study is to evaluate the utility of ultrasound in patients with Juvenile Recurrent Parotitis (JRP) on conservative treatment.

Methods: This prospective study included 22 patients who were referred for ultrasound with an initial clinical diagnosis of JRP. Ultrasound findings were scored as Type 1 – consistent with JRP, Type 2 – indeterminate for JRP and Type 3 – no features of JRP. The medical records of these patients were followed up for a period of 2 years from the time of first scan to record the final diagnosis, number of recurrent episodes and need for additional radiological investigations or sialoendoscopy.

Results: The final clinical diagnosis was JRP in 14 patients, intra-parotid lymphadenopathy in 6 patients and Sjogren’s syndrome in 2 patients. Ultrasound showed type 1 findings in 10/14 (71%) and type 2 findings in 4/14 (29%) patients with JRP. 12 patients with JRP remained on conservative therapy at the end of the follow up period of 2 years. 2 patients with JRP had insufficient control of symptoms and were referred to an outside institute for sialoendoscopy. 8 patients with JRP (57%) had unilateral involvement. Mean number of annual recurrent episodes was 2. During the recurrent episodes, ultrasound demonstrated similar findings as seen during initial presentation.

Discussion: In majority of patients with JRP, ultrasound can demonstrate characteristic imaging findings. While the ultrasound findings of JRP can overlap with Sjogren's syndrome, these two entities can be differentiated based on clinical presentation and presence of positive serological markers in the latter. Unilateral involvement can differentiate JRP from other entities such as intra-parotid lymphadenopathy and Sjogren's syndrome.

Conclusion: Ultrasound, when combined with clinical information can help establish a definitive diagnosis of JRP and can obviate the need for other radiological investigations in patients on conservative therapy.

Keywords
Parotitis, Pediatric, Ultrasound, Recurrent, Juvenile
redness of skin in the region of parotid gland and increased turbidity of saliva lasting for a few days at a time and can have several such episodes in a year. The etiology of JRP is not well understood, although genetic causes, immunologic disorders, congenital parotid duct malformation, dental abnormalities and autoimmune disease have been implicated [3-5].

Clinically it is important to exclude other causes for parotitis such as Mumps, Sialolithiasis and Sjogren’s syndrome. Radiologic investigations are also often directed towards excluding Sialolithiasis, parotid abscess or parotid mass and to identify parotid duct enlargement. Ultrasound has a distinct appearance in the form of multiple small hypoechoic foci within the parotid gland with or without enlargement of the gland and increased flow on color doppler imaging (CDI). These foci are thought to represent dilatation of peripheral salivary duct branches with surrounding lymphocytic inflammatory response.

The management of JRP is debated. Sialoendoscopy has been described to have both diagnostic and therapeutic role. However, in many patients, the treatment is conservative and consists of antibiotics and anti-inflammatory medications and symptomatic therapy such as cold compressions. Moreover, JRP may also show spontaneous resolution with advancing age in some patients [7]. Thus, it is important that patients be offered a trial of conservative therapy before performing invasive procedures like sialoendoscopy. In such patients, ultrasound can potentially be the only radiological investigation necessary for management.

The purpose of our study was to evaluate the performance of ultrasound in patients with JRP who are managed with conservative therapy.

Materials and Methods

Patient selection

We identified patients who were referred for ultrasound with a clinical suspicion of JRP, at the time of first presentation, from April 2011 to June 2014. We accessed the medical records of the 25 patients thus identified, for a follow up period of two years from the time of initial presentation.

Image analysis

The scans were initially done by a first year or second year resident and were subsequently reviewed by a faculty radiologist. All scans were done with a linear high frequency probe using the department’s standard settings. Color doppler imaging was used in all patients. The ultrasound findings were recorded into three types as follows:

Type 1- Consistent with JRP: These patients had typical ultrasound findings in the form of multiple hypoechoic foci in the parotid gland with or without enlargement of the gland and increased flow on CDI.

Type 2-Indeterminate for JRP: These patients had the following features in isolation or in combination: diffuse enlargement of the parotid gland, generalized heterogenous echogenicity, increased flow on CDI. No discrete focal lesion was seen in these patients other than benign appearing intra-parotid lymph nodes.

Type 3-No features of JRP: These patients had features consistent with an alternate diagnosis.

Clinical data analysis

We reviewed the medical records of these patients to determine the final clinical diagnosis. The number of recurrent episodes through the two year follow up period were noted. As pediatric sialoendoscopy was not performed at our institute at the time of this study, any patients with persistent symptoms who was thought to potentially benefit from sialoendoscopy was referred to another institute. Such patients were subsequently excluded from our study.

Results

Of the 26 patients who were initially included in our study, 4 were lost to follow up and were thus excluded from the study. 14 patients had a final clinical diagnosis of JRP based on clinical and ultrasound findings. In 6 patients the final diagnosis was intra-parotid lymphadenopathy. In these, percutaneous needle biopsy revealed mycobacterial infection in 2 patients and bacterial infection in 4 patients. In 2 patients the final diagnosis was Sjogren’s syndrome based on clinical features and positive serological markers. The study comprised of 14 males (75%) and 7 females (25%) with median age of 8 years (range 3-15).

All 14 patients with JRP were initially managed conservatively. 2 patients had persistent symptoms and were referred to another center for sialoendoscopy. The remaining 12 patients remained on conservative therapy at the end of the 2 year follow up.

10 patients with JRP (71%) showed type 1 ultrasound findings (Figure 1). 4 patients with JRP (29%) and 2 patients with Sjogren’s syndrome showed type 2 findings (Figure 2). All 6 patients with intra-parotid lymphadenopathy showed type 3 findings. The imaging findings and diagnoses are summarized in table 1.
characterized by similar clinical features and ultrasonographic findings as seen at the time of initial presentation.

Figure 2: Gray scale ultrasound of the right parotid gland in a 15-year-old female shows an enlarged gland with diffusely altered echotexture. This patient was eventually diagnosed to have Sjogren's syndrome. Few small intra parotid lymph nodes are seen (arrows).

8 patients with JRP (57%) had unilateral involvement (Figure 3). The remaining 6 patients with JRP (43%), 2 patients with Sjogren's and 6 patients with intra-parotid lymphadenopathy had bilateral involvement.

Table 1: Comparison of final clinical diagnosis and ultrasound findings.

| Final clinical diagnosis | Ultrasound findings |
|-------------------------|---------------------|
|                         | Type 1  | Type 2  | Type 3  |
| Juvenile recurrent parotitis | 10      | 4       | 0       |
| Intra-parotid lymphadenopathy - tubercular | 0       | 0       | 2       |
| Intra-parotid lymphadenopathy - bacterial | 0       | 0       | 4       |
| Sjogren's syndrome       | 0       | 2       | 0       |

Discussion

JRP is an often self-limiting condition and many patients benefit from non-invasive conservative therapy [4, 8]. In the past, conventional sialography has been used in the diagnosis of this condition [9]. More recent advances include MR sialography [10]. However, ultrasound is a simple, cheap and widely available tool that can be used to accurately diagnose JRP in the appropriate clinical setting. Besides, absence of ionizing radiation and non-requirement of contrast media makes it an ideal modality in the pediatric population that is affected by this condition [6, 11].

The typical symptoms and signs of JRP makes it relatively straightforward to diagnose clinically. However, it is important to exclude other cause of parotitis or parotid gland enlargement which can be seen in the pediatric population such as mumps, suppurative sialadenitis, sialolithiasis, etc. [1]. Ultrasound is usually used as a first line modality to exclude these mimickers. In our study, 10 patients (71%) with JRP had type 1 findings with typical ultrasound appearance of multiple small hypoechoic foci in the parotid gland with or without diffuse enlargement and increased vascularity of the gland. 4 patients with JRP (29%) and 2 patients with Sjogren's syndrome had nonspecific findings in the form of enlargement of the parotid gland and increased flow on color doppler. No focal lesions were seen in these patients. Ultrasound has been previously described as a useful tool in the evaluation of patients with Sjogren's syndrome [12]. Although the imaging findings of JRP can overlap with Sjogren's syndrome, the two entities can be quite readily differentiated based on clinical presentation and positive serological markers in the latter [8]. Also, JRP can be unilateral in its involvement while systemic conditions like Mumps and Sjogren's tend to be bilateral. Normal ultrasound appearance of one of the parotid glands points more towards JRP.

In 6 patients with initial clinical diagnosis of JRP, ultrasound showed type 3 findings and was able to both exclude JRP and help diagnose the correct underlying pathology.

Patients with JRP differ in the number of recurrences per year. In our study the mean recurrence was 2 episodes per year.

Our study has few limitations. Pediatric sialoendoscopy was not offered at our institute. Therefore, all patients were initially offered conservative therapy. The practice in other institutes may vary and in patients needing sialoendoscopy, imaging other than ultrasound may be considered necessary. It is also possible that we underestimated the number of recurrences as the patients may not have presented to the hospital for every episode of parotitis.

Conclusion

In conclusion, in the majority of patients with JRP, ultrasound can demonstrate very specific imaging findings. These findings, when combined with clinical information can definitively establish a diagnosis of JRP. In patients being offered conservative therapy for JRP, ultrasound can be the sole radiological investigation. Ultrasound can be used both for the initial diagnosis and for monitoring changes while on therapy.

Authors’ Contributions

All authors contributed to the conception or design of the work, the acquisition, analysis, or interpretation of the data.
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Conflict of Interest

None.

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