PEDIATRIC BODY MRI



Magnetic resonance imaging of obstructive sleep apnea in children

Robert J. Fleck ^{1,2,3} • Sally R. Shott ^{4,5} • Mohamed Mahmoud ⁶ • Stacey L. Ishman ^{4,5} • Raouf S. Amin ⁷ • Lane F. Donnelly ^{8,9}

Received: 17 March 2018 / Revised: 9 May 2018 / Accepted: 10 June 2018 $\hfill {\Bbb C}$ Springer-Verlag GmbH Germany, part of Springer Nature 2018

Abstract

Sleep-disordered breathing has a spectrum of severity that spans from snoring and partial airway collapse with increased upper airway resistance, to complete upper airway obstruction with obstructive sleep apnea during sleeping. While snoring occurs in up to 20% of children, obstructive sleep apnea affects approximately 1–5% of children. The obstruction that occurs in obstructive sleep apnea is the result of the airway collapsing during sleep, which causes arousal and impairs restful sleep. Adenotonsillectomy is the first-line treatment of obstructive sleep apnea and is usually effective in otherwise healthy nonsyndromic children. However, there are subgroups in which this surgery is less effective. These subgroups include children with obesity, severe obstructive sleep apnea preoperatively, Down syndrome, craniofacial anomalies and polycystic ovarian disease. Continuous positive airway pressure (CPAP) is the first-line therapy for persistent obstructive sleep apnea despite previous adenotonsillectomy, but it is often poorly tolerated by children. When CPAP is not tolerated or preferred by the family, surgical options beyond adenotonsillectomy are discussed with the parent and child. Dynamic MRI of the airway provides a means to identify and localize the site or sites of obstruction for these children. In this review the authors address clinical indications for imaging, ideal team members to involve in an effective multidisciplinary program, basic anesthesia requirements, MRI protocol techniques and interpretation of the findings on MRI that help guide surgery.

Keywords Adenoids \cdot Children \cdot Laryngomalacia \cdot Lingual tonsils \cdot Macroglossia \cdot Magnetic resonance imaging \cdot Obstructive sleep apnea \cdot Surgery

Electronic supplementary material The online version of this article (https://doi.org/10.1007/s00247-018-4180-2) contains supplementary material, which is available to authorized users.

Robert J. Fleck Robert.Fleck@CCHMC.org

- ¹ Department of Radiology, Cincinnati Children's Hospital Medical Center, University of Cincinnati College of Medicine, 3333 Burnet Ave., Cincinnati, OH 45229, USA
- ² Center for Pulmonary Imaging Research, Cincinnati Children's Hospital, Cincinnati, OH, USA
- ³ Imaging Research Center, Cincinnati Children's Hospital, Cincinnati, OH, USA
- ⁴ Division of Pediatric Otolaryngology–Head and Neck Surgery, Cincinnati Children's Hospital Medical Center, Cincinnati, OH, USA

- ⁵ Department of Otolaryngology–Head and Neck Surgery, University of Cincinnati School of Medicine, Cincinnati, OH, USA
- ⁶ Department of Anesthesia, Cincinnati Children's Hospital Medical Center, University of Cincinnati College of Medicine, Cincinnati, OH, USA
- ⁷ Division of Pulmonary Medicine, Cincinnati Children's Hospital Medical Center, University of Cincinnati College of Medicine, Cincinnati, OH, USA
- ⁸ Department of Radiology, Stanford University, Stanford, CA, USA
- ⁹ Quality and Safety, Lucile Packard Children's Hospital, Stanford, CA, USA

Introduction

Magnetic resonance imaging has been increasingly used to help localize and diagnose the site or sites of obstruction in children with obstructive sleep apnea that persist despite previous adenotonsillectomy. Donnelly [1] in 2005 described the protocol for using dynamic MRI for this purpose. The current review article serves to update this protocol.

Sleep-disordered breathing has a spectrum of severity that spans from snoring and partial airway collapse with increased upper airway resistance, to complete upper airway obstruction with apnea during sleeping. Snoring can be present with all levels of severity of obstructive sleep apnea and is the symptom that is most indicative of possible obstructive sleep apnea [2]. At its most severe, sleep-disordered breathing causes multiple obstructive episodes each hour resulting in arousal that disturbs restful sleep. When obstructions occur with associated drops in the oxygen saturation or an arousal from restful sleep, then obstructive sleep apnea is present.

Obstructive sleep apnea affects 1–5% of the pediatric population, although it can occur with much higher frequency in children with syndromes and craniofacial abnormalities [1]. Untreated pediatric obstructive sleep apnea can result in cognitive and behavioral problems that can affect academic performance as well as learning [2–5]. Untreated obstructive sleep apnea can also have more serious complications of pulmonary hypertension, systemic hypertension and cor pulmonale.

The polysomnogram, or sleep study, is the standard by which obstructive sleep apnea and other sleep disorders are diagnosed and severity determined [2]. Pediatric polysomnogram scoring commonly uses a 3% desaturation criterion, unlike adult scoring, which often uses a 4% desaturation criterion [3, 4]. In addition, pediatric obstructive sleep apnea severity definitions differ from those in adults. Severity is determined using the apnea hypopnea index, which is the number of obstructive apneas and hypopnea episodes that occur during sleep, divided by the total sleep time. In children, normal is defined as <1 event per hour. Mild obstructive sleep apnea is from 1 event to <5 events per hour. Moderate obstructive sleep apnea is 5 events to <10 events per hour, and severe obstructive sleep apnea occurs if there are 10 or more events per hour [5].

In general, first-line treatment for obstructive sleep apnea in children is adenotonsillectomy, and this is effective in most healthy nonsyndromic children. However in a large multicenter study of 578 "normal" children undergoing adenotonsillectomy, the average apnea hypopnea index decreased from 18.2 events/h to 4.1 events/h, and only 27% were "cured" (apnea hypopnea index < 1/h) after adenotonsillectomy [6]. Significant risk factors for persistent obstructive sleep apnea after adenotonsillectomy include obesity, severe obstructive sleep apnea preoperatively, and asthma in nonobese children [6]. In children who don't fully respond to adenotonsillectomy, the next line of treatment is frequently long-term continuous positive airway pressure (CPAP), but this is poorly tolerated by children because of its discomfort. Additionally, the contribution of CPAP to disruption of parental rest can contribute to poor adherence [2]. These children with persistent obstructive sleep apnea, poor adherence to CPAP, and a willingness to entertain surgical options are best served by dynamic cine MRI of the upper airway.

Imaging team

The majority of pediatric institutions do not routinely use MRI for treatment planning for persistent obstructive sleep apnea. At institutions that do have such a program, orders for these studies are generally driven by an otolaryngologist who is interested in offering surgical options to families of affected children and who is familiar with the use of MRI in guiding treatment decision-making [7]. A successful program also requires implementation of a standard MRI sleep study protocol (Online supplementary material). This requires an anesthesiologist who is willing to take the lead in providing general anesthesia in this group of children with critical airways [8]. The anesthesiologist must be comfortable in providing anesthesia to a child with a compromised airway without an artificial airway in place.

Magnetic resonance imaging technique

Imaging protocols should be set up and tested in awake volunteers prior to imaging a child under anesthesia. Imaging for obstructive sleep apnea can be performed on all MRI equipment, either 1.5 T or 3.0 T, using current commercially available sequences, hardware and coils. Generally, the best imaging characteristics are obtained from phased-array head and neck vascular coils, which allow for imaging acceleration with parallel imaging. As with all imaging, a 3-D localizer is performed to make certain that the head and neck are in neutral position (Fig. 1) and that the area of interest is centered in the MRI field and coil. The first sequence is a sagittal 3-D respiratory-triggered fast spin echo, proton-density sequence (PD CUBE by GE Healthcare, Waukesha, WI; VISTA [Volume ISotropic Turbo spin-echo Acquisition] by Philips Healthcare, Best, the Netherlands; or SPACE [Sampling Perfection with Application-optimized Contrast using different flip-angle Evolution] by Siemens Healthcare, Erlangen, Germany). This sequence covers from the tip of the nose anteriorly to the occiput posteriorly and the superior aspect of the nasal airway superiorly and to the subglottic trachea inferiorly. Specific imaging parameters used are: field of view (FOV)=20-24 cm, matrix 252 × 252, slice thickness 1.6 mm



Fig. 1 MRI in a 19-year-old man with Down syndrome and obstructive sleep apnea, with an apnea hypopnea index of 36 events per hour. **a** Midline sagittal T2-weighted image with fat saturation shows enlarged lingual tonsils (*) measuring 11-mm thick at midline. Very small adenoids are seen in the midline. Note that positioning of the head is not optimal for observing the airway in a neutral position. **b** Axial T2-weighted image with fat saturation shows the full extent of the airway narrowing and lingual tonsil enlargement (*). Both images were acquired with a radial

acquisition (PROPELLER [periodically rotated overlapping parallel lines with enhanced reconstruction] or MultiVane) and respiratory triggering, which substantially reduced the motion and blurring compared to the sagittal 3-D respiratory-triggered proton-density (PD) sequence (PD CUBE by GE Healthcare, Waukesha, WI; VISTA [volumetric isotropic T2W acquisition] by Philips Healthcare, Best, the Netherlands; or SPACE [sampling perfection with application optimized contrasts using different flip angle evolution] by Siemens Healthcare, Erlangen, Germany)

and gap 0.8 mm. The advantage of this sequence is that respiratory motion is minimized, voxels are isotropic, and reconstruction can be performed in any plane to visualize and measure structures.

Cine imaging is important for evaluating obstructive sleep apnea. This dynamic exam can show both the degree and the direction of airway collapse, e.g., anterior-posterior (AP) or circumferential. It also shows ways in which children are sometimes compensating or reacting to their periods of obstruction with tongue and jaw thrusting. Dynamic airway imaging is performed using a single-slice multi-phase gradient echo sequence acquired in the axial and sagittal planes. Imaging parameters are selected to obtain as high a spatial resolution as possible (approximately 1.68 mm \times 2.62 mm \times 5.00 mm in frequency, phase and slice, respectively) while maintaining a target temporal resolution of 300-400 ms/image over 30-35 s (to capture roughly 90 dynamic phases). The repetition time (TR) and echo time (TE; ideally fat and water in phase) are set accordingly, with the flip angle then selected to maximize signal-to-noise ratio. Representative acquisition parameters for an axial dynamic airway acquisition at 1.5 T are FOV= 215×210 mm (frequency x phase), matrix (frequency x phase)= 128×82 , TR/TE=5.49/3.24 ms, flip angle= 15° , acceleration factor=1.5; receiver bandwidth=434 Hz/pixel, signal averages=1.

Cine imaging is performed for approximately 30 s at each level with the rate of imaging of about three images per second, but faster is better. The rate of imaging and quality depends on the field strength, gradient strength, slew rate, matrix and signal-to-noise acceptability with acceleration techniques such as parallel imaging. Newer k-space sampling trajectories, such as compressed sensing, are coming available and are expected to accelerate the temporal resolution.

The initial slice is a sagittal midline cine of the airway set up on the reconstructed axial and sagittal views from the prior 3-D proton-density imaging. This allows for precision in bisecting the airway in a true midline sagittal plane (Fig. 2). This is important for accurate depiction of the multilevel obstruction that can occur in obstructive sleep apnea, often involving the nasopharynx and retroglossal region. Next, an axial cine, oriented perpendicular to the airway, is obtained of the retroglossal airway at the midpoint (superiorly to inferiorly) between the tip of the uvula and the epiglottis (Fig. 2). The final cine image is obtained perpendicular to the narrowest portion of the nasopharyngeal airway, which is usually the narrowest section of the entire upper airway (Fig. 2) [9]. More recently, we have begun performing an axial stack of cine images extending from the nasal choana to the glottis and we have observed changes in the pattern of airway motion along the axis of the airway. This is of unknown clinical significance at this time.

Axial and sagittal short tau inversion recovery (STIR), or T2-weighted MRI with fat saturation, are very good for visualizing palatine, adenoid and lingual tonsillar tissues, as well as lymphoid tissues and edema (Fig. 1), because these structures appear bright on a dark background. The use of a radial acquisition such as propeller or multivane is robust against motion and blurring. Typically these images are obtained with Fig. 2 MRI in a 15-year-old girl with Down syndrome and persistent obstructive sleep apnea after lingual tonsillectomy, with an apnea hypopnea index of 5.1 events per hour. a Midline sagittal 3-D proton-density respiratorytriggered image with 0.8-mm isotropic resolution. Solid line depicts placement of imaging plane for cine of the retroglossal airway and the dashed line shows placement of the plane for cine imaging of nasopharyngeal airway. b Coronal reformat of the data from (a) shows the length of the airway from the nasopharynx (A), uvula/soft palate (B), epiglottis (C) to subglottic trachea (D). Dashed line shows the orientation of the midline sagittal plane for cine of the upper airway. Using the reformatted image allows adjustment of the imaging plane so that the plane bisects the airway along the entire length, which can depict any dynamic multilevel obstruction that might occur. c Axial reformat of the data from the solid line in (a) shows the midline sagittal plane oriented to the mid retroglossal airway and apex of the mandible. Children can rotate their heads during imaging, so all planes should be checked on two images. d Sagittal midline cine gradient echo sequence available as a movie online shows soft-tissue movement at base of tongue causing intermittent obstruction at the level of the epiglottis. (Online supplementary material - Cine 1)



a FOV=20-24 cm, matrix= 240×128 , and slice thickness=5 mm. T1-weighted imaging is optional but can be performed as a routine if desired.

Some contraindications to MRI do exist. Screening for electronic devices such as pacemakers, defibrillators and vagal nerve stimulators should be routine for MRI because they are contraindications. Although the newest generation of hypoglossal nerve stimulators might be MRI-compatible, the older devices are not, and this must be clarified prior to starting the procedure.

Braces, metallic dental caps and palate expanders are a relative contraindication to MRI in obstructive sleep apnea

because the artifact produced can obscure the anatomy of interest. Because these studies are performed under anesthesia, it is wise to screen carefully for the presence of orthodontic hardware. We have found the amount of artifact to be variable, so we often put the child in the MR scanner prior to anesthesia to determine the extent of artifact and whether the relevant anatomy is visible and assessable (Fig. 3).

Children with severe obstructive sleep apnea who require CPAP might be unable to complete the study without unacceptable decreases in oxygen saturation. Anesthesia providers might want to use CPAP or a nasal trumpet to maintain



Fig. 3 Dental hardware in a 6-year-old girl with Pierre–Robin sequence, Chiari malformation and cleft palate post repair, and pharyngeal flap. The girl was positioned in the MRI scanner prior to imaging under druginduced sleep to determine the extent of susceptibility artifact created by the dental hardware. **a** Midline sagittal 3-D proton-density image shows the susceptibility artifact (*arrowhead*) only involving the anterior

portion of the mouth and allowing the imaging procedure to continue. Note the thick soft palate with heterogeneous signal intensity oriented horizontally because of the pharyngeal flap (*arrow*). **b** Axial reformatted slice from the same data set at the level of the soft palate shows the bilateral apertures (*arrows*) of the airway created by the pharyngeal flap procedure

acceptable oxygen saturation. CPAP is preferred over a nasal trumpet. However oral airways (Fig. 4) and laryngeal mask airways are not appropriate for this particular indication



Fig. 4 7-year-old male with fragile X syndrome and obstructive sleep apnea. Sagittal 3D proton density imaging was acquired with oral airway (outlined by arrowheads) still in place distorting the tongue and pushing the soft palate posteriorly. Additionally, the head is not in a neutral position. The oral airway was removed and imaging continued.

because of the extensive distortion they cause of the natural anatomy and motion.

If necessary, all sequences can be performed with the supplemental airway support (CPAP or nasal trumpet), including the sagittal and axial cine imaging. Then the supplemental airway support is removed and the sagittal and axial cine imaging is repeated after removal of the airway support so that the motion of the airway can be evaluated in its natural state. Airway support is discussed in detail in the next section.

Patient preparation and anesthesia

The children coming to MRI are usually those who have undergone adenotonsillectomy but still have obstructive sleep apnea on postoperative polysomnogram and are not tolerating CPAP or want to consider surgical options. Because symptoms are only present during sleep, MRI is performed during anesthesia, akin to drug-induced sleep endoscopy. Obtaining ideal dynamic airway images during sleep MRI studies requires that the child be in a state mimicking physiological sleep as closely as possible, while maintaining spontaneous breathing via the native airway. Maintaining cardiorespiratory parameters such as oxygen saturation, respiratory rate and blood pressure within a safe range can be a challenge during dynamic airway evaluation [10]. Historically, sedation has been performed under radiology's structured sedation program with pentobarbital [1, 11]. However, it is optimal for anesthesia specialists to supervise all such studies. Initially, we used propofol for these studies, but it was recognized that this agent has dose-dependent respiratory depression [9] and the agent of choice became dexmedetomidine, which does not cause respiratory depression; use of dexmedetomidine is included in the procedure describe next [12].

The child is placed in neutral neck position with the Frankfort angle (angle from the inferior orbital rim to the auditory canal relative to horizontal) at 90°. Neck extension, shoulder rolls and other maneuvers are avoided to maintain as natural of a supine position as possible. A head strap is positioned under the head so that a mask can be used for CPAP if required to maintain airway patency [13]. The child is put to sleep with sevoflurane in the induction room and intravenous access is secured. A bolus of dexmedetomidine (1 mcg/kg) is administered and then maintained with an infusion of 1 mcg/ kg/h [14]. Ketamine at 1 mg/kg might be added to the dexmedetomidine protocol, and this is used when a combination of drug-induced sleep endoscopy and cine MRI are performed together. The advantages of using this combination of drugs are beyond the scope of discussion for this review article; details of the sedation protocol are available in the references [15, 16].

Airway management during anesthesia

As stated, it is never desirable to have oral airways or laryngeal mask airways present when imaging the upper airway because it distorts and obscures the anatomy and physiology that is being imaged (Fig. 4). Usually an oral airway is placed during induction and is removed prior to commencement of imaging. We prefer CPAP via facemask to a nasal trumpet in order to maintain airway patency and oxygenation during cine MRI because it does not distort anatomy in an unnatural way, and pressure support is easily removed without arousing the child [13]. There is a caveat to using a full face mask: On rare occasions the air pressure administered orally by a full facemask can push the tongue posteriorly and narrow the retroglossal airway. This occurs when there is no open path for air from the open mouth anteriorly to the posterior pharynx, usually caused by an enlarged tongue obstructing the oral passage of air [17]. The next best option is a nasal trumpet, although this can distort the anatomy and is not visible on MRI.

When either CPAP or a nasal trumpet is needed to support the airway, it is best to perform all the imaging, including the cine sequences, with these in place. When image acquisition is completed, turn off the CPAP or remove the nasal trumpet and repeat the cine sequences to re-assess the native airway movement. CPAP is easiest to remove and children do not awaken with the change. Removing a nasal trumpet is more difficult and sometimes arouses the child.

Evaluation of airway motion

The normal airway changes during breathing because of physiological pressure changes created during the act of breathing and by the laws of fluid mechanics. For children with or without obstructive sleep apnea, the tendency for airway collapse during inspiration is countered by adequate neuromuscular action resulting in dilation; however, normal airway in a child without obstructive sleep apnea shows minimum change in caliber with breathing during sleep [18].

Because we do not perform sedated imaging in normal children without obstructive sleep apnea, it is difficult to make a statement about how much change in caliber is abnormal, but we can make some general statements. Severe obstructive sleep apnea is associated with greater change in the caliber of the airway during imaging than is seen in children without obstructive sleep apnea [18]. Generally a change in caliber of 50% is abnormal and a change of greater than 80% is very abnormal based on our experience. During hypopneas the airway usually decreases in size but doesn't totally obstruct, so if imaging occurs during a hypopnea the airway might appear relatively small and stable. It would be best to know airflow to better interpret the motion observed. After an obstruction or hypopnea, the airway of children with obstructive sleep apnea often balloons and then collapses repeatedly (Fig. 5) because of a physiological response known as "loop-gain" [19] before settling to a stable pattern. Loopgain is a term borrowed from electronics to explain the observed physiological response to the feedback from hypoxia caused by the apnea. The physiological response to the hypoxia is increased drive of breathing and arousal resulting in the observed changes of the airway.

The midline sagittal cine is the plane that allows for visualization of the entire airway at the same time and is best for detecting multilevel airway collapse (Fig. 5). The nasopharynx is typically the narrowest portion of the upper airway [9] and in children adenoid regrowth is a common cause of narrowing. The retroglossal airway is often very dynamic and on sagittal images appears to collapse anterior to posterior. However, it needs to be interpreted in conjunction with the axial retroglossal and nasopharyngeal cine images to rule out a circumferential collapsing pattern. Motion can be deceiving on the sagittal images alone because volume averaging in the sagittal slice makes circumferential airway collapse appear to be occurring in an anterior–posterior plane only.

Axial evaluation of airway motion is the most precise and can depict three types of pathological collapse: hypopharyngeal collapse (Fig. 5), glossoptotic collapse (Fig. 6) and lateral wall collapse. Hypopharyngeal collapse

Fig. 5 A 9-year-old boy with Down syndrome and severe obstructive sleep apnea with apnea hypopnea index of 142 events per hour. a Midline sagittal cine gradient recalled echo (GRE) image when the airway is open shows the enlarged, recurrent adenoids. b The next frames show the airway collapsed and closed. c Axial retroglossal cine GRE image of the airway (arrows) ballooned open. d Subsequent complete collapse in this boy demonstrates the concentric collapse seen in hypopharyngeal collapse (arrows). This boy underwent a midline posterior glossectomy, genioglossus advancement and revision adenoidectomy with postoperative apnea hypopnea index of 3.4. Online supplementary material: Sagittal and axial cine images show the movement of the tongue. If viewed only in sagittal this motion could be mistaken for glossoptosis (Cine 2) but Cine 3 shows the true nature of the collapse is circumferential





Fig. 6 A 15-year-old boy with obesity, obstructive sleep apnea and headaches with poor compliance to CPAP. **a** Axial gradient recalled echo (GRE) cine image of the retroglossal airway shows a wide patency and the arrows show posterior tongue surface. This is probably during peak expiration. **b** The next frame of the cine shows the tongue collapsing posteriorly (*arrows*). This is probably occurring during a period of cessation of airflow during transition to inspiration because of the rapid motion

indicated by the blurring of the tongue edge. **c** There is a small central airway opening and the posterior tongue is in contact with the posterior wall of the pharynx for three frames (about 1 s) during inspiration and then on expiration the airway "pops" open again. The boy and his family improved compliance to CPAP and the headaches resolved. Online supplementary material: An axial cine of the retroglossal segment shows the anterior posterior motion of the tongue consistent with glossoptosis (Cine 4)

occurs either from decreased neuromuscular activation of the dilator muscles or as a secondary phenomenon of a more superior obstruction. Circumferential hypopharyngeal collapse is a pattern of collapse where the lateral, posterior and anterior walls all move centrally, narrowing the airway (Fig. 5). This type of collapse is the hardest to address surgically and these children are often recommended to consider bony framework surgery, such as a 2-jaw surgery, or to reconsider the use of CPAP.

Glossoptosis collapse occurs with anterior and posterior motion of the tongue (Fig. 6). Glossoptosis is a dynamic process of the tongue in which the tongue moves posteriorly and then anteriorly. If the tongue contacts the posterior wall the surface adhesion might be more difficult for the dilator muscles to overcome. Glossoptosis is not necessarily correlated with worse obstructive sleep apnea when compared to hypopharyngeal collapse. Both can result in significant obstruction. Often the tongue is retro-positioned because of the size of the tongue or poor neuromuscular tone, but the static retro-position of the tongue is different from glossoptosis. This can be addressed surgically with hyoid suspension or a tongue suspension suture, which can be coupled with reduction in the tongue volume through posterior midline glossectomy.

Lateral wall collapse is the least common pattern of airway collapse [20] and seems to be related to the child's ability to keep the tongue anteriorly positioned. In some cases there is an increase in the anteroposterior diameter while the lateral walls collapse. Seen with this is forward "thrusting" of the tongue in an effort to maintain a patent airway. This is typically addressed with lateral expansion pharyngoplasty or bony surgeries that expand the posterior airway such as genioglossal advancement, hyoid suspension and 2-jaw surgery.

Tongue thrusting is one of several compensatory motions (head bobbing, jaw thrusting and neck extension) that can be observed on the cine imaging. Head bobbing either up and down or back and forth in a nodding motion and neck motion often manifests as an accentuation of the extension of the neck. In children with Down syndrome, the tongue is often sticking out of the confines of the oral cavity between the teeth and the anterior portion is moving forward with each respiratory cycle in a thrusting motion. These motions are often very dramatic. A consistent relationship between these compensatory motions and severity of obstructive sleep apnea has not been proved. It could be that this is an effort to keep the airway open and the effectiveness of the movement is variable for each child.

One of the real difficulties in interpreting the movement of the airway during imaging is that it is unclear whether airway motion is secondary to forces related to upstream obstruction because the direction and magnitude of airflow at the time of imaging are unknown, or whether it is a result of neuromuscular activation intended to keep the airway open. Figure 6 illustrates the narrowing of the airway related to airflow. Alternatively, the effect of neuromuscular control is illustrated in Fig. 7. This figure shows the cross-sectional area of an upper airway in a child with obstructive sleep apnea in relation to tidal volume, which is depicted as an idealized loop. In this illustration, the upper airway expands at the beginning of expiratory air flow (C-D) and then collapses when expiration airflow ceases at point E [21]. The difference between segments E-D and A-C is exaggerated in a child with obstructive sleep apnea when compared to those without obstructive sleep apnea and is dependent on the volume of airflow. Segment B represents the period of airway recovery (inspiration) during which the dilator muscles are activated to allow airflow through a patent airway [21]; this segment is dysfunctional in many children with obstructive sleep apnea. Unfortunately, during clinical imaging we can only make assumptions regarding the direction of the air flow and really do not know the magnitude of airflow; however advances in real-time monitoring of airflow during imaging are being developed [22].

Evaluation of anatomical structures

Anatomical evaluation is an important component of the MRI evaluation in obstructive sleep apnea in children who



Fig. 7 This is an idealized loop of airway motion in children with obstructive sleep apnea adopted from Schwab et al. [21] and more recent work of our own. The segment extending from A to C is during inspiration; segment C–D is during the transition from inspiration to expiration airflow; segment D–E is during expiration when the airway is largest. The difference between segments E–D and A–C is exaggerated in obstructive sleep apnea compared to normal and depends on the volume of airflow. Segment B is airway recovery caused by the dilator muscles to allow flow through the airway on inspiration [21], and this is dysfunctional in many children with obstructive sleep apnea

have persistent obstructive sleep apnea despite previous adenotonsillectomy because hypertrophied lingual tonsils and adenoid regrowth are all commonly seen in these children. Macroglossia, with the tongue enlarged relative to the size of the oral cavity, is also common [23]. The T2weighted images show regrowth of adenoids and enlarged lingual tonsils because of their bright signal as compared to a dark background. The isotropic proton-density 3-D images allow for precise reconstruction in any plane to show the relationship of anatomical narrowing to the thickness of these tissues. The palatine tonsils are generally absent because adenotonsillectomy is the first-line treatment for obstructive sleep apnea in this population, but if they are present, they can be assessed.

Evaluation for recurrent adenoidal tissue narrowing the nasopharyngeal airway is important because this is typically the narrowest portion of the upper airway [9]. Adenoids can mainly be removed at midline only because of close proximity to the Eustachian tubes and desire to avoid damage to these important structures (Fig. 8). The Eustachian



Fig. 8 Residual adenoid tissue in a 16-year-old boy with Down syndrome post adenotonsillectomy with poor adherence to CPAP and an apnea hypopnea index of 9.4. This axial oblique 3-D proton-density MR image through the nasal pharyngeal airway while on 12 cm of CPAP nicely shows the recess of the Eustachian tubes outlined by adenoid tissue (*short arrow*) centrally and the tubal tonsils laterally (*long arrows*). The close association of the torus tubarius and the desire to not cause damage during adenoidectomy is why adenoid tissue is not completely removed and can recur. *CPAP* continuous positive airway pressure

tubes also have tonsils of their own. Residual adenoidal tissue and the lymphoid tissue around the Eustachian tubes can hypertrophy and form a new, obstructing pad of tonsillar tissue. The recurrence is often visible on a lateral pharyngeal radiograph but thickness and detailed anatomy are better appreciated on MRI. In the sagittal plane, recurrence is implied by a thick adenoid pad, >12 mm, and is often associated with midline collapse or narrowing of pharyngeal airway by >70% [24].

In the past, recurrence of the palatine tonsils was not a concern because the traditional tonsillectomy technique removed the entire encapsulated tonsil. However, more recently some surgeons are performing intracapsular partial tonsillotomies, and in these cases recurrence of the palatine tonsils can occur.

The soft palate and adenoid tissue can interact and cause airway obstruction at the level of the nasopharynx as the palate collapses against the adenoid tissue. A vertical-oriented, elongated palate extending past the mid tongue or close to the epiglottis is generally considered abnormal. Abnormal soft palates demonstrate mild increase in the T2 signal relative to the tongue and sometimes the soft palate is thickened. A number of surgical palatoplasty techniques (e.g., lateral expansion pharyngoplasty) used by otolaryngologists are intended to decrease retropalatal obstruction and improve airflow.

In addition, obstruction can occur from an enlarged or glossoptotic tongue. Frequently, the tongue pushes on the soft palate, either due to the tongue's large size or its movement. The soft palate is then pushed up against the adenoid tissue or the nasopharyngeal ceiling, resulting in obstruction [23]. In these cases, tongue reduction or forward displacement of the tongue through hyoid suspension, tongue suspension or bony surgery might be indicated.

Lingual tonsils lie on the posterior surface of the tongue and are typically less than 5 mm thick and often barely seen on T2-weighted MRI in normal children (Fig. 1). Enlargement might be related to removal of other tonsillar tissue, reflux and obesity [25, 26]. Gastroesophageal reflux is often exacerbated, with greater changes in intrathoracic pressure seen with obstructive sleep apnea from breathing against obstruction. When enlarged, the lingual tonsils fill the retroglossal airway, causing narrowing in the AP direction and filling the vallecular space, pushing the epiglottis posteriorly. Because of extreme motion at the base of the tongue in some children, the lingual tonsils can be difficult to evaluate if respiratory triggering is marginal, but we have found that T2-weighted imaging in the sagittal and axial planes with propeller technique are robust in reducing motion artifact. A recent meta-analysis by Kang et al. [27] looked at the effect of lingual tonsillectomy on obstructive sleep apnea in four pediatric studies and include 73 subjects. The meta-analysis showed a mean reduction of the apnea hypopnea index by 8.9, but an apnea hypopnea index of less than 5 was obtained in 51%, and an apnea hypopnea index less than 1 in only 17% [27].

Enlargement of the tongue is present in many of the children imaged for obstructive sleep apnea, especially in those with Down syndrome. Evaluation of tongue size is difficult, but helpful signs of significant enlargement include the posterior aspect of the tongue abutting and displacing the soft palate. Another helpful sign is the degree to which the tongue protrudes posteriorly over the line that extends from the anterior border of the trachea (Fig. 2). If the tongue is abnormally posteriorly positioned and the position is static, it is said to be retropositioned, and when there is AP motion as judged on the axial cine, it is glossoptosis. The tongue can be both retropositioned and glossoptotic.

Fatty infiltration of the tongue is often present on proton-density images when there is no fat suppression. This has been studied in adults and shown to be increased in obese children with obstructive sleep apnea compared to age- and weight-matched controls without obstructive sleep apnea. The same group of researchers showed decreased metabolism in these tongue tissues [28, 29]. This decrease in metabolism is probably related to increased visceral fat that is associated with metabolic syndrome and a prediabetic state. In general, pediatric patients are at risk of recurrent obstructive sleep apnea if they are obese.

Summary

Cine MR anesthesia-induced sleep studies are a useful adjunct to clinical assessment when evaluating children with persistent obstructive sleep apnea despite first-line treatment with adenotonsillectomy. In children who are intolerant of CPAP and entertaining surgical options, cine MR studies are especially helpful in identifying anatomical and physiological causes of airway collapse that can be addressed surgically.

Acknowledgments This paper was supported by National Institutes of Health (NIH) grant RO1HL105206-01. The authors have indicated no financial conflicts of interest.

The project described was supported by the National Center for Advancing Translational Sciences of the NIH, under award number 5UL1TR001425-03. The content is solely the responsibility of the authors and does not necessarily represent the official views of the NIH.

Compliance with ethical standards

Conflicts of interest None

References

- 1. Donnelly LF (2005) Obstructive sleep apnea in pediatric patients: evaluation with cine MR sleep studies. Radiology 236:768–778
- Marcus C, Brooks L, Ward S et al (2012) Diagnosis and management of childhood obstructive sleep apnea syndrome. Pediatrics 130:E714–E755
- 3. Berry R, Brooks R, Gamaldo C et al (2017) AASM scoring manual updates for 2017 (version 2.4). J Clin Sleep Med 13:665–666
- Berry RB, Brooks R, Gamaldo DE et al (2017) The AASM manual for the scoring of sleep and associated events: rules, terminology and technical specifications. Version 2.4. American Academy of Sleep Medicine, Darien
- Berry R, Budhiraja R, Gottlieb D et al (2012) Rules for scoring respiratory events in sleep: update of the 2007 AASM manual for the scoring of sleep and associated events. J Clin Sleep Med 8:597– 619
- Bhattacharjee R, Kheirandish-Gozal L, Spruyt K et al (2010) Adenotonsillectomy outcomes in treatment of obstructive sleep apnea in children a multicenter retrospective study. Am J Respir Crit Care Med 182:676–683
- Shott SR, Donnelly LF (2004) Cine magnetic resonance imaging: evaluation of persistent airway obstruction after tonsil and adenoidectomy in children with down syndrome. Laryngoscope 114:1724–1729
- Mahmoud M, Gunter J, Donnelly LF et al (2009) A comparison of dexmedetomidine with propofol for magnetic resonance imaging sleep studies in children. Anesth Analg 109:745–753
- Arens R, McDonough J, Corbin A et al (2003) Upper airway size analysis by magnetic resonance imaging of children with obstructive sleep apnea syndrome. Am J Respir Crit Care Med 167:65–70
- Chatterjee D, Friedman N, Shott S et al (2014) Anesthetic dilemmas for dynamic evaluation of the pediatric upper airway. Semin Cardiothorac Vasc Anesth 18:371–378
- Donnelly LF, Strife JL, Myer CM (2001) Is sedation safe during dynamic sleep fluoroscopy of children with obstructive sleep apnea? AJR Am J Roentgenol 177:1031–1034
- Mahmoud M, Gunter J, Sadhasivam S (2009) Cine MRI airway studies in children with sleep apnea: optimal images and anesthetic challenges. Pediatr Radiol 39:1034–1037
- Fleck RJ, Amin RS, Shott SR et al (2014) MRI sleep studies: use of positive airway pressure support in patients with severe obstructive sleep apnea. Int J Pediatr Otorhinolaryngol 78:1163–1166
- Mahmoud M, Radhakrishman R, Gunter J et al (2010) Effect of increasing depth of dexmedetomidine anesthesia on upper airway morphology in children. Paediatr Anaesth 20:506–515
- Luscri N, Tobias J (2006) Monitored anesthesia care with a combination of ketamine and dexmedetomidine during magnetic resonance imaging in three children with trisomy 21 and obstructive sleep apnea. Pediatr Anesth 16:782–786
- Kandil A, Subramanyam R, Hossain M et al (2016) Comparison of the combination of dexmedetomidine and ketamine to propofol or propofol/sevoflurane for drug-induced sleep endoscopy in children. Pediatr Anesth 26:742–751
- Fleck RJ, Mahmoud M, McConnell K et al (2013) An adverse effect of positive airway pressure on the upper airway documented with magnetic resonance imaging. JAMA Otolaryngol Head Neck Surg 139:636–638
- Donnelly LF, Surdulescu V, Chini BA et al (2003) Upper airway motion depicted at cine MR imaging performed during sleep: comparison between young patients with and those without obstructive sleep apnea. Radiology 227:239–245
- Chen W, Gillett E, Khoo M et al (2017) Real-time multislice MRI during continuous positive airway pressure reveals upper airway response to pressure change. J Magn Reson Imaging 46:1400–1408

- Donnelly LF, Strife JL, Myer CM (2000) Glossoptosis (posterior displacement of the tongue) during sleep: a frequent cause of sleep apnea in pediatric patients referred for dynamic sleep fluoroscopy. AJR Am J Roentgenol 175:1557–1560
- Schwab RJ, Gefter WB, Hoffman EA et al (1993) Dynamic upper airway imaging during awake respiration in normal subjects and patients with sleep disordered breathing. Am Rev Respir Dis 148: 1385–1400
- 22. Bates A, Schuh A, Amine-Eddine G et al (2018) Assessing the relationship between movement and airflow in the upper airway using computational fluid dynamics with motion determined from magnetic resonance imaging. Clin Biomech. https://doi.org/10. 1016/j.clinbiomech.2017.10.011
- Guimaraes CV, Donnelly LF, Shott SR et al (2008) Relative rather than absolute macroglossia in patients with down syndrome: implications for treatment of obstructive sleep apnea. Pediatr Radiol 38: 1062–1067
- 24. Donnelly LF, Shott SR, LaRose CR et al (2004) Causes of persistent obstructive sleep apnea despite previous tonsillectomy and

adenoidectomy in children with down syndrome as depicted on static and dynamic cine MRI. AJR Am J Roentgenol 183:175-181

- Guimaraes CV, Kalra M, Donnelly LF et al (2008) The frequency of lingual tonsil enlargement in obese children. AJR Am J Roentgenol 190:973–975
- 26. Fricke BL, Donnelly LF, Shott SR et al (2006) Comparison of lingual tonsil size as depicted on MR imaging between children with obstructive sleep apnea despite previous tonsillectomy and adenoidectomy and normal controls. Pediatr Radiol 36:518–523
- Kang K, Koltai P, Lee C et al (2017) Lingual tonsillectomy for treatment of pediatric obstructive sleep apnea: a meta-analysis. JAMA Otolaryngol Head Neck Surg 143:561–568
- Kim C, Jackson N, Chawla S et al (2011) Quantification of tongue fat by novel MRI techniques and its relationship to BMI. Am J Respir Crit Care Med 183:A3678
- Kim A, Keenan B, Jackson N et al (2014) Metabolic activity of the tongue in obstructive sleep apnea: a novel application of FDG positron emission tomography imaging. Am J Respir Crit Care Med 189:1416–1425