

Is a Multidisciplinary Aerodigestive Clinic More Effective at Treating Recalcitrant Aerodigestive Complaints Than a Single Specialist?

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Abstract

Objective: To determine the utility of a pediatric multidisciplinary aerodigestive clinic (ADC) in treating recalcitrant aerodigestive conditions.

Methods: Longitudinal observational study of presenting complaints, evaluation, management, and outcome of patients seen during 12 monthly ADCs beginning August 2013.

Results: Fifty-five patients were seen by the ADC team (otolaryngology/gastroenterology/pulmonology/speech pathology/nurse practitioner) and followed for a mean 17.6 months (range, 12–26 months). Mean age was 4.3 years (range, 0.5–19 years). All were seen by at least 1 specialist before ADC referral but without significant improvement. Chronic cough was the most common primary symptom (44%). Clinic evaluation included flexible nasopharyngolaryngoscopy (FFL, 53%) and pulmonary function testing (36%). FFL influenced management in 79%. An operative procedure usually combined endoscopy was warranted in 58%. Endoscopy provided high diagnostic yield, detecting laryngeal cleft (8), adenoid hypertrophy (8), vocal cord dysfunction (4), pulmonary infection (4), reflux disease (3), laryngomalacia (3), tracheomalacia (2), cilia abnormality (2), celiac disease (1), *Helicobacter pylori* (1), duodenal web (1), and eosinophilic esophagitis (1). Outcome was available for 48 of 55 patients, with 73% reporting resolved to markedly improved symptoms and 27% minimal to no improvement.

Conclusions: The ADC team approach resulted in resolved to markedly improved symptoms in 73% of patients whose symptoms persisted despite seeing a single specialist prior to referral.

Keywords

aerodigestive disorders, direct laryngoscopy, endoscopy, GERD, laryngology, otolaryngology, adenotonsillectomy, eosinophilic esophagitis, reactive airway disease

Introduction

Complex aerodigestive disorders in children pose diagnostic and treatment challenges. These patients often present with a variety of diagnoses, which may include chronic cough, gastroesophageal reflux disease, asthma, allergic rhinitis, recurrent pulmonary infections, sleep apnea, feeding disorders, congenital airway anomalies, and/or airway malacia.¹ Children with recalcitrant aerodigestive problems often have extensive evaluations and tend to bounce between clinics and providers as their symptoms often overlap specialty fields. Lack of a unified diagnostic evaluation and treatment plan can negatively impact patient satisfaction and quality of life and drive up health care costs.²

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The purpose of a pediatric multidisciplinary clinic is to coordinate care between multiple providers during a single office visit. The interdisciplinary approach may result in earlier diagnosis and treatment, leading to improved patient outcome and family quality of life. The benefits of an interdisciplinary approach have been demonstrated in the management of several pediatric conditions, including obesity,³ asthma,^{4,5} diabetes,⁶ and cystic fibrosis.^{7,8} Additionally, some pediatric interdisciplinary clinics have demonstrated a reduction of costs^{2,9-13} and in instances where operative management is necessary may reduce the number of separate episodes of anesthesia and thus decrease exposure to repeated anesthetics.¹⁴

The multidisciplinary aerodigestive clinic (ADC) team at Children's National Health System (CNHS, formerly CNMC) in Washington, DC, is comprised of a dedicated pediatric otolaryngologist, pediatric gastroenterologist, pediatric pulmonologist, advanced registered nurse practitioner, and 2 pediatric speech and language pathologists, the latter of whom alternate attending each clinic. All ADC team members see patients at a single combined appointment and develop an integrated and coordinated treatment plan after a roundtable discussion of the patients' condition. During the rare event that a dedicated ADC team member is unable to attend the ADC, a colleague from the same specialty covers the clinic.

This study aims to assess the success and utility of such a clinic by determining the most common presenting complaint, diagnostic workup, management, and outcome in patients who presented to the ADC with recalcitrant aerodigestive complaints despite prior specialist evaluation. We define *recalcitrant complaints* as symptoms that failed to improve despite evaluation by a single specialist (either a pulmonologist, gastroenterologist, or otolaryngologist) prior to referral to the multiple specialists of the ADC. Interestingly, despite the prevalence of ADCs nationwide, a literature search failed to show a similar study with relatively long-term follow-up.

Materials and Methods

This longitudinal observational study was approved by the Children's National Health System Institutional Review Board, Washington, DC (CNHS 00005677). All participants underwent their first CNHS ADC evaluation during the 12 monthly consecutive clinics conducted between August 2013 and October 2014. November and December 2013 clinics were cancelled due to hospital holidays. Patients were typically referred by their pediatrician or ADC members who had seen patients in their own specialty clinic and suggested referral for a multidisciplinary evaluation. All ADC team members remained the same throughout the study period. Parents completed a preclinical survey summarizing their child's symptoms, past medical history, family history, prior diagnostic tests, and treatment. Data

Table 1. Presenting Primary Symptoms (n = 55).

Presenting Primary Complaint	Frequency (%)
Chronic cough	25 (44)
Chronic nasal congestion	9 (17)
Stridor	7 (13)
Dysphagia	6 (11)
Recurrent infection	6 (11)
Aspiration	1 (2)
Choking episodes	1 (2)

were collected retrospectively from the parent-completed, preclinical screening surveys, initial clinic evaluations, operative reports, diagnostic test results, and follow-up clinic evaluations. Patients whose diagnostic workup disclosed multiple conditions would be seen for a follow-up appointment in the combined multidisciplinary clinic; however, follow-up appointments for most patients were conducted in a single specialty clinic. Clinic designation was based on the patient's condition; for example, a patient diagnosed with eosinophilic esophagitis would follow up with the gastroenterologist while a patient undergoing adenotonsillectomy would follow up with the otolaryngologist. Patients for whom follow-up clinic evaluations were not available, either because the patient missed an appointment or because an in-person clinic evaluation was not felt necessary, were contacted by telephone to assess their condition.

Results

Fifty-five patients were seen during the study period. All patients were seen by at least 1 specialist before ADC referral but failed to show significant improvement in their clinical picture, thus prompting the referral. The mean age at presentation was 4.6 years (range, 0.5-19 years), and 64% (n = 35) were male. Most patients had multiple symptoms on presentation, with chronic cough being the most common and the primary symptom in 44%. The primary presenting symptoms are listed in Table 1. At the time of presentation, 67% of patients (n = 37) reported that they had been previously diagnosed with gastroesophageal reflux disease (GERD), 75% of them by clinical evaluation and 25% by GERD testing, but ADC symptoms persisted despite medication trial. Eighty-two percent of patients (n = 45) presented with a diagnosis of reactive airway disease. Among this group, 16 patients were over the age of five, 9 of whom had the diagnosis of asthma confirmed by pulmonary function testing.

Diagnostic Workup

The diagnostic procedures completed in the 55 patients are listed in Table 2. Clinic evaluation always included a

Table 2. Diagnostic Evaluation by Aerodigestive Clinic (n = 55).

Diagnostic Evaluation	Frequency (%)
Flexible nasopharyngolaryngoscopy (FFL)	29 (53)
Pulmonary function testing (PFTs)	20 (36)
Operative procedure	33 (60)
Combined endoscopy ^a	12 (22)
Direct laryngoscopy and bronchoscopy (DLB) alone	6 (11)
Esophagogastroduodenoscopy (EGD) alone	5 (9)
DLB and flexible bronchoscopy with lavage	5 (9)
DLB and EGD	5 (9)
Flexible bronchoscopy with lavage alone	0 (0)
Polysomnography	10 (18)
Multichannel intraluminal impedance (MII)	9 (16)
Immune evaluation	8 (15)
Sweat chloride testing	5 (9)
Cilia biopsy	4 (7)
Functional endoscopic evaluation of swallow (FEES)	2 (4)

^aCombined endoscopy consisted of DLB, flexible bronchoscopy with lavage, and EGD.

thorough history and physical examination completed by each provider. Based on clinical presentation, flexible fiberoptic nasopharyngolaryngoscopy (FFL) was completed in 53% and pulmonary function testing in 36%. Of those who underwent FFL, the procedure influenced management in 23 of the 29 patients, with findings of adenoid hypertrophy (n = 17), laryngomalacia (n = 2), and vocal cord dysfunction (n = 4).

An operative procedure, usually single or combined endoscopy, was deemed indicated to evaluate the presenting symptoms in 60%, polysomnography in 18%, intraluminal impedance testing in 16%, immune evaluation in 15%, sweat chloride testing in 9%, cilia biopsy in 7%, and functional endoscopic evaluation of swallow (FEES) in 2%. The most common operative procedure was combined triple endoscopy consisting of direct laryngoscopy and bronchoscopy (DLB), flexible bronchoscopy with bronchoaveolar lavage (BAL), and esophagogastroduodenoscopy (EGD), performed in 22% of patients. Eleven percent underwent DLB alone while DLB and BAL, DLB and EGD, and EGD alone were each performed in 9% of patients (Table 2).

Of all patients who underwent bronchoscopy with BAL, 75% demonstrated abnormal findings. Forty-eight percent (n = 6) had lipid-laden macrophages present, 19% had positive viral panel results, 13% (n = 2) had evidence of tracheomalacia, and 6% (n = 1) had elevated neutrophil counts.

Multichannel intraluminal impedance (MII) testing was abnormal in 3 patients, including 2 who had normal EGD findings and normal histology on EGD biopsy. The MII was normal in 6 patients, despite 1 patient with diagnosis of GERD by EGD biopsy. Two of the 4 cilia biopsies showed

Table 3. Patient Diagnoses (n = 55).

Patient Diagnoses	Patients Seen, No. (%)
Based on diagnostic results	
Adenoid hypertrophy	8 (15)
Laryngeal cleft	8 (15)
Vocal cord dysfunction	4 (7)
Pulmonary infection	4 (7)
Reflux disease	3 (5)
Laryngomalacia	3 (5)
Tracheomalacia	2 (4)
Cilia abnormality	2 (4)
Celiac disease	1 (2)
Helicobacter pylori	1 (2)
Duodenal web	1 (2)
Eosinophilic esophagitis	1 (2)
Based on clinical assessment	
Asthma	8 (15)
GERD or EERD	5 (9)
Allergic rhinitis	4 (7)
Munchausen by proxy	1 (2)
Periodic fever and aphthous stomatitis	1 (2)

Abbreviations: EERD, extra-esophageal reflux disease; GERD, gastroesophageal reflux disease.

low numbers of cilia with oligocilia and microcilia. Sweat chloride testing was positive in 1 patient.

Establishing the Diagnosis

A diagnosis leading to improvement in symptoms was determined in 85% of patients based on clinical evaluation and results of testing, with some being new and some building on prior working diagnoses. Several patients also had more than 1 diagnosis (Table 3). Clinic and operative endoscopies provided the highest diagnostic yield, detecting laryngeal cleft (n = 8), marked adenoid hypertrophy (n = 8), vocal cord dysfunction (n = 4), pulmonary infection (n = 4), reflux disease (n = 3), laryngomalacia (n = 3), tracheomalacia (n = 2), cilia abnormality (n = 2), celiac disease (n = 1), Helicobacter pylori (n = 1), duodenal web (n = 1), and eosinophilic esophagitis (n = 1). Other diagnoses included Munchausen by proxy (n = 1), periodic fever and aphthous stomatitis syndrome (n = 1), asthma (n = 8), clinical GERD (n = 5), and allergic rhinitis (n = 4).

Treatment

The diagnoses of the patient population were broad. Treatment was tailored for each patient according to the diagnoses listed in Table 3. For example, patients with marked adenoid hypertrophy underwent adenoidectomy, patients with vocal cord dysfunction underwent breathing

Table 4. Patient Treatment or Interventions (n = 55).

Treatment/Intervention	No. Patients (%) ^a
Reflux medication	
Initiated	6 (11)
Advanced to proton pump inhibitor therapy	9 (16)
Reactive airway disease medication	
Increased dosage and/or administration	5 (9)
Antihistamine medication	
Initiated	2 (4)
Increased dosage and/or administration	4 (7)
Adenoidectomy ± tonsillectomy	17 (31)
Laryngeal cleft injection	7 (13)
Supraglottoplasty	1 (2)
Vocal cord training	4 (7)
Gluten-free diet	2 (4)
Antibiotic therapy	3 (5)

^aSome patients received more than 1 treatment modality; each % is independent value.

and voice training per our speech and language pathologists, patients with celiac disease underwent dietary modifications and referral to our celiac center, and patients with suspected allergic rhinitis were treated with antihistamines often in conjunction with nasal steroid sprays. The treatments described in Table 4 were deemed to be the most effective. However, it is important to note that some patients had multiple symptoms and multiple treatment modalities and that some patients did not improve.

Interestingly, at time of presentation to the ADC, 60% of patients had at least 1 previous trial of anti-reflux medication. Following evaluation in the ADC, reflux medications were advanced to proton pump inhibitor therapy (PPI) in only 16% of those aforementioned patients and started in another 11% of patients (Table 4). At initial presentation, 82% of patients were already prescribed medication for reactive airway disease, and these medications were increased in 9% of those patients by our pulmonologist. Forty-nine percent were taking antihistamines for nasal congestion prior to evaluation. Antihistamines were increased in 7% and started in 4%. Surgical intervention included adenoidectomy ± tonsillectomy in 31%, Type I A laryngeal cleft injection in 13%, and supraglottoplasty in 2% of patients.

Outcome

Treatment outcome was available for 48 of 55 patients. Mean follow-up was 17.6 months (range, 12-26 months). Medical and surgical management was tailored according to each patient's clinical and operative findings described in Table 3. Following the medical or surgical management noted in Table 4, 73% of responding patients reported

resolved or markedly improved symptoms, and 27% reported minimal or no improvement. Patients showing minimal improvement typically had developmental delay and multiple medical problems, including chronic conditions such as asthma, cilia abnormality, and/or vocal cord dysfunction.

Discussion

This study uniquely investigates the diagnostic evaluation, management, and relatively long-term outcome of a multidisciplinary pediatric ADC for recalcitrant aerodigestive complaints. Such a comprehensive review does not exist in the literature. This study demonstrated a high diagnostic yield, providing a new diagnosis in 85% of our patients. Of these, approximately 2 of 3 were established by results of endoscopy in the clinic or operating room, supporting the value of these diagnostic procedures. Furthermore, the majority of patients treated in the ADC had significant improvement in their symptoms over a relatively long follow-up period. These points demonstrate the true utility of a multidisciplinary approach to diagnosis and treatment of recalcitrant airway conditions, particularly because patients had already been seen by 1 ADC pediatric specialist prior to referral.

Diagnoses of this cohort of patients encompassed a broad range of conditions; however, the diagnoses were usually not rare conditions but rather common conditions that are seen in each of the individual specialty clinics of this group. A multidisciplinary approach is valuable because a potentially common etiology of a recalcitrant condition may not be easily recognized if the patient first presents to the wrong specialist. It is likely that patients who benefitted from adenotonsillectomy would have been scheduled for the procedure if they first presented to an otolaryngologist. Similarly, for patients with confirmed GERD, reflux disease might not have been considered by non-gastroenterologists. A combined eye is often more effective than a single eye in evaluating patients with aerodigestive complaints.

Chronic cough was the primary complaint in almost half of the patients seen in the ADC. Several clinical guidelines have been developed to evaluate pediatric chronic cough,¹⁵⁻¹⁸ and studies support a tailored approach similar to ours that systematically assesses the nature, duration of cough, and coexistent conditions.¹⁹

Although a large number of patients carried a diagnosis of GERD at presentation, only 10% were ultimately felt to have reflux disease. This assessment may slightly underestimate the role of reflux in causing aerodigestive conditions because our patient population would not have included patients who improved on empiric anti-reflux medications prescribed by their primary care provider because such patients would never have been referred to our ADC. Clinicians often use symptomatic improvement with an

empiric trial of anti-reflux medications as a diagnostic tool for reflux disease, leading to a marked overprescribing of anti-reflux medications.²⁰ The lack of a single true “gold standard” for diagnosis of GERD and EERD perpetuates this practice, and even our small series revealed histologically confirmed GERD in a patient with a normal MII-pH probe study. Our ADC gastroenterologist agrees with initiating an empiric trial of reflux medications for clinical histories and symptoms highly suggestive of reflux disease and reserves testing for those who fail to improve with therapy but for whom reflux disease is still suspected.^{21,22} Consistent with other literature, we have found that evaluation by endoscopic examination in conjunction with pH monitoring is complementary and may increase sensitivity in detecting reflux disease.^{23,24}

Vocal cord dysfunction was diagnosed in 7% of our patients. While this may seem like a relatively low yield, it deserves mentioning as it was a critical finding with a high rate of marked improvement in our patient population. Vocal cord dysfunction in pediatric patients is often misdiagnosed as asthma or exercise-induced reactive airway disease, resulting in improper treatment, sometimes for years, before the correct diagnosis is established.²⁵⁻²⁹ Vocal cord dysfunction should always be considered in patients with reactive airway disease, particularly exercise-induced, that does not respond to traditional bronchodilator therapy.

In our study, approximately 1 of 3 patients underwent adenoidectomy with or without tonsillectomy. Interestingly, of those patients who had the procedure, 76% also carried a diagnosis of reactive airway disease at presentation. While obstruction and recurrent infections of the upper respiratory tract are the major indications for removal of the adenoids and/or tonsils in children, several studies have shown concomitant improvement in asthma control and decreased medication and health care utilization.³⁰⁻³² In our study, of the 17 patients who had adenoidectomy ± tonsillectomy, 76% reported resolved or marked improvement in symptoms. It is difficult to pinpoint whether the improvement was a direct result of surgery itself versus surgery and medication changes in some patients, but it does highlight that adenotonsillectomy may be of benefit in certain patients with refractory reactive airway disease symptoms and chronic cough.

Through the coordination of care, patients were able to undergo operative procedures with multiple specialties at once, thereby limiting the number of anesthetic exposures to our pediatric population. This is a definite advantage as parents and health care providers have increasing concerns of the potential effects on neurocognitive development of multiple exposures to anesthesia in early life.³³⁻³⁷ It is important for multidisciplinary teams to only perform surgical procedures that are indicated and tailor surgical recommendations according to the patient's needs. Although not directly assessed in this study, coordination of care in

multidisciplinary clinics could theoretically decrease the health care cost burden to families. This is done not only by minimizing the number of office visits and avoiding repetitive testing by multiple providers but also by determining a definitive diagnosis and providing appropriate treatment.^{2,9-13} Reduction in health care costs of a multidisciplinary ADC have been shown theoretically,³⁸ and we recently have demonstrated that such clinics can operate in a cost-positive fashion when taking into account downstream revenue.¹³

Our study highlights the diagnostic and therapeutic benefits of a multidisciplinary clinic for patients with recalcitrant aerodigestive complaints. Despite having been seen first by a single ADC specialist, the majority of patients showed significant improvement after joint evaluation. Other studies reviewing ADC experiences are limited to assessing the outcome of a specific condition, such as laryngeal cleft management,³⁹ as opposed to an overview of all patients seen in the clinic. Critical diagnoses were able to be made in this clinic, redirecting such patients to the appropriate management. We are undertaking an intensive review of the conditions of patients who failed to show significant improvement to further improve our treatment outcomes and hope to report these findings in the future.

A limitation of our study is its retrospective nature and the resultant nonstandardized method of follow-up that was tailored to the patients' severity of disease and the patients' diagnoses. Patients with more severe symptoms were seen in follow-up appointments while patients with less severe symptoms were contacted by phone to determine the outcome of their therapy. For example, a patient with allergic rhinitis was not necessarily scheduled for a follow-up appointment to reassess the outcome of the antihistamine and nasal steroid therapy but instead instructed to contact us if symptoms did not improve with the prescribed therapy. Similarly, a patient with chronic cough who was found to have a bacterial cause on bronchoalveolar lavage would have been prescribed antibiotics over the telephone when culture results were available and then contacted by telephone 2 to 3 weeks later to determine the outcome of the antibiotic therapy. If the condition had resolved, a follow-up clinic appointment was not deemed necessary. We employ this tailored method of follow-up because we have found that parents prefer not to miss work/school for a simple discussion of treatment. A prospective study could arrange for standardized follow-up evaluations for all patients but would be very costly to fund.

Aerodigestive clinic providers as well as our patients have provided positive feedback on our multidisciplinary approach to patient management. Given that our study population was referred for specialized care at a dedicated tertiary care pediatric hospital, it may be difficult to implement this form of comprehensive care in the general population and smaller practices where resources may be more limited.

Conclusions

Further investigations are needed to confirm the impact of multidisciplinary care on aerodigestive disease and how multidisciplinary clinics can be improved, perhaps with the development of validated treatment algorithms. This study demonstrates that through coordinated care, physicians are able to establish a high diagnostic yield and significant improvement in the majority of patients with favorable implications on patient safety, morbidity, and quality of life.

Authors' Note

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Declaration of Conflicting Interests

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