Aerodigestive Programs Enhance Outcomes in Pediatric Patients

Christopher T. Wootten, MD, MMHC\textsuperscript{a,b,*}, Ryan Belcher, MD\textsuperscript{a,b}, Christian R. Francom, MD\textsuperscript{c}, Jeremy D. Prager, MD, MBA\textsuperscript{c}

INTRODUCTION

Beginning in the mid-1950s, indexed reports exist that use the term aerodigestive to describe the anatomic location of inhaled or ingested foreign bodies in children. Calcaterra and Maceri\textsuperscript{1} seem to be the first investigators to incorporate the term aerodigestive dysfunction into the title of a publication. This was in the context of

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\textsuperscript{a} Department of Otolaryngology, Vanderbilt University Medical Center, 2200 Children’s Way, Doctor’s Office Tower 7th Floor, Nashville, TN 37232, USA; \textsuperscript{b} Pediatric Otolaryngology, Vanderbilt University Medical Center, 2200 Children’s Way, Doctor’s Office Tower 7th Floor, Nashville, TN 37232, USA; \textsuperscript{c} Department of Otolaryngology, University of Colorado School of Medicine, Children’s Hospital Colorado, University of Colorado, 13123 East 16th Avenue B-411, Aurora, CO 80045, USA

* Corresponding author. Monroe Carell Jr. Children’s Hospital at Vanderbilt, Doctors’ Office Tower, 7th Floor 2200 Children’s Way, Nashville, TN 37232-9307.

\textit{E-mail address:} christopher.t.wootten@vumc.org


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compressive or invasive thyroid tumors affecting airway, breathing, feeding, and swallowing. Yet, by the early-1990s, considerable experience with airway reconstruction had led Cotton and colleagues\(^2\) to hypothesize about aerodigestive dysfunction or disease in children and, in particular, to articulate the importance of a thorough assessment of the entire airway before undertaking laryngotracheal reconstruction. Host or foreign body reaction to stents and the importance of systemic and inhaled steroids to mitigate airway granulomatous reaction were being discussed as a way to reconcile success and failure for the hundreds of children who had been operated on at Cincinnati Children’s Hospital over the preceding decades. In 1999, the same institution opened the first Pediatric Aerodigestive Center with the charge to colocate specialists from multiple disciplines to diagnose and manage the interrelated conditions known as aerodigestive disease. From the perspective of the otolaryngologist, the Center’s multidisciplinary purpose was to diagnose and control for disease states that may have detrimental effects on surgical success rates. Controlling gastroesophageal reflux disease (GERD) was the initial multidisciplinary task; however, as experience and communication increased so did the list of aerodigestive diseases to identify and treat.

In 2002, Hartnick and colleagues\(^3\) published a case report demonstrating the clinical value of this multidisciplinary approach. The investigators describe a young airway reconstruction patient whose preoperative workup included a normal pH study and esophageal eosinophilia on esophagastroduodenoscopy (EGD) with biopsy. She was placed on antireflux medications but struggled to maintain an adequate airway postoperatively until she was placed on steroid therapy and demonstrated resolution of eosinophilia and improvement in her airway. This was the first published association between what the investigators called allergic esophagitis and subglottic stenosis. Although the understanding of the entity now known as eosinophilic esophagitis continues to evolve, its association with large and small airway disease states and varied clinical presentation is the subject of continued publication, requiring awareness that crosses disciplines.\(^4\)

Through experiences such as these, and the integration of previously separate disciplines, aerodigestive programs continued to spread within individual health care organizations as well as between organizations. During the same time period, the method of delivering complex, coordinated care to particular populations began to undergo its own evolution. Pediatric multidisciplinary care as administered in the medical home model has been broadly and repeatedly endorsed by influential medical societies, including the American Academy of Pediatrics and the American Academy of Family Practice, since the mid-1960s. Likewise, the Centers for Medicare and Medicaid Services supports medical homes for children with complex health care needs in which there is a satisfaction of the triple aim: improved care, improved health, and lower costs. The concept of the medical home is sound for optimizing outcomes while lowering costs in caring for children with medical complexity.\(^5\) There are publications that specifically address the medical home concept in the management of pediatric airway problems (see later discussion). Highlights from these publications include lower overall cost of care, fewer days in-hospital per year, and a shift of care from inpatient environments to outpatient environments.

However, the current practice of pediatric multispecialty aerodigestive care has grown in sophistication beyond the medical home concept. In 2018, Boesch and colleagues\(^6\) published the first contemporary and multicentric assessment of the landscape of aerodigestive care as practiced across the United States. More a reflection of than a directive to aerodigestive centers, the investigators delineate the structure and functions of aerodigestive programs via the Delphi method to resolve
expert opinions. What is clear from multicentric, multidisciplinary expert input is that aerodigestive care is increasingly performed in an integrated practice unit (IPU) model.\(^7\) In the prevailing IPU model, the specific aerodigestive population, defined as the aerodigestive patient, is being diagnosed and managed by a multidisciplinary team colocated in time and space in dedicated facilities and supported by dedicated staff. The staff and providers convene regularly outside of the patient care arena to assess IPU operations, including a discussion of how health information and technologies are being leveraged to improve care and to measure the cost of that care. Ideally, throughout the aerodigestive IPU, there exists a focus on increasing care value.

Although 1 of several disorders qualify a child as an aerodigestive patient, these disorders are collectively rare. The unifying theme among the seemingly disparate aerodigestive conditions is not the primary organ system affected, a set of common embryologic errors, or a unified locoregional inflammatory response to some antigen. Rather, aerodigestive disorders are unified by a common contemporary approach to their management. The efflorescence of aerodigestive programs is not driven by an increased incidence of aerodigestive conditions. Instead, it is driven by a nationwide tendency toward care delivery models that provide integrated care.

Although the provision of integrated care through an increasing array of aerodigestive centers improves on the inefficiencies of piecemeal medicine, challenges remain. For example, as health care systems increasingly provide contemporary management of aerodigestive disease in children, it remains possible that more centers does not equal greater access. Furthermore, the authenticity of the IPU and its processes may be degraded by poor incentive alignment within and among providers, the organization, and payers, as well as insufficient community need due to IPU service area overlap. Fortunately, aerodigestive centers are communicating with each other. An international organization of pediatric aerodigestive care, the Aerodigestive Society (www.aerodigestive.us), is convening annually to promote valuable activities and to reduce unnecessary testing and intervention.\(^8\) Where there is a deficit of guidance, expert opinion has been garnered through Delphi studies. These opinions are forming the basis of multicenter research into the data objects, outcomes, and resource utilizations that define multidisciplinary aerodigestive care. Engagement with health care providers and patients is occurring at a quickening pace through the cooperation of the Aerodigestive Society, the various allied medical subspecialty groups, and the American Academy of Pediatrics. Authentic, appropriate, and accessible aerodigestive care is the mantra moving forward.

**DISCUSSION**

**Evolving Models of Aerodigestive Care Delivery**

Integrated care is organized around the patient. However, there are several different models of multidisciplinary care that tout patient-centricity. Indeed, colocated multispecialty care represents a convenience to families of children with aerodigestive conditions. Coordinating visits helps minimize travel costs, time off work, and lost revenue. Going a step further, the patient-centered medical home (PCMH) model cares for children with complex conditions using a multidisciplinary team that hinges on a primary pediatrician. Indeed, the concept of a PCMH was initially developed in order to meet the challenges of coordinating and delivering primary care. Those goals have then been applied to more complex medical needs and populations. In these settings, the pediatrician, embedded in the team, both administers and coordinates care for the patient to ensure patient-centricity, minimize waste, and maximize clinical
effectiveness. Medical homes lean on a primary care provider in a care coordination role to shepherd the patient to appropriate services integral to the patient’s treatment. These services include medical services; surgical services; social work; physical, occupational, and speech therapy; and community resources, to name a few. In the PCMH, the core primary care provider integrates and organizes the recommendations from the various medical home team members for the benefit of the patient in a way that is easy for the family to understand and to follow. Effective medical homes have open lines of communication among team members and through the primary care provider. Casey and colleagues describe their experience with a hospital-based multidisciplinary clinic for medically complex children. In the investigators’ experience, their medical home resulted in a significant decrease in Medicaid costs to provide care. Since that writing, the tertiary care-based medical home has been proposed as viable for pediatric aerodigestive care.

The IPU does not feature a core health care provider tasked with coordinating and integrating multidisciplinary care. Rather, the IPU cares for children through a dedicated, multidisciplinary team of clinicians who devote a significant portion of their time to the medical condition. In this model, providers see themselves as part of a common organizational unit. The team takes responsibility for the full cycle of care for the condition. Exactly how care is integrated is a key distinction between the PCMH and the IPU. The IPU also mandates effective communication. Indeed, education, engagement, and follow-up are managed by the IPU’s single scheduling and administrative structure. In the IPU, the value of the administrative structure, that is, the extent to which it has managed costs, achieved meaningful patient-reported outcomes, and leveraged the information platform, is the subject of deliberate self-scrutiny.

**Enhanced Outcomes**

An integrated care model offers many opportunities to improve the value of care. Using a rudimentary understanding of value (ie, value = [quality/cost]), aerodigestive programs have steadily added to the literature, demonstrating how they accomplish this goal. A sampling of this literature follows, many examples of which address both the numerator and denominator of this value equation.

**Reducing unnecessary testing**

Hart and colleagues reviewed patients who underwent pH multichannel intraluminal impedance (pH-MII) probe testing before airway reconstruction to determine if the results of pH-MII were associated with surgical outcomes. The investigators noted that fewer patients than anticipated (17.5%) had their management adjusted based on pH-MII results, particularly those patients with a prior history of fundoplication. For those patients without a history of fundoplication, pH-MII remained a valuable tool in decision-making for the investigators. DeBoer and colleagues evaluated the yield of gastrointestinal testing in pediatric patients in an aerodigestive clinic. For all comers over a 3-year period, 144 of 193 pH-MII were normal (74.6%). EGD with biopsy was negative for histologic abnormalities in 188 of 295 patients (63.7%). Upper gastrointestinal fluoroscopy (UGI) was normal in 47 of 54 patients (87%). Children with feeding difficulty, tracheoesophageal fistula or esophageal atresia, and asthma were most likely to have an abnormal EGD or pH-MII. Although many of the patients were on acid suppression medication at the time of testing, the low yield of pH-MII testing demonstrates a gap between the current state of clinical diagnosis and test result. Given the growing concerns regarding overuse and risk of acid suppression medication, the investigators modified their protocol for aerodigestive patients, removing...
pH-MII from routine use. In addition, the investigators comment that their investigation allowed for discontinuation of medication in those who did not need it. Data did support ongoing EGD and UGI use in this study.

Wentland and colleagues reviewed their patients’ experience undergoing videofluoroscopic swallow study (VFSS) after laryngeal cleft repair. The investigators’ multidisciplinary group reviewed the available literature and their own experience, leading to a modification of their previously published algorithm for swallow evaluation after cleft repair and a reduction in the number of VFSSs per patient. Though there was a small number of patients in this study, multidisciplinary discussion led to modification of an algorithm and a reduction in VFSS charges and radiation exposure.

Reducing charges and costs, risk, and time to diagnosis while maintaining viability
As aerodigestive programs have grown and spread, publications regarding their inherent efficiency, charge, and cost-reduction have increased in number. Collaco and colleagues described the potential opportunity for reduction in cost for families, as well as the potential reduction in anesthetic exposures and procedural charges due to enrollment in an aerodigestive program. In 2016, Skinner and colleagues demonstrated a shift from inpatient to outpatient care and a subsequent reduction in charges after enrollment in an aerodigestive program.

A 2017 retrospective examination of a single aerodigestive center revealed that after enrollment in the program there was a decrease in inpatient days and direct costs, extrapolated from an average for the state. The study had several limitations, including that an overall shift from inpatient to outpatient care has been occurring, as well as that the patients may have been clinically improving or may have been seen at other institutions.

A recent single institution study examined patient and family experience before and after the creation of an aerodigestive program. Patients with similar diseases achieved a reduction in time to diagnosis (6 vs 150 days) with fewer required specialist consultations (5 vs 11) compared with those seen in the same institution before creation of the aerodigestive program. These patients also underwent fewer radiology studies and anesthetic exposures. Charges for evaluations were also reduced from a median of $10,374 to $6055.

Although these studies focus on charges to the payer, cost to the institution to deliver care is also reduced. Costs for time-dependent resources (operating rooms, supplies, medications) were reduced approximately 40% when procedures were combined into 1 anesthetic event versus separated into 3 customary procedures (flexible bronchoscopy, rigid laryngoscopy and bronchoscopy, EGD). Charges were similarly reduced. These effects can be achieved while maintaining a positive revenue stream when the program is defined as the sum of the clinic encounter and the endoscopic procedures.

Developing new techniques
Medical innovation occurs by many different methods, among which is colocating multiple specialties and techniques. Colocation and sharing of ideas and techniques, when applied to aerodigestive diseases, can have significant impacts on care value. Unsedated transnasal esophagoscopy (TNE) with biopsy for pediatric eosinophilic esophagitis was discussed, then piloted, through the efforts of providers within the aerodigestive program. After demonstrating that mucosal biopsies were appropriate for diagnosis when obtained in this manner, patients and parents were surveyed about their experience of TNE. All parents and 76.2% of subjects would undergo the TNE again. TNE was preferred over EGD by 85.7% of parents and 52.4% of subjects.
Charges associated with TNE were 60.1% lower than for previous EGD. A follow-up study of 294 TNE demonstrated a consistent reduction in charges, no adverse events, and 71 minutes average check-in to check-out compared with 3 hours for EGD.\textsuperscript{22} Charge reduction occurred largely through the absence of anesthesia and facility fee. Risk reduction is obvious given the absence of anesthetic exposure.

Tracheopexy for tracheomalacia is yet another example of new techniques being applied through aerodigestive program care delivery methodology. First described as a treatment of tracheomalacia in 2015, the technique has been applied to those patients with congenital tracheomalacia most commonly due to tracheoesophageal fistula or esophageal atresia.\textsuperscript{23} The investigators describe a multidisciplinary team of pediatric surgeons, pediatric cardiothoracic surgeons, and pediatric pulmonologists primarily driven by pediatric surgeons. Publication and communication regarding this technique coalesced with an increasing commitment among aerodigestive programs to better treat the population of patients with severe tracheobronchomalacia. As a result, multiple groups have adapted this surgical method through the use of a thoracoscopic approach.\textsuperscript{24,25}

The number of aerodigestive IPUs has grown, and coordinated care is taking place across a broader geography.

Over the past decade, the number of identifiable aerodigestive teams providing care to children has increased. A recent publication noted 34 programs, 31 in the United States, most of which had been in existence for 5 years or less.\textsuperscript{26} A separate study by the first 2 authors of the present work, based on an Internet survey of aerodigestive programs in each state (search term aerodigestive followed by a state name) and their dates of establishment (Christopher Wootten, 2019, unpublished work), demonstrates the broad geographic distribution of these programs. Paralleling the high population density in the New England and mid-Atlantic regions, those same areas demonstrate the highest concentration of aerodigestive teams. The pattern of growth is demonstrated by comparing program density in 2007, 2010, 2013, and 2016 (Figs. 1–4).

The question arises, does an increasing number and geographic distribution of aerodigestive centers correspond to a commensurate increase in airway reconstructive operations? To attempt to answer this question, the Pediatric Health Information System (PHIS) was interrogated (www.childrenshospitals.org/phis) by the first 2 authors of the present study. The PHIS database is an administrative database that contains inpatient, emergency department, ambulatory surgery, and observation encounter–level data from more than 45 not-for-profit, tertiary care pediatric hospitals in the United States.

The PHIS database was queried for pediatric patients 18 years or younger who had undergone airway reconstruction surgery between 2007 and 2016 using coding that contains groups of \textit{International Statistical Classification of Diseases and Related Health Problems}, 10th revision, procedural coding system codes and current procedural terminology codes. The frequency of the airway reconstruction surgeries in each year were evaluated. The data were placed in a United States heat map format for the following years: 2007, 2010, 2013, and 2016 (see Figs. 1–4). The PHIS data are not provider-generated, so it is not possible to establish a 1-to-1 correlation between PHIS data points and specific types of airway reconstruction. However, over the decade spanning 2007 to 2016, the efflorescence and geographic dissemination of airway cases as detected by PHIS suggests that aerodigestive centers’ growth parallels growth in PHIS-reported case volume.

With a broadening geographic distribution of airway cases being performed over the past decade, one might assume improved access to care. The quality of that care is less known. For certain operations, the center’s experience and even the surgeon’s
individual experience with an intervention has been correlated with outcomes.²⁷–²⁹ It is imperative that surgeons and centers study their outcomes for complex airway reconstructions. Further, communication between aerodigestive programs should include a frank disclosure of best practices that seem to yield improved outcomes, as well as a disclosure of practices that are best avoided to soften the learning curve in airway surgery. Professional societies facilitate this communication and benchmarking.

**THE IMPORTANCE OF PROFESSIONAL SOCIETIES IN THE MAINTENANCE OF QUALITY AND THE CREATION OF FOCUS**

The elevation of evidence-based medicine over the past 4 decades has shown that, even among the best institutions and physicians, there has been significant unexplained variation in the quality and volume of care.³⁰ In the midst of this evolution of medicine toward focusing on patient-centered experiences, the clinical effectiveness of interventions, meaningful outcomes, and efficient health care delivery, IPUs have been developed and shown to provide patient-centered care.³¹

IPU and PCMH providers have made great strides in improved care for their patients, especially for patients with complex medical problems and comorbidities. Communication and association between disease-specific IPUs across the country has created collaborative improvement networks. These networks, often uniting as medical societies, can serve as catalysts to advance knowledge through meetings, communications, and research initiatives, which allows IPUs to share best practices,

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**Fig. 1.** Pediatric Health Information System (PHIS) heat map data indicating the number and geographic locations of unique patients with laryngotracheal surgical interventions in calendar year 2007 (*upper*) and the contemporaneous geographic distribution of aerodigestive centers (*stars*) that were verifiable by Internet query (*lower*).
advocate for recognition in academic and training programs, focus research efforts, raise money, and engage patient populations for improved patient-centered care. Increasingly, medical societies may become a source for medical research funding for their own members’ research initiatives because the National Institutes Health funding is at an historic low and continues to decrease.32

An often-cited example of treatment advances and improvement of morbidity and mortality due to medical society collaboration, benchmarking, and implementation of patient registries is found in the cystic fibrosis (CF) population. A national patient registry for CF patients in the United States was initially established in the mid-1960s. In the 1990s, epidemiologists started appreciating the registry’s usefulness to evaluate risk factors for disease progression. The CF Foundation has also long supported a benchmarking program that identifies CF centers that have excellent outcomes according to their registry data, then studies these CF centers’ exceptional organizational and structural features that contribute to these findings. This has ultimately led to less variation in the spread of effective treatment strategies for the CF population.33

In the realm of pediatric aerodigestive care, integrated care teams voted, in 2014, to organize as the Aerodigestive Society. However, the impetus for this organization was not patient-driven as in the case of the CF Foundation; instead, it was provider-driven. Unlike CF, which is a discrete, genetically linked multiorgan condition, aerodigestive conditions are many disorders with many etiologic factors. What links them are the common integrated care teams that have formed nationwide to help patients and populations realize value in the treatment of aerodigestive disease.

Fig. 2. PHIS heat map data indicating the number and geographic locations of unique patients with laryngotraceal surgical interventions in calendar year 2010 (upper) and the contemporaneous geographic distribution of aerodigestive centers (stars) that were verifiable by Internet query (lower).
The Aerodigestive Society has benefited from Delphi projects that defined the structure and function of aerodigestive teams and the airway outcomes they seek from laryngotracheal operations. The Society will continue to use Delphi methodology to assess and focus expert opinion on intake practices, diagnostic practices, interventions, clinical outcomes, and patient-reported outcomes. These agreed-on data objects will form the basis for an aerodigestive collaborative database that is multicentric and powered to answer nuanced questions about the inherently rare aerodigestive diseases and their treatments.

Certainly, the Aerodigestive Society is not the only professional organization that will help shape the quality and availability of care for children with aerodigestive conditions. National and international organizations within otolaryngology, pulmonology, gastroenterology, speech pathology, occupational therapy, esophageal atresia–tracheoesophageal fistula, and other fields have created sections and/or committees that educate their membership on the contemporary landscape of pediatric aerodigestive care. The journals affiliated with these allied professional organizations will continue to be the most appropriate venue for publishing original research into aerodigestive disorders.

Finally, the nature of aerodigestive research will not be restricted to clinical inquiry. Already, considerable work has been done on the economic logic underpinning integrated care delivery for aerodigestive disease. A 4-center value-based health care

**THE FUTURE OF AERODIGESTIVE CARE**

![Map showing PHIS heat map data indicating the number and geographic locations of unique patients with laryngotracheal surgical interventions in calendar year 2013 (upper) and the contemporaneous geographic distribution of aerodigestive centers (stars) that were verifiable by Internet query (lower).](image)
analysis in the management of oropharyngeal dysphagia is underway. What is clear from such health care business studies is the importance of leveraging emerging technologies such as nationally visible electronic medical records, telehealth, and automated patient-reported outcomes and biometrics that continually recenter the aerodigestive care delivery model on the patient.

SUMMARY
Aerodigestive programs fit into the modern medical trend of incorporating disparate disciplines into 1 location and team in order to provide more comprehensive, family-centered and patient-centered care in an efficient and cost-effective manner. Combining disciplines, by its nature, leads to improved communication and the development of improvements and innovation in care delivery. By deconstructing the siloed approach to care delivery, these programs and others may function as IPUs for disease states and populations, tracking outcomes that may include both clinical quality measurements and other metrics of care value (eg, cost of care delivery, time to diagnosis, reduction in unnecessary testing). Time will tell whether or not aerodigestive programs continue to develop and thrive. The external pressures that helped create the environment in which these programs have developed are subject to change from political and economic forces that may reinforce program development or serve to dissuade organizations, payers, and providers from coalescing into IPUs.

Fig. 4. PHIS heat map data indicating the number and geographic locations of unique patients with laryngotracheal surgical interventions in calendar year 2016 (upper) and the contemporaneous geographic distribution of aerodigestive centers (stars) that were verifiable by Internet query (lower).
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