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Key Findings and Experience in the Management of Juvenile Recurrent Parotitis with Sialoendoscopies—A Retrospective Study

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Abstract: Introduction: Juvenile recurrent parotitis (JRP) is characterized by intermittent swelling of one or both parotid glands, accompanied by pain and associated with non-obstructive sialectasia. Sialoendoscopy is considered a significant treatment for JRP, being a safe and minimally invasive treatment method with good clinical outcomes. The purpose of this study was to assess the procedure of sialoendoscopy as a treatment modality for JRP and discuss the relevant literature on this topic. Methods: This study retrospectively reviewed clinical records of children diagnosed with JRP who were treated in the Maxillofacial Department of the Tzafon Medical Center, Poriya, Israel, with sialoendoscopy between May 2016 and March 2023. Data on demographics, the site of symptoms, follow-up duration, treatment outcome, and complications were collected for each patient. The inclusion criteria were patients not older than 16 years of age, with at least two episodes of swelling of the parotid gland unilaterally or bilaterally during a time period of 6 months. The exclusion criteria were patients older than 16 years of age, and patients who were diagnosed with sialolithiasis or Sjogren's syndrome. Results: The study included 17 children, of whom 12 (70%) were boys and 5 (30%) were girls. The mean age of the children at the time of the procedure was 6.7 years and ranged from 3 to 16. The affected parotid gland was the left in 7 children (41%), right in 9 children (53%), and bilateral in 1 child (6%). The average follow-up time was 45.17 months and ranged from 5 to 81 months. A total of 15 children (88%) were deemed to have a successful treatment result regarding the frequency of episodes after therapy and subjective improvement. Full resolution was seen in 10 children (59%). The sialoendoscopy findings were avascularity in the walls of the parotid duct, strictures, and mucous plugs. Pathological findings in sialoendoscopy were found in 11 out of 12 asymptomatic contralateral glands (p-value > 0.001). Conclusions: Sialoendoscopy under general anesthesia is a worthwhile and practicable treatment method for treating JRP. Satisfactory results were seen in the vast majority of patients. It is recommended to perform bilateral sialoendoscopy also in cases of a unilateral symptomatic gland. This treatment method is effective, safe, and potentially repeatable.

Keywords: sialoendoscopy; parotitis; JRP; juvenile recurrent parotitis

Citation: Mohana, A.; Bar, O.; Porat Ben Amy, D.; Abdelraziq, M.; Abu El-Naaj, I. Key Findings and Experience in the Management of Juvenile Recurrent Parotitis with Sialoendoscopies—A Retrospective Study. *Appl. Sci.* **2023**, *13*, 10780. https://doi.org/10.3390/app131910780

Academic Editors: Andrea Scribante, Federica Rocchetti, Ahmed Mohsen and Gianluca Tenore

Received: 13 July 2023
Revised: 23 September 2023
Accepted: 26 September 2023
Published: 28 September 2023

1. Introduction

Juvenile recurrent parotitis (JRP) is the second most common salivary gland disease among children, the first being mumps, and in areas where mumps vaccine is available, JRP is the most common parotid disease amongst children [1]. Still, no decisive etiology exists to precisely explain the occurrence of this phenomenon [2]. Some suggested etiologies include recurrent infections, allergies, deficient immune systems, and genetic factors or hereditary mutations [3–5]. Various types of viruses are associated with JRP, the most common being mumps, followed by others such as adenovirus, Epstein–Barr virus, parainfluenza, and human herpes virus type 6 (HHV-6) [6]. Infection of the upper respiratory tract may trigger JRP via dehydration that leads to hyposalivation, resulting in mucosal inflammation that

increases vulnerability to secondary infection. Yet JRP is still considered an inflammatory response rather than a result of recurrent bacterial infection [7]. The common time of onset is from 3 to 6 years of age, typically self-limiting by puberty, and more common amongst males [8–10]. Symptoms of JRP include intermittent swelling of one or both parotid glands accompanied by pain, local erythema, masticatory difficulty, and local or systemic fever. An episode of JRP will most likely last a few days, will seldom persist for weeks, and intervals between episodes may be as long as a few years [9]. A preliminary diagnosis is usually made based on clinical findings and elimination of other diseases such as dental or viral infections, or Sjogren's disease [2]. Diagnostic criteria for JRP were suggested in a meta-analysis by Garavello et al. [11]. According to this meta-analysis, the inclusion criteria for JRP are recurrent unilateral or bilateral swelling under the age of 16 years, with at least two episodes over 6 months. Exclusion criteria are obstructive lesions in the parotid gland, dental malocclusion, Sjogren's syndrome, and congenital IgA immunodeficiency. In general, the diagnosis of JRP is made mostly clinically, based on physical examination and personal history. Laboratory investigation is also included, as it was shown that the activity of serum amylase may be used in assessment of the salivary gland function, or even as a diagnostic marker [12]. Diagnosis may be confirmed with imaging methods, including sialography, sialoendoscopy, ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI). US is in common use and may reveal hypoechoic inclusions in the affected gland, as well as possibly enlarged cervical lymph nodes [13,14].

Currently, there is no absolute consensus on a precise treatment guideline for JRP [15]. Common treatments are antibiotics and analgesics, sialogogues, hydration, and gland massage, which are in common use for the acute phase [9,16]. For the chronic phase, diagnostic sialography was shown to have a therapeutic effect, which was credited to the irrigation and the potential anti-bacterial activity of the iodine-based contrast material [17,18]. This was the origin of the idea of implicating sialoendoscopy for therapeutic purposes; like sialography, sialoendoscopy enables the dilation and irrigation of the ductal system and the application of anti-inflammatory solutions. The hypothetic idea of the therapeutic effect of sialoendoscopy is irrigation, which may cut the inflammatory cycle by cleaning the duct of debris and mucous plugs [19]. In JRP, the inflammation of the gland reduces salivary flow, which aggravates inflammation, causing ductal metaplasia, further decreased salivary flow, and progressive parotid atrophy, thus creating a vicious cycle [20]. Factors that might increase the probability of this inflammatory cycle are reduced salivary flow due to dehydration, inflammation caused by infection, and ductal abnormalities of a congenital or autoimmune origin [20]. Although many reports and publications show success in treating JRP with sialoendoscopy [8,21,22], the superiority of this treatment modality in comparison with other techniques is not statistically proven [22].

The aim of this article is to present the experience of the Maxillofacial Department of the Tzafon Medical Center, Poriya, Israel in the treatment of JRP with sialoendoscopy, to assess the procedure of sialoendoscopy as a treatment modality for JRP, and to discuss the relevant literature on this topic.

2. Materials and Methods

This retrospective study included 17 pediatric patients (aged 3–16 years; mean age 6.7 years), who were diagnosed with juvenile recurrent parotitis according to radiologic and clinical features and were treated with sialoendoscopy between May 2016 and March 2023 in the Maxillofacial Department of the Tzafon Medical Center, Poriya, Israel.

This study was approved by the Tzafon Medical Center institutional review board (approval 0027-23-POR) and was performed under the Declaration of Helsinki principles.

All the patients approached the Maxillofacial Department of the Tzafon Medical Center with a complaint of recurring events of swelling. All the patients underwent a clinical examination, and most underwent a radiological procedure (US) of the face and neck before a surgical procedure was performed. Three patients did not undergo US due to their young age (3 years old), and one additional patient did not undergo US since the

processing of this patient was conducted in early 2018 before the protocol was implemented; since then, US has been performed on all patients, if cooperative. No other imaging was performed on these four patients before sialoendoscopy.

JRP was diagnosed in cases of a repeating acute unilateral or bilateral episode of parotitis, in some children accompanied by a characteristic US imaging of the parotid gland, showing hypoechoic areas with or without swollen cervical lymph nodes.

In the acute phase, conservative treatment was given, consisting of hydration, sour foods, gland massage, analgesics (paracetamol), and antibiotic treatment (amoxicillin–clavulanate). This was also recommended as a conservative treatment for future acute episodes.

After recovery from the acute phase, the patients underwent a thorough evaluation of their medical history and a physical examination, helping to confirm the diagnosis of JRP and to assess the severity of their condition. The parents of children who suffered from two swelling events or more in one year were advised to choose one from two treatment options, the first to wait until the child reaches the age of 16 years, when JRP usually spontaneously resolves, and to manage acute phases in a conservative fashion until then. The second option is to perform a sialoendoscopy. If swelling occurs again, another sialoendoscopy treatment is suggested.

The parents of 17 children decided to advance with that treatment, due to the potential benefits and good clinical treatment results.

The inclusion criteria were patients not older than the age of 16 years, who were diagnosed with JRP based on at least two episodes of swelling of the parotid gland unilaterally or bilaterally. All the patients had undergone at least one sialoendoscopy for one of the parotid glands. Most of the children (15 out of 17) were treated under general anesthesia. Treatment under local anesthesia was performed on two children, aged 13 and 16 years.

The exclusion criteria were patients older than 16 years of age, and patients who were diagnosed with sialolithiasis or Sjogren's syndrome.

Assessments of medical charts were conducted, as data was collected from the patient's records during pretreatment and follow-up examinations. For each patient, the obtained data included demographics (sex, age at the time of surgical intervention), medical history (background of chronic diseases), US findings (if available), number of sialoendoscopy procedures, sialoendoscopy findings, complications, and clinical outcome.

The criteria for successful treatment were defined either as total resolution, no swelling episodes in the follow-up period, or a distinct improvement in the frequency or intensity of swelling episodes, subjectively noted by children and their parents, leading to a better life quality during the follow-up period.

2.1. Technique of the Sialoendocopy Procedure

All the procedures were performed on an outpatient basis. The procedures were performed during asymptomatic periods and never during an acute phase of JRP. After detection of the orifice of the parotid duct, a local infiltration anesthetic was then injected with lidocaine 2% into the papilla area. Then, the duct orifice was gradually dilated with the use of lacrimal probes of increasing diameter (Figure 1).

These probes were used until a diameter of 1.2 mm was reached, which matched the outer diameter of the sialoendoscope later used. Afterward, a 1.2 mm Erlangen sialoendoscope (Karl Storz) was inserted intraductally and advanced inwards up to the end point of the ductal system (Figure 2).

The advancement was done with continuous lavage, beginning with 10 mL of lidocaine 1% and later up to 50 mL of isotonic saline solution. If strictures of the duct were seen, dilatation of these was performed with hydrostatic pressure. After establishing dilation of the ductal system, a rinse was performed intraductally with 10 mL of dexamethasone. After the operation, prophylactic antibody therapy was given with Augmentin (amoxicillin–clavulanate), according to the patient's weight, for 1 week.

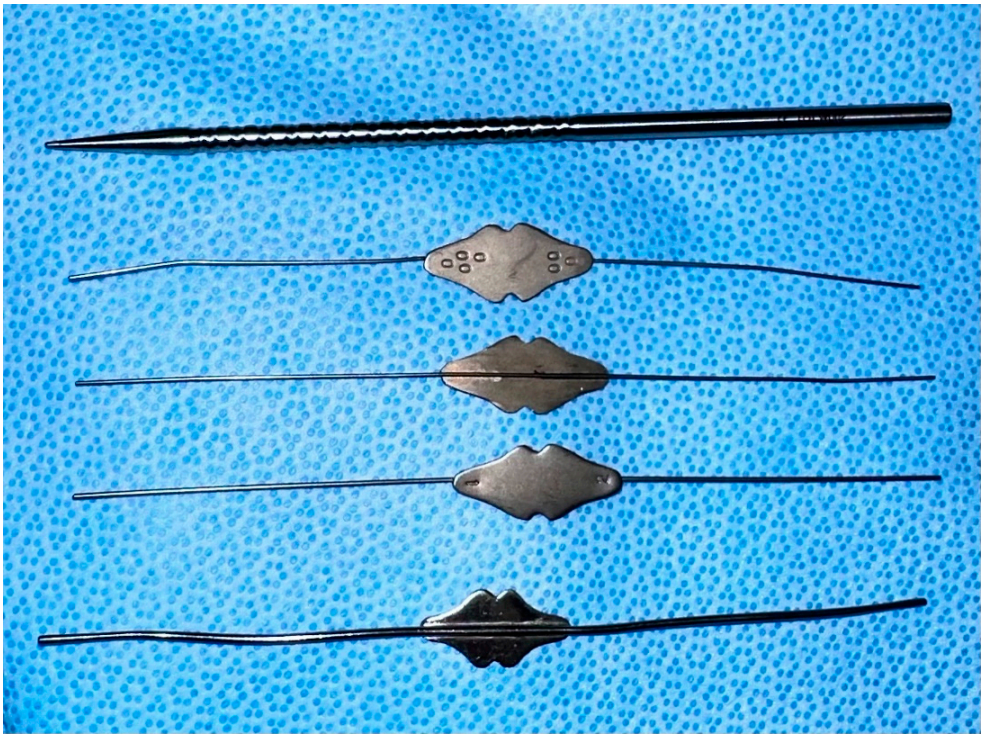

Figure 1. Lacrimal probes of increasing diameter.

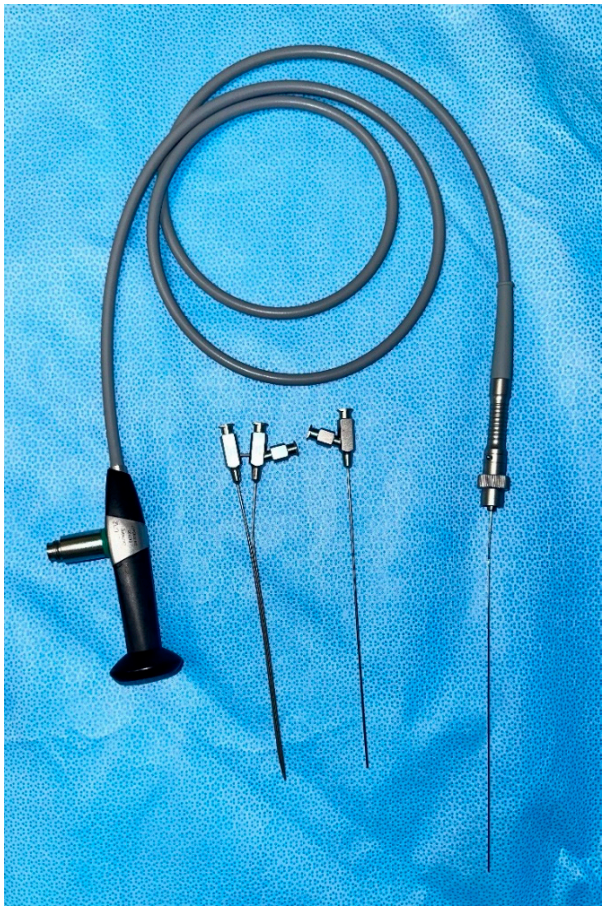

Figure 2. A 1.2 mm Erlangen sialoendoscope (Karl Storz).

2.2. Statistical Analyses

Descriptive statistics were employed to summarize demographic characteristics and relevant clinical factors. Categorical variables, including gender, the US findings, and the sialoendoscopy findings, were analyzed by Fisher's exact test (due to the small dataset). Continuous variables such as age were assessed using the Mann–Whitney U test due to the limited sample size. All the statistical analyses were conducted using SPSS v25.0. Significance was set at $p < 0.05$ (two-sided).

3. Results

Tables 1 and 2 show the patients' demographics, background, US findings, and treatment results.

Table 1 describes the characteristics of the patients, US findings, and treatment results.

Table 2 shows the results of the statistical analysis.

The study included 17 children, of whom 12 (70%) were boys and 5 (30%) were girls.

The mean age of the children at the time of the procedure was 6.7 years and ranged from 3 to 16. All the children were generally healthy; 1 had asthma.

The affected parotid gland was the left in seven children (41%), right in nine children (53%), and bilateral in one child (6%).

A pre-operative US was performed in 21 glands, 14 of which were symptomatic and 7 asymptomatic. Typical US findings were of a heterogenic appearance and strictures or dilation of the ducts.

Most of the children were treated with sialoendoscopy only once. Only two children were treated twice, due to a continuation of symptoms, and one child was treated with 4 endoscopies.

The majority, 15 of the 17 children, were treated under general anesthesia. Treatment under local anesthesia was performed on two children, aged 13 and 16 years.

The average follow-up time was 45.17 months and ranged from 5 to 81 months.

Complications were not seen in any of the patients. Immediate short-term post-operative swelling of the parotid gland was expected and considered an innocent surgical sequel that resolved spontaneously and was not counted as a complication.

All the patients were discharged on the same day as the procedure.

A full resolution was defined as a complete lack of symptoms after the sialoendoscopy and was seen in 10 children (59%). One of these patients had a single event of swelling, 70 months after the endoscopy procedure. Due to the long period of time that had passed, this patient's treatment result was defined as a full resolution. Full resolution with episodes of pain but without any swelling was seen in an additional two patients. Full resolution with a single event of swelling was seen in two additional patients, one of whom suffered from a single event of swelling 11 months after the procedure, and the second patient suffered from a single swelling event 1 month after the procedure, which was accounted as a natural post-operative course, and another single event of swelling 4 months after the procedures. One patient is still suffering from recurrent swellings, yet at a smaller frequency, and the parents report a subjective improvement and are not interested in additional treatment. All the above-mentioned patients, a total of 15 children (88%), are considered to have a successful treatment result.

Table 1. Patients' characteristics and treatment results.

Case Number	Sex	Age at Treatment (Years)	Medical Background	Gland Involved	Gland Treated	Episodes before Treatment	US Findings (Left)	US Findings (Right)	Number of Treatments	Anesthesia	Length of Follow-Up (Mo)	Complications	Episodes after Last Endoscopy (Time after Endoscopy in Mo)	Treatment Result **
1	M	6	Healthy	Right	Right	Recurrent	N/A	Normal	2	General	81	No	1 (70)	0
2	M	9	Healthy	Right	Right	Recurrent	N/A	Normal	2	General	73	No	0	0
3	F	5	Healthy	Bilateral	Bilateral	Recurrent	Heterogenic	Heterogenic	1	General	69	No	0	1
4	M	13	Healthy	Right	Right	Recurrent	N/A	Normal	1	Local	69	No	0	0
5	F	3	Asthma	Left	Bilateral	Recurrent	Normal	Normal	1	General	68	No	0	0
6	M	16	Healthy	Left	Left	Recurrent	Normal	N/A	1	Local	67	No	0	0
7	F	10	Healthy	Left	Bilateral	Recurrent	N/A	N/A	1	General	58	No	0	1
8	M	3	Healthy	Left	Bilateral	Recurrent	N/A	N/A	1	General	56	No	1 (55)	0
9	M	5	Healthy	Right	Bilateral	Recurrent	Strictures	Strictures	4	General	56	No	2 (1.4)	2
10	M	3	Healthy	Left	Bilateral	Recurrent	Heterogenic	Heterogenic	1	General	55	No	Multiple (6)	4
11	F	5	Healthy	Left	Bilateral	Recurrent	Heterogenic	Heterogenic	1	General	38	No	0	0
12	F	6	Healthy	Left	Bilateral	Recurrent	Heterogenic	Normal	1	General	26	No	0	0
13	M	7	Healthy	Right	Bilateral	Recurrent	Heterogenic	Heterogenic	1	General	14	No	Multiple *	3
14	M	3	Healthy	Left	Bilateral	Recurrent	N/A	N/A	1	General	14	No	1 (11)	2
15	F	11	Healthy	Left	Bilateral	Recurrent	Heterogenic	Normal	1	General	12	No	0	0
16	M	9	Healthy	Right	Bilateral	N/A	N/A	Dilation	1	General	7	No	0	0
17	M	3	Healthy	Right	Bilateral	N/A	N/A	N/A	1	General	5	No	Multiple	4

M: Male; F: Female; Mo: Months; N/A: Not applicable. * Parental report of recurrent swelling every month that is treated independently with self-massaging. The parents say that the general condition has improved, and they are not interested in any additional therapy. ** Treatment result: 0—Full resolution; 1—Full resolution with episodes of pain, no swelling; 2—Full resolution with a single event of swelling, months after endoscopy; 3—Partial resolution (few events of swelling, patients report subjective improvement); 4—Symptomatic (additional treatment needed).

Table 2. Summary of patients' (no. = 17) characteristics, treatment results, and sialoendoscopy findings.

Parameter	Results	*p* Value
Gender, no. (%)		
Male	12 (70.5)	0.03
Female	5 (29.5)	
Age, years		
mean	6.7	--
range	3–16	
Side, no. (%)		
Right	9 (53%)	0.2
Left	7 (41%)	
Bilateral	1 (6%)	
No. of treatments, no. (%)		
1	14 (82.3)	--
2	2 (11.8)	
3	1 (5.9)	
Results, no.(%)		
Full resolution	10 (59)	--
Pain without swelling	2 (12)	
Pain with swelling	2 (12)	
Treatment failure	2 (12)	
Findings, no.(%)		
Decreased vascularization	5 (16)	0.05
A-vascularization	19 (63)	
Strictures	14 (46)	
Mucus plugs	19 (63)	
Pathological finding in A-symptomatic gland	11 of 12	

The remaining two children (12%) are still symptomatic and have been prescribed additional therapy.

Sialoendoscopy was performed on 30 parotid glands in 17 patients. Common findings were A-vascularization, mucus plugs, and strictures.

Twelve children with a unilateral affected gland underwent bilateral sialoendoscopy, in eleven of whom, pathological findings were found on the asymptomatic side. Only one child out of twelve had no pathological findings in the sialoendoscopy performed in the contralateral asymptomatic gland. A pathological finding in an a-symptomatic gland was statistically significant. This is further described in Table 3, which shows the different findings.

Table 3. Description of sialoendoscopy findings.

Case Number	Gland Involved	Gland Treated	Endoscopy Findings (Left)	Endoscopy Findings (Right)
1	Right	Right	N/A	Few mucous plugs
2	Right	Right	N/A	Vast mucous plugs Avascularity
3	Bilateral	Bilateral	Constrictions Vast mucous plugs Decreased vascularity	Constrictions Vast mucous plugs Decreased vascularity
4	Right	Right	N/A	Constrictions Few mucous plugs
5	Left	Bilateral	Few mucous plugs Avascularity	Constrictions Few mucous plugs

Table 3. *Cont.*

Case Number	Gland Involved	Gland Treated	Endoscopy Findings (Left)	Endoscopy Findings (Right)
6	Left	Left	Decreased vascularity	N/A
7	Left	Bilateral	Constrictions Avascularity	Constrictions Avascularity
8	Left	Bilateral	Few mucous plugs	Few mucous plugs Avascularity
9	Right	Bilateral	Constrictions Avascularity	Few mucous plugs Avascularity
10	Left	Bilateral	Constrictions Few mucous plugs Avascularity	Constrictions Few mucous plugs Decreased vascularity
11	Left	Bilateral	Few mucous plugs Avascularity	Avascularity
12	Left	Bilateral	Constrictions Few mucous plugs Decreased vascularity	Few mucous plugs
13	Right	Bilateral	Constrictions Vast mucous plugs Avascularity	Constrictions Few mucous plugs Avascularity
14	Left	Bilateral	Avascularity	Vast mucous plugs
				Avascularity
15	Left	Bilateral	Avascularity	Constrictions Avascularity
16	Right	Bilateral	Constrictions Avascularity	Vast mucous plugs Avascularity
17	Right	Bilateral	No pathological findings	Avascularity

N/A: Not applicable.

Five glands (16% of glands, 4 patients) showed ducts with decreased vascularization, while nineteen glands (63% of glands, 12 patients) showed avascular ducts (Figure 3).

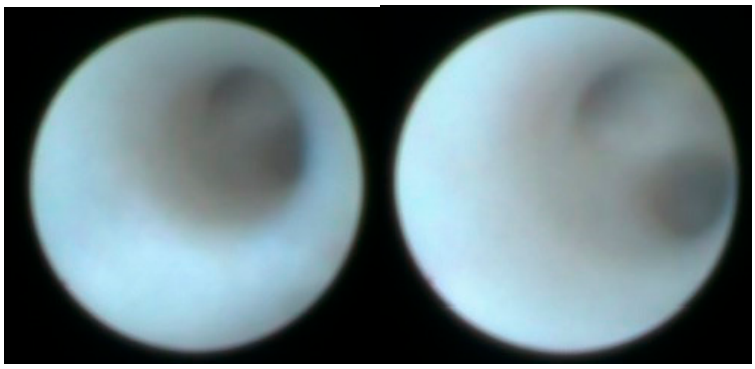

Figure 3. Sialoendoscopic appearance of the parotid duct: ducts showing avascularity.

Avascularity is a sign of chronic inflammation in the ducts.
Strictures were seen in 14 glands (46% of glands, 10 patients), (Figure 4).

Figure 4. Sialoendoscopic appearance of the parotid duct: a duct showing stricture.

A stricture in the duct negatively affects the salivary flow and may cause additional inflammation and the formation of mucous plugs.

Mucous plugs (few or vast) were seen in 19 glands (64% of glands, 13 patients), (Figure 5).

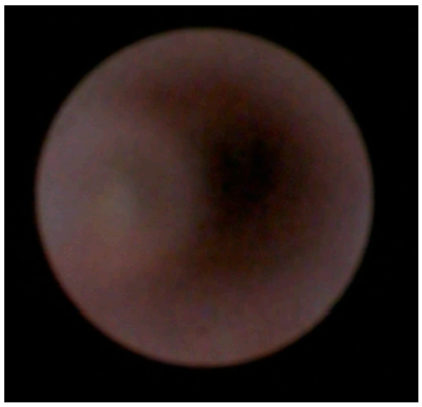

Figure 5. Sialoendoscopic appearance of the parotid duct: a duct showing a mucous plug.

A mucous plug in the duct is evidential to improper salivary flow and may cause additional duct inflammation.

4. Discussion

JRP mostly presents in the swelling of one, or sometimes, both parotid glands, enduring from several days to a week, with an asymptomatic period between episodes. Episodes tend to stop in early adulthood, for unknown reasons [1,23]. As mentioned above, the first episode commonly occurs between the ages of 3–6 years old and is more common in males [1,8,24–27]. Equal distribution between genders was also reported [28]. The data in this research shows the mean age of the children was 6.7 years, with a male predominance (70%).

Ultrasonography (US) is a common, cost-effective, noninvasive method, broadly used in the pediatric population for many indications. This modality can detect intra- and extra-glandular lesions, lymph nodes, and features that suggest the diagnosis of JRP, such as multiple areas of 2–4 mm hypoechoic lesions that represent ductal ectasia, lymphocytic infiltration, and sialectasis [13,14]. US is suggested as the first-line imaging method when suspecting JRP [29–31] and is very efficient in the acute phase. Between episodes, US findings will usually appear normal. In this study, pre-operative US was performed in 21 glands, 14 of which were symptomatic and 7 asymptomatic. The US findings were a heterogenic appearance and strictures of dilation in the ducts, yet no specific US characteristics were observed.

Various treatment options have been suggested for JRP over the years. These options can be classified as invasive or noninvasive and are utilized for either the acute or chronic phases.

The management of the acute phase is mostly aimed at relieving the symptoms of pain, swelling, and fever. Conservative measures for the acute phase are stimulating salivary flow by hydration or with the use of sialagogues, massaging the gland, and relieving the pain with anti-inflammatory medications and analgesics [22]. The prescription of antibiotics during the acute phase is considered controversial and is not proven to affect the course of the disease. Yet if signs of infection arise, antibiotic treatment should be given [32]. Sialoendoscopy is contra-indicated during the acute phase and might be more harmful than helpful [22].

Management of the chronic phase, which follows the acute phase, is aimed at reducing the frequency and severity of swellings, if they tend to reoccur. In the past, patients who did not benefit at all from conservative treatment may have proceeded to surgery, which included ablative measures like tympanic neurectomy, parotid duct ligation, and for severe refractory cases, gland excision or radiation. Parotidectomy, once considered the gold standard treatment for severe JRP, alongside the aforementioned invasive treatments, had numerous risks and potential side effects including facial nerve injury, a bad esthetic outcome, numbness of the earlobe, and Frey's syndrome. These techniques are not routinely in use today for treating JRP, except in strictly selected severe cases [1,18,33]. An additional procedure used previously that was considered less invasive was an injection of sclerosing materials into the ductal system [33,34]. In the last couple of decades, many studies have reported the increased efficacy of different minimally invasive treatments, namely sialography and sialoendoscopy, which have been shown to be effective, safe, clinically helpful, and with lower rates of complications [22].

In sialography, the anatomical structure of the parotid duct is depicted, and anatomical malformations can be detected by injecting a sialographic contrast dye through the ductal system. This produces a therapeutic effect that is attributed to the flushing of the ductal debris, the dilation of the duct, or the antibacterial effect of the iodine-based contrast material [22,35–37]. A study on sialography with iodine in hundreds of JRP patients found a symptom-free period of 1 year after the procedures, without any complications [18]. In a series of 110 patients treated with sialography, an improvement in symptoms was noted in 89% of patients, with a 67% mean decrease in the number of episodes of acute parotitis in the first year after treatment [38]. Sialography is considered to be safe, yet cases of extravasation of contrast material into the gland or buccal space have been reported [39]. This research reports success rates of 88%, which is comparable to those of sialography.

Another common treatment modality is that of sialoendoscopy, in which a specialized endoscope is used to assess the salivary glands and ducts minimally invasively, which was first described in 1993 [40]. It was also described for the treatment of sialolithiasis [41]. The advantage of sialoendoscopy over sialography has not yet been decided, with both modalities having a therapeutic benefit due to duct dilation and debris irrigation [22]. The surgeon's and the institution's preferences should play a role in choosing a treatment modality between the two, as reports from the literature do not clearly point to a preferred modality.

Sialoendoscopy is simultaneously diagnostic and therapeutic and enables irrigation and dilation of the parotid duct with a direct view of any ductal pathologies [24]. Typical findings during the sialoendoscopy that are consistent with a diagnosis of JRP are mucus plugs, debris and ductal stenosis, scarce vascularity in the ductal lamina, dilations, strictures, and a whitish ductal mucosa which is evidence of inflammation [8,21,26,27,42–45]. In this study, common endoscopic findings were decreased vascularization (5 glands, 4 patients), no vascularization (19 glands, 12 patients), strictures (14 glands, 10 patients), and few or vast mucous plugs (19 glands, 13 patients).

Numerous studies have shown improvement in or resolution of JRP symptoms following sialoendoscopy, although some of these have had a rather short follow-up period, which might not reveal the long-term efficacy of sialoendoscopy [8,26,28,45].

A systematic review of JRP stated that the rate of recurrence of swelling after sialoendoscopy was as high as nearly 26%, but with lower severity [11]. In this study, similar results were seen, as five children (29%) had at least one recurrent swelling in the follow-up period.

The reports in the literature demonstrate significant variability in percentages of symptom-free patients after sialoendoscopy. Some studies suggest that even one sialoendoscopy might be useful in healing the patient [8,28,31,46,47]. Others only saw an improvement and not a total resolution [48]. A retrospective analysis showed a complete resolution of symptoms in only 35% of the patients after one sialoendoscopy procedure, while all the remaining patients (65%) had a partial remission that was characterized by a decrease in the number and intensity of episodes [28]. On the other hand, another study showed that 90% of participants were free of symptoms after only one endoscopy procedure [46]. A recent study [27] described 76% of patients as symptom-free, but only for the first year after the procedure. A meta-analysis that included seven studies showed a success rate with no further sialoendoscopy needed at 87% [26]. The findings of this study are quite similar, as 15 children (88%) in this research had successful treatment results; 12 of them (80%) were treated with one sialoendoscopy procedure and did not need a further one. Complete resolution was seen in 10 children (65%); 8 of them (80%) were treated only once.

It should be noted that the need for repeated sialoendoscopy treatments to completely resolve symptoms is commonly described in the literature [1,20,49–52], even up to five times. It is reported that the recurrence of swellings after treatment, if it does occur, tends to be less intense and less painful and the treatment can be performed again in cases of several recurrences of swellings [8,24,27]. In this study, two children (12%) needed a second intervention, and one child (6%) needed up to four interventions in order to reach a successful treatment result.

A meta-analysis [26] reported bilateral symptoms in 39% of cases. Another study [28] showed bilateral involvement in almost 50% of cases. Yet, bilateral symptoms were also reported to be as low as 14% [49]. In this study, only one child (6%) had bilateral symptomatic glands.

All the children in this study were treated within two months of diagnosis, the vast majority of them bilaterally, even though only one gland was symptomatic. Some institutions perform sialoendoscopy bilaterally regardless of whether both glands are involved [46]. Yet lately, it was noted that in cases of a unilaterally affected gland, it is not required to operate bilaterally [53]. In another study, all the children who underwent a repeated sialoendoscopy due to a recurrence were those who first presented and were treated for unilateral parotitis [49]. In this study, 12 children with unilaterally affected glands underwent bilateral sialoendoscopy. In 11 of these, pathological findings were found on the asymptomatic side (p-value < 0.001). Only one child out of twelve had no pathological findings in sialoendoscopy in the contralateral asymptomatic gland. Therefore, bilateral treatment is recommended also when only one gland is symptomatic, and such prophylactic treatment may prevent the currently asymptomatic gland from becoming symptomatic later.

Potential side effects that might occur as a result of sialoendoscopy are swelling of the gland, perforation which might lead to swelling of the parapharyngeal space, and obstruction of the upper airway or breaching of the duct [22,44,54,55]. Complications of this procedure are rather rare and are not commonly reported in the literature, especially in the context of JRP. Yet, possible complications may be that of perforation of the duct, formation of duct stricture, and post-operative swelling of the operated gland [54]. In this study, no intra- or post-operative complications were seen. Immediate short-term post-operative swelling of the parotid gland was expected and was considered an innocent surgical sequel that resolves spontaneously or with the help of self-massaging and was not considered a complication.

Different solutions for gland irrigation are reported in the literature. In this study, the glands were washed with saline, and later with dexamethasone. However, irrigation with saline only, saline with steroids, saline with antibiotics, or saline with both steroids and antibiotics have been reported [8,26,33,50,53]. As similar effectiveness has been seen irrespective of the solution composition, it is considered that the main effect of irrigation is breaking the cycle of mucous secretion and stasis through the evacuation of intra-ductal mucous plugs and debris [1,19].

Performing the sialoendoscopy under local rather than general anesthesia is age-dependent, and among children, it is mostly performed under general anesthesia [4,8,24,56,57]. A recent study [27], which performed sialoendoscopy on patients with JRP, performed it under local anesthesia only with patients older than 16 years, and two additional studies [43,50] suggested that sialoendoscopy under local anesthesia may be useful for patients older than 8 years of age. Sialoendoscopy under sedation was also reported [49]. In this study, two children, the eldest in the study population, aged 13 and 16, were treated under local anesthesia. Both children had a successful treatment result and did not suffer from any episodes after the endoscopy. Still, the majority of the children, 15 out of 17, were treated under general anesthesia, as is often required in the pediatric population [21,31,46,48,50,57]. General anesthesia enables the optimization of the endoscopy procedure, shortening it and increasing its efficacy. It also prevents unexpected movements of the child, which could cause traumatic complications of the procedure. Additionally, performing the sialoendoscopy under general anesthesia provides the opportunity to treat both parotid glands in one session.

The main limitation of sialoendoscopy is the sensitive technique requirement. The procedure is operator-dependent and requires specific training. Moreover, associated costs are substantial, and the equipment needed is sensitive and prone to damage, which might limit its extensive use [58]. The need for general anesthesia in younger children is also a limitation but can also be of value, as performing the procedure under general anesthesia enables the surgeon to work under more convenient conditions in terms of cooperation, and the sialoendoscopy can be done more thoroughly and bilaterally.

Many of the reports in the literature lack a proper follow-up time. The follow-up time described in the literature, both in single studies and in a meta-analysis, is reported to be as much as 36 months or less [8,26,31]. Some studies focus on the follow-up period of the first year after the procedure [48,50]. That is not enough, as it was shown in this study that recurrence of swelling may appear as late as 55 or even 70 months after treatment. In this study, the follow-up time ranged up to 81 months, with a mean follow-up time of 45.17 months. Nevertheless, a recent study claimed that the majority of recurrences occur in the first year after the endoscopy, and, afterward, recurrence rates decreased [27].

The weaknesses in the present research are similar to the majority of other studies in this area and are those of a rather small study population, a retrospective research design, and the absence of a control group for comparison. Since JRP is inclined to spontaneously heal, a control group could be particularly important. Without such a control group, distinguishing the therapeutic effect of the treatment from the ordinary course of the disease is more challenging. Considering the results of this study, it is indicated that bilateral sialoendoscopy under general anesthesia is an effective and safe treatment method and should be anchored as part of a treatment plan for JRP, also in cases of unilateral gland involvement. This study's results showed a clear clinical improvement and even complete resolution and recommend the combination of sialoendoscopy with an intraductal injection of corticosteroids for any child suffering from JRP, both as a diagnostic tool and as a treatment modality.

5. Conclusions

This study indicates that the use of sialoendoscopy under general anesthesia is a worthwhile and practicable treatment method for treating JRP. This shows satisfactory results, with an improvement in swelling episodes for the vast majority of patients. Sialoen-

doscopy can be effective both as a diagnostic tool and as a therapeutic one, and is safe and minimally invasive, and therefore is a modality of choice for treatment of JRP in the Maxillofacial Department of the Tzafon Medical Center. This treatment method is effective, safe, and potentially repeatable. It is followed by fast recovery and scarce peri-operative complications. Moreover, a good clinical outcome is the main advantage of this treatment. According to the findings of this study, it is highly recommended to perform bilateral sialoendoscopy also in unilateral cases, in one session, under general anesthesia. Performing the treatment under general anesthesia enables bilateral treatment and is helpful in the pediatric population, in which cooperation is mostly not optimal. Further trials, especially randomized prospective ones, could help in determining the differences in the efficacy of the treatment comparing different methods to control groups and reaching higher levels of evidence.

Author Contributions: Conceptualization, A.M., D.P.B.A., M.A. and I.A.E.-N.; Investigation, O.B.; Data curation, O.B.; Writing—original draft, O.B. and A.M.; Supervision, A.M. and I.A.E.-N.; Project administration, O.B. All authors have read and agreed to the published version of the manuscript.

Funding: This research received no external funding.

Institutional Review Board Statement: The study was conducted in accordance with the Decleration of Helsinki, and approved by the Tzafon Medical Center institutional review board (approval 0027-23-POR-14.6.2023).

Informed Consent Statement: Patient consent was waived due to the retrospective nature of the research, anonymization of the data.

Data Availability Statement: The data presented in this study are available on request from the corresponding author. The data are not publicly available due to privacy restrictions.

Conflicts of Interest: The authors declare no conflict of interest.

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