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Clinical efficacy of low-temperature radiofrequency ablation of pharyngolaryngeal cyst in 84 Chinese infants

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Abstract

The aim of the study is to investigate the advantages and disadvantages of low-temperature radiofrequency ablation of pharyngolaryngeal cyst.

The study population was composed of 84 children diagnosed with pharyngolaryngeal cyst who underwent surgical treatment at the Department of Otolaryngology, Wuhan Children's Hospital, Wuhan, China, between January 1984 and December 2013. All patients were operated using a self-retaining laryngoscope and were divided into 3 groups: traditional cystectomy group (N = 9), dynamic cutting system group (N = 18), and low-temperature radiofrequency ablation group (N = 57). Clinical outcomes were analyzed to assess the efficacy of low-temperature radiofrequency ablation in treatment of pharyngolaryngeal cyst.

Compared with traditional cystectomy group or dynamic cutting system group, operation time was shorter, bleeding was less and one-year recurrence rate was much lower in low-temperature radiofrequency ablation group. However, operation time and bleeding was not statistically different between traditional cystectomy and dynamic cutting system group.

Low-temperature radiofrequency ablation may be an effective substitute for treating pharyngolaryngeal cyst.

Abbreviations: CO_2 = carbon dioxide, CT = computerized tomography, MRI = magnetic resonance imaging, SICU = surgical intensive care unit.

Keywords: infant, laryngeal cyst, low-temperature radiofrequency ablation

1. Introduction

Congenital pharyngolaryngeal cyst is a rare laryngeal-occupying lesion.[1,2] Ever since this condition was first reported by Abercrombie,[3] an increasing number of studies have investigated the clinical characteristics, diagnosis, and treatment of pharyngolaryngeal cysts.[1–3] Pharyngolaryngeal cyst can arise in any region of laryngopharynx, such as glottis, appendices ventriculi laryngis, aryepiglottic folds or subglottic area.[1–3] Pharyngolaryngeal cysts can lead to respiratory tract obstruction in children, and if not diagnosed accurately and treated in a timely manner, could even result in death from suffocation.[1–3] Several therapeutic methods have been used in the treatment of pharyngolaryngeal cysts. These include open surgery (median laryngotomy approach), needle aspiration, conventional endoscopic cystectomy (using laryngeal forceps and powered microdebrider), and carbon dioxide (CO_2) laser resection.[1–3,4] Open operation is usually used when cysts that are large and extend beyond the larynx.[2] With the development of endoscope, endoscopic operation has become the main mode of therapy. Conventional endoscopic operation has the advantage of being less invasive, thus resulting in a quicker recovery.[3,4] However, there are diverging opinions regarding this issue. Some studies have suggested that pharyngolaryngeal cysts treated via endoscopy are more likely to leave residual tissue that can cause recurrence. Moreover, the operative visual field can be easily blurred by blood accumulation during conventional endoscopic operation.[5,6] Therefore, there is an urgent need to develop new treatment modalities that are less traumatic and also ensure a thorough resection.

In recent times, low-temperature radiofrequency ablation has been widely used in cardiology,[7] otorhinolaryngology,[8] and oncology.[9] Bäck et al reported a shorter operating time and lower intraoperative blood loss with low-temperature radiofrequency ablation as compared with traditional tonsillectomy.[10] Low-temperature radiofrequency ablation of the base of tongue is considered an acceptable therapeutic method for treating supine-position-associated obstructive sleep apnea.[11] The advantage of low-temperature radiofrequency ablation in the treatment of pharyngolaryngeal cyst include a clear operative field, less bleeding, shorter operation time, and lower recurrence rate. Liu et al[12–14] found that the operation time was of 5- to 10-minute duration, and the volume of blood loss during surgery was 1 to 5 mL in a case series of 14 patients with tongue base cyst treated by low-temperature radiofrequency ablation.[14]

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However, there is no systematic study with a large sample to compare the differences between low-temperature radiofrequency ablation and conventional endoscopic cystectomy of pharyngolaryngeal cyst. In this retrospective study, the treatment outcomes were analyzed to clarify the advantages and disadvantages of low-temperature radiofrequency ablation of pharyngolaryngeal cyst compared with traditional cystectomy and resection using dynamic cutting system.

2. Materials and methods

2.1. Patients

In this retrospective study, 84 children with pharyngolaryngeal cyst underwent surgery at the Department of Otolaryngology, Wuhan Children's Hospital, Wuhan, China, between January 1984 and December 2013. The study was established according to the ethical guidelines of the Helsinki Declaration and approved by the Ethics Committee of the Wuhan Children's Hospital. Written informed consent was obtained from all parents/guardians of the patients prior to surgery.

The inclusion criteria were presence of at least 1 of the following clinical manifestations: inspiratory stridor, dyspnea, snoring, feeding difficulty, choking during eating, weak cry, or neck mass; and laryngoscopy or imaging examination was used for diagnosing pharyngolaryngeal cyst (described later). Patients with pharyngolaryngeal cyst not suitable for laryngoscope operation were excluded from the study.

2.2. Diagnosis of pharyngolaryngeal cyst

Before operation, patients underwent the following investigations: palpation of neck, blood gas analysis, B-mode ultrasonography of thyroid, and laryngoscopy. Diagnostic imaging (computerized tomography [CT] or magnetic resonance imaging [MRI]) was used when needed. Blood gas analysis showed hypoxemia and/or CO_2 retention. B-mode ultrasonography excluded aberrant thyroid lesions. Neck CT showed round or oval low density lesions with clear demarcation by surrounding tissues and uniform density inside the pharyngolaryngeal cyst (Fig. 1A). Neck MRI showed round or oval changes of long T1

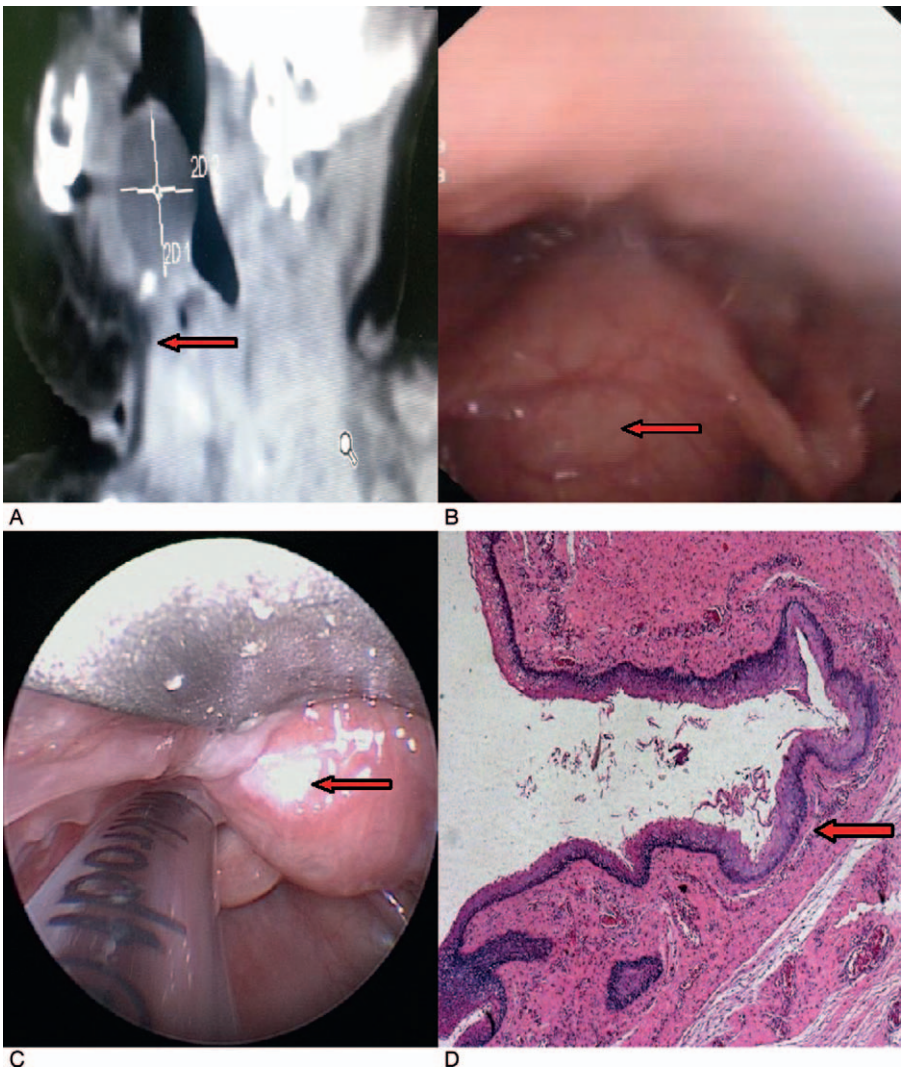

Figure 1. Diagnosis of pharyngolaryngeal cyst. (A) Computed tomography of the neck showing round or round-like low density lesions with clear demarcation from surrounding tissues and uniform density in pharyngolaryngeal cyst (red arrow). (B, C) Laryngoscopic findings of round or round-like lesion with clear demarcation, a thin layer of connective tissue capsule wall, and white lacteal or limpid secretions within the cyst. (D) Histopathological examination showing cystic dilatation with white lacteal or limpid secretions and squamous epithelium.

Table 1

Demographic and clinical characteristics of study subjects.

	Low-temperature radiofrequency ablation group (N=57)	Dynamic cutting system group (N=18)	Traditional cystectomy group (N=9)
Age, d	311±24	218±11	232±25
Sex (M/F)	32/25	9/9	5/4
Weight, kg	8.55±0.69	6.21±0.55	6.65±0.59
Course of disease, d	5.6±1.12	5.7±1.79	7.5±1.91
Cyst diameter, cm	1.76±0.33	1.8±0.32	1. 48±0.18

and long T2 signals. Laryngoscopy showed a rounded lesion with clear demarcation, a thin connective tissue capsule, and white lacteal or limpid secretions within the cyst (Fig. 1B and C). Histopathological examination showed cystic dilatation with white lacteal or limpid secretions and squamous epithelium (Fig. 1D).

2.3. Treatment of pharyngolaryngeal cyst

All 84 patients underwent endotracheal intubation and intravenous combined general anesthesia. Twenty-one cases with large cysts underwent paracentesis before endotracheal intubation. The operation was performed using a self-retaining laryngoscope in all 84 cases. Patients were divided into 3 groups: traditional cystectomy group (N=9), dynamic cutting system group (N=18), and low-temperature radiofrequency ablation group (N=57). Nine cases underwent paracentesis before endotracheal intubation followed by traditional cystectomy using goblet laryngeal forceps (STORZ, Germany). Eighteen cases underwent resection using powered microdebrider (Spiggle, Germany). In low-temperature radiofrequency ablation group, the pharyngolaryngeal cyst were excised as far as possible along its root, and the basal part of the cyst wall was ablated (the excision gear was set at 7, and the ablation gear was set at 3) using ENT Coblater II system (ArthroCare, Austin, Texas). In all patients, a tissue specimen was obtained for histopathological examination.

After operation, the patients received nasogastric feeding on days 1 to 3 and were administrated antibiotics and atomization. Children with congenital heart disease, bronchopneumonia, younger age, low birth weight, lower blood oxygen saturation before and during operation, repeated endotracheal intubation before operation, difficulty in extubation, or obvious laryngeal obstruction post-extubation were transferred to surgical intensive care unit (SICU) and referred back to the Department of Otolaryngology after stabilization. The average length of hospital stay was 6 days.

2.4. Statistical analyses

Statistical analyses were performed using the SPSS software (version 13.0; SPSS Inc, Chicago, IL). Continuous variables were expressed as mean±standard deviation. One-way ANOVA was performed to assess differences in continuous variables among groups. The chi-square test statistic was used for categorical variables. A P value $<.05$ was considered statistically significant.

3. Results

3.1. Baseline characteristics

There were no statistically significant differences among the 3 groups in terms of age, sex, weight, cyst size, course of disease, rate of dyspnea, and symptoms (Tables 1 and 2). The mean age of 3 groups are 311±24 days (N=57, low-temperature radiofrequency ablation group), 218±11 days (N=18, dynamic cutting system group), and 232±25 days (N=9, traditional cystectomy group).

Clinical manifestations included: inspiratory stridor (61/84), dyspnea (39/84), snoring (29/84), feeding difficulty (41/84), choking during eating (28/84), weak cry (6/84), and neck mass (4/84) (Table 2).

The location of pharyngolaryngeal cyst were as follows: 34 at tongue base, 25 at lingual surface of epiglottis, 15 at epiglottic vallecula, 2 at valleculae linguae, 6 at the aryepiglottic fold, and 2 at subglottic region. The pharyngolaryngeal cyst had varying diameter (range 1.0–2.0 cm) and size (range $0.5\times0.5\times0.3$ to $2.6\times2\times2.4\,cm^3$).

3.2. Surgical characteristics of low-temperature radiofrequency ablation

Pathological examination revealed a mucous cyst. There were no obvious postoperative complications. There were no statistically significant differences among the 3 groups in terms of

Table 2

Clinical symptoms of pharyngolaryngeal cyst in 3 groups.

	Low-temperature radiofrequency ablation group (N=57)		Dynamic cutting system group (N=18)		Traditional cystectomy group (N=9)	
	Before	After	Before	After	Before	After
Stridor	71.9% (41)	0	77.8% (14)	0	66.7% (6)	0
Dyspnea	38.6% (22)	0	72.2% (13)	0	44.4% (4)	0
Snoring	29.8% (17)	3 (3.57%)	50% (9)	1	33.3% (3)	1
Feeding difficulty	47.4% (27)	0	55.6% (10)	0	44.4% (4)	0
Choking during eating	31.6% (18)	0	38.9% (7)	0	33.3% (3)	0
Weak cry	5.3% (3)	0	11.1% (2)	0	11.1% (1)	0
Neck mass	5.3% (3)	0	5.56% (1)	0	0% (0)	0

Table 3

Surgical characteristics and follow-up of patients with pharyngolaryngeal cysts who were treated with low-temperature radiofrequency ablation or conventional operation.

	Low-temperature radiofrequency ablation group (N = 57)	Dynamic cutting system group (N = 18)	Traditional cystectomy group (N = 9)
Operation time, min	13 ± 2.01*	20.6 ± 3.57	40.8 ± 5.73
Intraoperative bleeding, mL	0.45 ± 0.3*	2.6 ± 0.74	5.2 ± 1.02
Postoperative hospital stay, d	6.7	6.9	7.5
Proportion of patients transferred to SICU, %	45.6	46.4	49.8
One-year recurrence rate, %	1.75	5.5	11.1

SICU = surgical intensive care unit.
* $P < .05$, compared with conventional endoscopic group or dynamic cutting system group.

postoperative hospital stay or the ratio of cases that had to be transferred to SICU (Table 3).

Compared with traditional cystectomy group or dynamic cutting system group, operation time was shorter and bleeding was less in the low-temperature radiofrequency ablation group ($P < .05$, Table 3). The average operation time was 13 minutes and the average blood loss was 0.45 mL. However, operation time and bleeding was not statistically different between traditional cystectomy and dynamic cutting system group. In addition, low-temperature radiofrequency ablation had a clearer intraoperative visual field, a smoother and more regular surgical incision, more complete ablation of basilar epithelium, and less injury to the surrounding tissues (Fig. 2). After operation, 2 patients in the low-temperature radiofrequency ablation group continued to complain of snoring, and while in the conventional endoscopic group, the corresponding figure was 3. The other symptoms, however, were relieved (Table 2).

3.3. Follow-up of low-temperature radiofrequency ablation

Postoperative follow-up duration was from 6 months to 7 years in 77 cases, of which 45 cases were followed-up for 6 months to 1 year, 24 cases for 1 to 2 years, 7 cases for 2 to 5 years, and 1 case for 7 years (11 ± 2.49 months in low-temperature radiofrequency ablation group and 20 ± 3.57 months in conventional endoscopic group). Seven patients withdrew from the follow-up. In 3 cases (1 case/group), the pharyngolaryngeal cyst recurred at about 1 year after surgery; the others had no complications during postoperative follow-up. Because cases with follow-up of >1 year are too few to be analyzed, we only analyzed 1-year recurrence rate. One-year recurrence rate of low-temperature radiofrequency ablation group (1.75%) was significantly lower than that of traditional cystectomy (11.1%, $P < .05$) and dynamic cutting system group (5.5%, $P < .05$). The 1-year recurrence rate of dynamic cutting system group was significantly lower than that of traditional cystectomy ($P < .05$). The recurrent case in low-temperature radiofrequency ablation group underwent a second low-temperature radiofrequency ablation operation. The recurrent case in the conventional endoscopic group is still being followed-up.

4. Discussion

In this retrospective study, we aimed to evaluate clinical efficacy of low-temperature radiofrequency ablation in the treatment of pharyngolaryngeal cyst, in a sample of 84 Chinese infants. Patients were operated using a self-retaining laryngoscope, and divided into 3 groups: traditional cystectomy group (N = 9), dynamic cutting system group (N = 18), and low-temperature radiofrequency ablation group (n = 57). Clinical characteristics of surgery and follow-up analysis showed that there were no statistically significant differences among the 3 groups in terms of

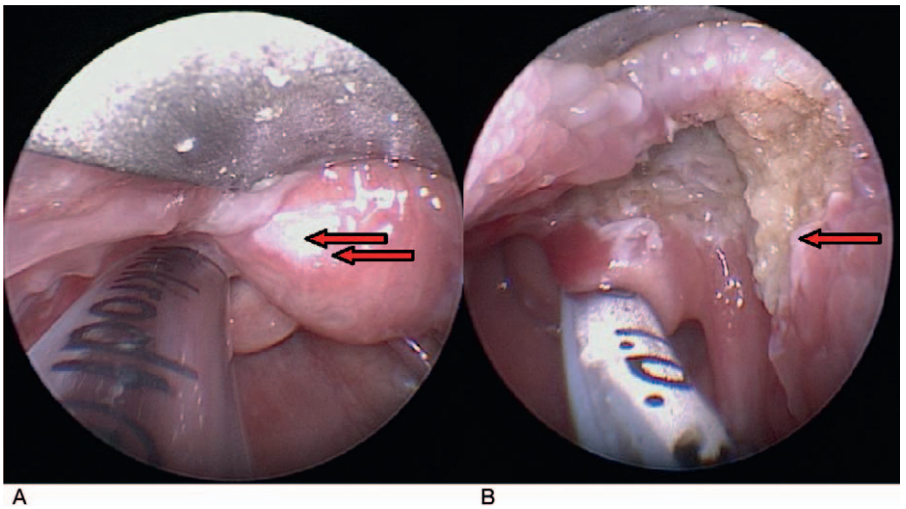

Figure 2. Surgical characteristics of low-temperature radiofrequency ablation. (A) Before surgery, red arrow showed the pharyngolaryngeal cyst; (B) after surgery, cyst has been completed ablated. Low-temperature radiofrequency ablation had a clearer operative visual field, ensured a smoother and more regular surgical incision, and effected complete ablation of basilar epithelium with much lesser degree of trauma to the surrounding tissues.

age, sex, weight, cyst size, course of disease, dyspnea, postoperative hospital stay, and ratio of patients transferred to SICU. However, the operation time was shorter and there was lesser bleeding and 1-year recurrence rate in the low-temperature radiofrequency ablation group as compared with traditional cystectomy group or dynamic cutting system group. Therefore, low-temperature radiofrequency ablation appears to be a more effective treatment for pharyngolaryngeal cyst as compared with the conventional therapy.

Pharyngolaryngeal cyst in infants is usually congenital and is caused by pathologic dilatation of sacculus of pharyngolarynx.[1–3] In the present study, 34 pharyngolaryngeal cysts were located at the tongue base, 25 on the lingual surface of epiglottis, 15 in the epiglottic vallecula, 2 at valleculae linguae, 6 at the aryepiglottic fold, and 2 cases in the subglottic region. The previous reports also detected laryngeal vallecular cyst[15] and Thornwald cyst.[16] This suggests that the pharyngolaryngeal cyst can occur in any region of pharyngolarynx. Symptoms of pharyngolaryngeal cyst are related to its size, location, and development speed, including, inspiratory stridor, inspiratory dyspnea, feeding difficulty, and snoring, etc.[1–3] In our study, 61 patients had inspiratory stridor, 39 patients had dyspnea, 29 patients had snoring, and 41 patients experienced feeding difficulties. Pharyngolaryngeal cyst should be considered in the differential diagnosis in patients with these clinical symptoms.

Pharyngolaryngeal cysts frequently induce airway obstruction and may even cause suffocation in infants.[15–17] Hence, it is very important to diagnose these patients accurately and in a timely manner. The previous reports as well as our own experience suggests that the diagnosis of pharyngolaryngeal cysts is usually made on palpation of neck, blood gas analysis, B-mode ultrasonography of thyroid, diagnostic imaging (CT or MRI), and laryngoscopy.[1–3] If round or round-like cyst with clear demarcation, a thin layer of connective tissue capsule wall, and white lacteal or limpid secretions within cyst are observed by laryngoscope, the diagnosis of pharyngolaryngeal cyst can be established.

While pharyngolaryngeal cysts may induce symptoms at birth, most of the patients become symptomatic by the age of 6 months.[1–4] Several therapies for pharyngolaryngeal cyst have been developed, such as open operation, needle aspiration, endoscopic extended cystectomy, and endoscopic CO_2 laser excision.[1–3,4] Needle aspiration of cyst is simple and one of the most effective measures for rescue, because it can quickly relieve the upper airway obstruction and dyspnea.[3,18,19] But the recurrence rate after needle aspiration is reported to be higher than with other therapies.[3,18,19] Conventional cystectomy using laryngeal forceps is difficult to perform, the bleeding is difficult to control, and is more traumatic for surrounding tissue.[3,5,20] CO_2 laser marsupialization was reported to have a good efficacy with almost no bleeding. However, the equipment is expensive and is difficult for wider use.[21,22] Because pharyngolaryngeal cyst is rare and therapies investigated in previous reports have widely varied, the treatment of choice for pharyngolaryngeal cysts is as yet undecided.

Recently, low-temperature radiofrequency ablation has been used to treat soft tissue lesion in otorhinolaryngology.[8,10,11] Carney et al reported a much lower resection time with low-temperature radiofrequency ablation of head and neck malignancies as compared with the CO_2 laser.[23] Pierson et al reported that low-temperature radiofrequency ablation allowed for a controlled debulking of the tumors with less blood loss and without the need for multiple instruments for the treatment of advanced juvenile nasopharyngeal angiofibroma.[24] However, these studies had a small sample size.

In our study, there were no statistically significant differences among the 3 groups in terms of age, sex, cyst size, postoperative hospital stay, and the proportion of patients requiring transfer to SICU, low-temperature radiofrequency ablation was associated with a shorter operation time, decreased amount of blood loss, and lower 1-year recurrence rate as compared with traditional cystectomy group or dynamic cutting system group. The conclusion was supported by two other studies on epiglottic cysts.[6,25] In one study, the intraoperative blood loss is less than 2 ml, and all patients recover without any complications after using coblation of epiglottic cyst. The other study claims that coblation excision includes direct contact with vallecular cyst, improved targeting of the cyst, and preservation of normal tissue. These results suggested that low-temperature radiofrequency ablation involves less damage to the surrounding tissues resulting in improved surgical outcome.

The underlying mechanism in low-temperature radiofrequency ablation involves generation of ion oscillations between the bipolar radiofrequency electric field and the formation of low temperature plasma layer (40–70°C).[7–9,26] Subsequently, the highly active radicals within the low temperature plasma cause chemical disintegration of the tissues into carbohydrates and oxide ions, resulting in tissue coagulation necrosis.[7–9,26] This mechanism of low-temperature radiofrequency ablation confers the advantage of a better visual operative field, more smooth and regular surgical incision, complete ablation of basilar epithelium, less trauma to surrounding tissues. All these advantages were observed in the present study.[7–9]

In addition, the surgical cost of each patient was recorded. The average surgical expense incurred by each patient in the low-temperature radiofrequency ablation group, dynamic cutting system group, and traditional cystectomy group was 29,900 RMB, 28,400 RMB, and 13,500 RMB, respectively. It should be noted that at our institution, low-temperature radiofrequency ablation and dynamic cutting system have been in common use over the last 10 years, but use of traditional cystectomy was discontinued about 10 years ago. Taking inflation into account, it might be concluded that the low-temperature radiofrequency ablation was not more expensive than dynamic cutting system and traditional cystectomy. A key limitation of our study was that it was a single-center retrospective study. Multicenter prospective randomized clinical trials would provide robust evidence for confirming the comparative efficacy of low-temperature radiofrequency ablation in relation to the other therapeutic modalities.

In conclusion, low-temperature radiofrequency ablation of pharyngolaryngeal cystitis is associated with shortened operation time, decreased blood loss, and lower 1-year recurrence rate. Our findings underline the efficacy of this technique in treating pharyngolaryngeal cysts. Its role as a valuable substitute to conventional therapy needs to be validated in prospective studies.

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