The Journal of Laryngology & Otology (2009), ***123***, *642–647.*

doi:10.1017/S0022215109004599

Main Article

Management of dysphonia in children

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Abstract

Background: Dysphonia is common in children, but practice varies considerably regarding what, if any, investigations are performed and how the condition is managed. Although childhood dysphonia is mostly due to non-serious causes such as voice misuse, very serious pathology such as papillomatosis or malignancy needs occasionally to be excluded, and treatable congenital anomalies such as webs and cysts can be missed. Voice clinics and voice therapy services are now well established in most adult health services in the developed world, but equivalent services for children are less common, at least in the UK.

Methods: We retrospectively reviewed the records of all children presenting to our large children's hospital with a primary complaint of dysphonia between January 2001 and October 2007, in order to determine their management, investigations and final diagnosis.

Results: We identified 142 children. Case records were found for 137 (97 per cent). Eight-three children were male (61 per cent) and 54 female (39 per cent). Ages ranged from two months to 15 years (median 5.3 years). In 10 children (7 per cent), hoarseness was congenital, presenting as a hoarse, weak cry at birth. In 15 children (11 per cent), onset of hoarseness was related to a specific surgical procedure. The larynx was visualised by mirror alone in 23 children (17 per cent), by awake fibre-optic laryngoscopy in 27 (20 per cent) and by microlaryngoscopy-bronchoscopy under anaesthesia in 42 (31 per cent). Forty children (29 per cent) did not undergo laryngeal visualisation at any time and were diagnosed based on history alone. A further five (4 per cent) were scheduled for direct laryngoscopy but this was not performed due to resolution of symptoms. Voice abuse accounted for 62 (45 per cent) of all diagnoses.

Conclusions: Childhood dysphonia accounts for a large number of referrals. There is considerable variation in how these children are managed. A more structured approach to diagnosis and investigation would be beneficial, perhaps within the setting of a dedicated paediatric voice clinic.

Key words: Dysphonia; Children; Paediatrics; Hoarseness

Introduction

Impairment of voice quality is common in children. The term dysphonia is used to describe disorders of voice generation at laryngeal level, as opposed to disorders of vocal resonance (involving the nose and nasopharynx) and disorders of articulation (involving the oral cavity). Dysphonia can encompass a range of situations in which the child or their parents are dissatisfied with the child's quality of voice, often described as 'hoarse', 'husky', 'weak', etc. In a large community study in Avon, UK,[1] 11 per cent of 7389 children aged eight years were reported by their parents to be hoarse. Dysphonia was more common in boys and in those with older siblings. It is unlikely that all these children's parents would seek hospital referral for investigation and treatment; however, enough do for dysphonia to be a common presentation in paediatric otolaryngology practice.

Practice varies considerably in what, if any, investigations are performed and how the condition is managed in otolaryngology clinics. Although childhood dysphonia is mostly due to non-serious causes such as voice misuse and consequent vocal fold nodules,[2,3] very serious pathology such as papillomatosis or malignancy occasionally needs to be excluded, and treatable congenital anomalies such as webs and cysts can be missed. The role of laryngopharyngeal reflux (LPR) in dysphonia has been increasingly discussed in recent years.[4–6] Although the optimal means of assessment is still a subject of debate (methods include history alone, endoscopic examination, contrast swallow, single- and dual-channel pH studies, and intraluminal impedance measurement), the proportion of dysphonic children diagnosed with LPR seems to depend on how aggressively the diagnosis is pursued. It is not known whether *Helicobacter pylori* has a role in paediatric

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Accepted for publication: 31 October 2008. First published online 16 February 2009.

voice disorders, but there is some preliminary evidence from adults suggesting that *H pylori* may have a role in patients with vocal fold polyps.[7]

Guidelines for the management of paediatric dysphonia have been suggested, but there is currently no general consensus on the extent of investigation required. In one US study,[6] most, but not all, children underwent endoscopic examination of the larynx. Of those who did, 36 per cent had LPR alone, 29 per cent had vocal fold nodules alone, and 20 per cent had both LPR and vocal fold nodules. A more intensive policy of universal direct laryngoscopy, rigid bronchoscopy with bronchoalveolar lavage, and oesophagoscopy with biopsy was retrospectively reviewed in 127 children presenting with hoarseness in another US study.[8] Eight-two per cent of these children had vocal nodules, 43 per cent had endoscopically visualised laryngitis, 28 per cent had tracheobronchial inflammatory changes, 37 per cent had an abnormal bronchoalveolar lavage result and 30 per cent had an abnormal oesophageal biopsy result. Although the various findings did not correlate with each other, it is clear that positive bronchoalveolar lavage results and abnormal oesophageal biopsy findings are prevalent among children with hoarseness.

Previous studies have focused on a particular pathology or investigation, but no study has yet examined outcomes for all children presenting with dysphonia to a UK tertiary care unit. Therefore, it was thought worthwhile to review our current practice in order to produce evidence regarding workload and outcomes, which could inform future management of children with dysphonia (particularly since voice clinics, which are now well established in most adult health services in the developed world, are uncommon in children's services in the UK).

Methods

We retrospectively reviewed the records of all children (aged zero to 16 years) presenting to our unit with dysphonia as their primary complaint between January 2001 and October 2007. Cases were identified by performing a keyword search on the departmental computers, which contained a copy of every typed letter produced by our secretarial staff, using the keywords 'hoarse/hoarseness', 'dysphonia', 'gruff', 'croaky', 'guttural' and 'husky/huskiness'. As a typed letter is produced after every out-patient clinic visit and every surgical procedure, this provided us with a comprehensive database of the workload of our department. The patient records were scrutinised and relevant data extracted regarding patient demographics, clinical features, diagnostic investigations and final diagnoses.

Results

Patient characteristics

A total of 142 children were identified as having presented primarily with dysphonia over the eight years reviewed. Case records were found for 137 children (97 per cent). Eighty-three were male (61 per cent) and 54 female (39 per cent). The children ranged in age from two months to 15 years (median 5.3 years).

In 10 children (7 per cent), hoarseness was congenital, presenting as a hoarse, weak cry at birth. In 15 children (11 per cent), onset of hoarseness was related to a specific surgical procedure. In the remaining children (82 per cent), onset of hoarseness was spontaneous.

Associated symptoms at presentation included stridor (n = five), apnoeic episodes (two), dysphagia (two), foreign body sensation (three), failure to thrive (one), neck pain (one), post-nasal drip (11), sore throat (16), snoring (11), cough (17), heartburn (six), wheeze (three) and vomiting (three).

Laryngeal examination

Thirty children underwent mirror examination of the larynx in the out-patient clinic (22 per cent). Of these 30, seven children required further investigation with fibre-optic laryngoscopy or microlaryngoscopy-bronchoscopy under general anaesthesia. Laryngeal visualisation in the remaining 23 children (17 per cent) comprised only indirect laryngoscopy. Mirror examination was performed most often in children aged six years or older (84 per cent).

Twenty-three children (17 per cent) underwent awake fibre-optic laryngoscopy as their first examination, and four more underwent fibre-optic laryngoscopy after an initial mirror examination, giving a total of 27 children (20 per cent) for whom the most detailed examination was fibre-optic laryngoscopy. Thirty-nine children (29 per cent) underwent microlaryngoscopy-bronchoscopy under general anaesthesia as their first examination, and an additional three underwent microlaryngoscopy-bronchoscopy after mirror examination in the clinic, giving a total of 42 (31 per cent) who underwent microlaryngoscopy-bronchoscopy at some time. Forty children (32 per cent) underwent no laryngeal visualisation at any time and were diagnosed based on history alone. A further five children (4 per cent) were scheduled for direct laryngoscopy but this was not performed, either because the patient did not attend for the procedure or because the parents cancelled the procedure due to resolution of symptoms.

Therefore, 92 children (67 per cent) received some form of laryngoscopy and 45 (33 per cent) did not. Table I shows the children's final diagnoses according to whether or not the larynx was visualised.

Laryngoscopy was always carried out for children who presented with associated symptoms of stridor (n = five), apnoeic episodes (two), dysphagia (two), foreign body sensation (three), failure to thrive (one) or neck pain (one).

Diagnosis

The children's final diagnoses are listed in Table II.

The most common diagnosis was voice abuse, accounting for 62 (45 per cent) of all diagnoses. Of these children, 36 (58 per cent) had a diagnosis of vocal fold nodules confirmed by laryngoscopy. Four (11 per cent) of these children underwent indirect

https://doi.org/10.1017/S0022215109004599 Published online by Cambridge University Press

TABLE I

CHILDREN'S FINAL DIAGNOSES, BY LARYNGOSCOPY *VS* NO LARYNGOSCOPY

Diagnosis	Laryngoscopy?	
	Yes*	No†
Congenital anomalies	10	0
Iatrogenic	8	7
Inflammatory	10	8
Neoplastic	1	0
Voice abuse	49	13
No specific diagnosis	14	17

Data represent number of children. *$n = 92$; †$n = 45$.

laryngoscopy in the out-patient clinic, 11 (31 per cent) underwent fibre-optic laryngoscopy and 21 (58 per cent) received microlaryngoscopy-bronchoscopy. Fourteen children (23 per cent) were diagnosed based on history alone. A diagnosis of voice abuse was more common in boys than girls (39 males (63 per cent) versus 23 females (37 per cent)).

The second largest diagnostic group comprised children in whom no specific diagnosis was recorded in the case records. In 14 children, this was despite the fact that some form of laryngoscopy had been performed.

Table III shows the children's diagnoses by age. In children aged <2 yrs or younger, a wide range of pathologies was seen, including papillomas, congenital anomalies (cysts and webs), recurrent laryngeal nerve injury, reflux and intubation trauma. Children aged two to six years most commonly had voice abuse or no specific diagnosis recorded (69 per cent). Seven children in this age group presented with congenital anomalies (webs, cysts and congenital vocal fold palsies). While these lesions present more commonly in infants, the presentation in those cases is usually with stridor and such children would not have been included in this study. The children in this series presented with hoarseness, hence the older age at presentation than one might expect. Children aged seven years or older most commonly had a diagnosis of voice abuse or no specific diagnosis (80 per cent), but inflammatory conditions and intubation trauma were also seen.

TABLE II

CHILDREN'S FINAL DIAGNOSES

Diagnostic category	Specific diagnosis and notes	Children (*n*)
Voice abuse	Presumed on history alone	14
	Suggestive history, normal exam	12
	Nodules on laryngoscopy	36†
Inflammatory	Secondary to URTI	1
	Infective (associated with inhaled steroids)	1
	Reflux (presumed, no pH study)	7
	Reflux (demonstrated on pH study)	1
	Reflux (in association with laryngomalacia)	2
	Secondary to chronic rhinosinusitis	6
Neoplastic	Viral papillomas	1
Congenital	VF cyst	5‡
	Congenital VF palsy (unilateral)	2
	Cystic hygroma involving larynx	1
	Congenital glottic web	2
Iatrogenic	Minor intubation injury, self-limiting	4
	VF palsy after cardiac surgery	2
	VF palsy after excision of vagal neurofibroma	1
	Ant commissure granulation after tracheostomy	1
	After reconstruction for subglottic stenosis	7
No specific diagnosis	Laryngoscopy planned, not done	6
	Laryngoscopy done, normal	13
	Laryngoscopy not done	12

*n=137. †Two also had reflux. ‡One with secondary nodule formation. Exam = examination; URTI = upper respiratory tract infection; VF = vocal fold; ant = anterior

Management of voice abuse

Voice therapy under the direction of a speech and language therapist was arranged for 46 of the 62 children with a diagnosis of voice abuse (74 per cent). Voice therapy was not arranged for the other 16

TABLE III

CHILDREN'S DIAGNOSES BY AGE

Diagnosis	Age		
	<2 yrs	2–6 yrs	≥7 yrs
Voice abuse	3	26	33
No specific diagnosis	3	17	11
Neoplastic			
Viral papillomas	1		
Congenital			
Congenital glottic web	1	1	
Congenital VF palsy (unilateral)		2	
Cystic hygroma involving larynx	1		
VF cyst	1	4	
Iatrogenic			
Ant commisure granulation after tracheostomy	1		
Post-intubation	1	2	1
After laryngeal reconstruction for subglottic stenosis	2	4	1
VF palsy after cardiac surgery	2		
VF palsy after excision of plexiform neurofibroma of vagus nerve			1
Inflammatory			
Infective (associated with inhaled steroids)			1
Reflux (demonstrated on pH study)	3	4	3
Secondary to chronic rhinosinusitis	1	2	3
Secondary to URTI			1
Total	20	62	55

Mths = months; yrs = years; VF = vocal fold; ant = anterior; URTI = upper respiratory tract infection

https://doi.org/10.1017/S0022215109004599 Published online by Cambridge University Press

children, presumably because they were felt to be too young to comply with therapy.

Discussion

In any health survey, the range of diagnoses found will reflect the nature of the population being studied. In our large children's hospital, we provide a referral service for airway disorders for Scotland, and we support a cardiac surgery service that also covers the whole of Scotland. Accordingly, we encounter a number of children in whom dysphonia coexists with airway problems or is a consequence of surgery. However, regardless of the study population, voice abuse is the commonest reason for children to have a hoarse voice, as seen in our series and those reported by others.[6,8]

The proportion of children diagnosed with LPR depends to a certain extent on the enthusiasm with which this diagnosis is pursued, via laryngoscopy and other investigations. Laryngopharyngeal reflux was a minority diagnosis in our series, because we have only recently been persuaded that it has a significant role to play in the aetiology of dysphonia in children. In any event, there is conflicting evidence as to whether laryngoscopic findings are a reliable means of establishing a diagnosis of LPR.[9–11] This raises the question of whether suspected LPR should always be investigated by means of pH studies or intraluminal impedance studies, or whether an empirical trial of therapy is justified in the first instance. We have always tended towards the latter, pragmatic approach, hence the number of presumed diagnoses of LPR in Table II. Initially, such empirical therapy would comprise an H_2-antagonist, with or without an alginate, but more recently we have started to use proton pump inhibitors as first-line therapy.

The role of investigation in the management of children with dysphonia is also unclear when considering whether every child with dysphonia requires laryngoscopy. While such a hard-and-fast rule seems attractive, dysphonia is most prevalent in children aged two to six years, in whom laryngoscopy usually requires a general anaesthetic. We must at least ask the question of whether the information gathered justifies a policy of routine anaesthetic examination, when the majority of children have a non-surgical, non-life-threatening condition (i.e. voice abuse, with or without nodule formation). It would be reasonable to argue, for example, that children aged two to six years who have a clear history of regular shouting, and who have an intermittent voice problem that resolves completely at times, are at very low risk of significant pathology and could be managed initially by voice therapy with early clinical review.

Congenital lesions (such as cysts and webs) and tumours can only be diagnosed on visualisation of the larynx. Persistent dysphonia, or dysphonia associated with any airway obstruction, pain or dysphagia, should be investigated promptly, and visualisation of the larynx is clearly mandatory in such cases. Regarding assessment of the children in our series aged two to six years, we suggest that those with a history of recent intubation or laryngeal surgery would be easy to identify, and those with congenital lesions would have persistent dysphonia that does not recover to normal at any point. It should be possible to identify that the latter children require microlaryngoscopy-bronchoscopy (Table IV). The remaining two- to six-year-olds all had non-serious pathology, and it would be reasonable to avoid anaesthesia for them if possible.

- **Dysphonia is common in children**
- **The cause is generally voice abuse, but occasionally the cause is serious (e.g. papillomas or tumour) or requires surgical treatment (e.g. cysts or webs)**
- **While laryngeal visualisation of every case would be ideal for diagnosis, in children this often requires general anaesthesia, the risks of which must be weighed against the benefits of the additional information obtained**
- **In this series of dysphonic children, in a large UK children's hospital, laryngoscopy was not routinely performed**
- **Although voice abuse was the most common diagnosis encountered, a wide range of pathology may present primarily with dysphonia**
- **There may be a small group of children with clear evidence of voice abuse who can be managed initially with a trial of voice therapy; however, in all other cases some form of laryngoscopy is mandatory**

The infants in our series had a wide variety of diagnoses, some serious, and we feel that examination of the larynx is essential in this age group. Infants tolerate awake transnasal fibre-optic laryngoscopy sufficiently well for us to suggest that mandatory laryngoscopy should be possible without problems. In most cases, topical anaesthesia and lubrication are unnecessary and unhelpful, but a small-bore endoscope should be used (diameter 1.8 mm). If fibre-optic laryngoscopy fails to provide an adequate view or a clear diagnosis, then microlaryngoscopy under general anaesthesia is required.

In our series, many older children (aged approximately seven years and older) tolerated the laryngeal mirror without topical anaesthesia. Children in this

TABLE IV

'RED FLAG' SYMPTOMS MANDATING PROMPT AND THOROUGH ENDOSCOPIC ASSESSMENT

Throat pain or referred ear pain
Dysphagia
Stridor
Age <2 yrs
Weight loss or failure to thrive
Persistent (rather than intermittent) dysphonia
Lack of reponse to voice therapy

https://doi.org/10.1017/S0022215109004599 Published online by Cambridge University Press

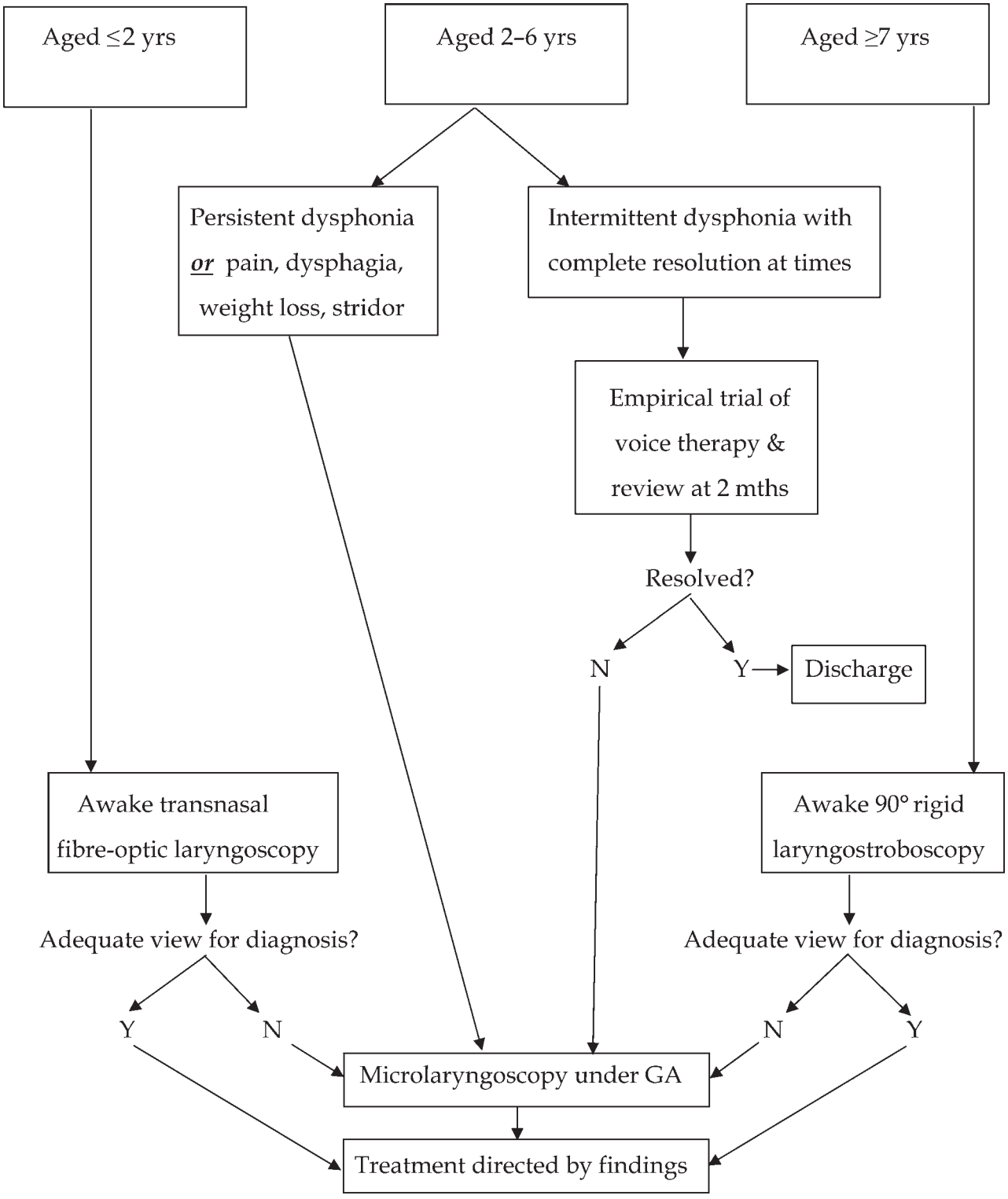

FIG. 1
Suggested algorithm for laryngoscopy in the child with dysphonia. Mths = months; yrs = years; N = no; Y = yes; GA = general anaesthesia

older age group can often be gently persuaded to tolerate awake laryngoscopy with either a transnasal flexible laryngoscope or (ideally) a rigid 90° instrument with stroboscopy, as long as the child is handled with sensitivity. Topical anaesthesia of the oropharynx is less important than gentle technique, but it can be used if required. Again, failure to obtain an adequate view for diagnosis should lead to microlaryngoscopy-bronchoscopy under anaesthetic.

https://doi.org/10.1017/S0022215109004599 Published online by Cambridge University Press

Based on our experience, we propose a pragmatic management algorithm for the dysphonic child, presented in Figure 1.

It is clear that the majority of children to whom this algorithm is applied would undergo some form of laryngoscopy, but that a small number of children aged two to six years with a clear history of voice abuse would be spared an anaesthetic. We believe this is a fair compromise. In any event, it was only on collating the figures for this paper that we became aware of the high proportion of children attending our service who did not undergo any visualisation of the larynx. In many cases, this will have been because the symptoms had resolved spontaneously before the child was seen, but such a high proportion is still unexpected. We have no evidence that any child came to harm from this, and we have no reason to believe that any important diagnoses were missed. We do feel, however, that what happens in our hospital may well be happening in many others.

Conclusion

Childhood dysphonia is a common reason for referral, but it's management can be variable. A more systematic approach to the investigation of dysphonia in children can only be beneficial, and we believe there is a strong case for organising this through a dedicated paediatric voice clinic.

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Mr H Kubba takes responsibility for the integrity of the content of the paper.
Competing interests: None declared