Diagnosing Tongue Base Obstruction in Infants with Pierre Robin Sequence: Is Sleep Endoscopy Superior to Awake Endoscopy?

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Abstract

Objectives
To investigate whether awake endoscopy can diagnose base-of-tongue obstruction as reliably as sleep endoscopy in infants with Pierre Robin sequence (PRS).

Study Design
Individual case-control study.

Methods
A retrospective review of 141 infants with PRS managed between 2005 and 2015 was conducted. Thirty-five underwent both bedside awake flexible fiberoptic nasopharyngolaryngoscopy and drug-induced sleep endoscopy in the operating room. Base-of-tongue collapse was considered pathologically obstructed if described as moderate or severe or classified as Yellon grade 2 or 3. Sensitivity, specificity, and positive likelihood ratio of awake endoscopy findings were calculated with 95% confidence intervals (CI). McNemar’s test was also utilized to examine inter-test differences.

Results
Awake endoscopy was performed at a median age of 5.5 days (range 0 to 61), and drug-induced sleep endoscopy was performed at a median age of 21 days (range 0 to 216). Awake endoscopy had 50.0% sensitivity (CI 27.2-72.8%) and 86.7% specificity (CI 59.5-98.3%) for detecting base-of-tongue obstruction compared to drug-induced sleep endoscopy; false negative rate was 28.6% (N=10). Positive likelihood ratio was 3.75 (CI 0.96-14.65). Compared to awake endoscopy, sleep endoscopy demonstrated significantly more cases of base-of-tongue obstruction (p = 0.039).

Conclusion
Bedside awake endoscopy has low sensitivity for detecting base-of-tongue collapse in infants with Pierre Robin sequence. Due to the substantial number of false negatives, awake endoscopy may not be a reliable diagnostic modality for ruling out tongue base obstruction in this susceptible population. Drug-induced sleep endoscopy may be indicated in high-risk patients in order to avoid missing the diagnosis of upper airway obstruction.

Introduction
• Pierre Robin sequence (PRS): microretrognathia, glossoptosis, & possible cleft palate (Fig 1) → upper airway obstruction, feeding difficulty, & growth retardation.1
• Upper airway obstruction may be subtle, manifesting only during sleep. 85-100% of PRS infants have concurrent sleep-disordered breathing.2,3
• Awake flexible fiberoptic nasopharyngolaryngoscopy has been used to assess for sites of obstruction and to grade base-of-tongue (BOT) collapse in patients with PRS.4
• Drug-induced sleep endoscopy (DISE) in adults and children has been shown to identify more cases of obstruction than awake endoscopy (AE).5,6
• Use of DISE in infants with PRS to has not been described.
• Can awake endoscopy diagnose base-of-tongue obstruction as reliably as sleep endoscopy in infants with PRS?

Methods
• Retrospective case series with chart review of the 141 consecutive infants with PRS managed between January 1, 2005 and July 31, 2015.
• 35 patients underwent both AE and DISE.
• Awake flexible fiberoptic nasopharyngolaryngoscopy was performed and interpreted by pediatric otolaryngologists at the bedside.
• DISE was performed and interpreted by pediatric otolaryngologists in the operating room.
• Sedation was achieved using continuous intravenous propofol infusion.
• BOT collapse was graded as normal, mild, moderate, or severe or 0, 1, 2, or 3 according to the Yellon classification7.
• Moderate (2) or severe (3) BOT collapse was considered pathologically obstructed (Fig 2).
• Sensitivity, specificity, positive predictive value, negative predictive value, positive likelihood ratio, and negative likelihood ratio of AE findings compared to the gold standard of DISE were calculated with 95% confidence intervals (CI).
• McNemar’s test was utilized to examine inter-test differences between AE and DISE.

Results
• Awake endoscopy performed at a median age of 5.5 days (range 0 to 61 days).
• DISE performed at a median age of 21 days (range 0 to 216 days).

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Table 1. Prevalence of BOT collapse as determined by AE and DISE in all 35 patients.

• AE had 50.0% sensitivity and 86.7% specificity for detecting BOT obstruction compared to DISE (Tables 1 & 2).
• False negative rate = 28.6% (N=10).

Discussion & Conclusions
• Bedside awake endoscopy has low sensitivity for detecting BOT obstruction in infants with PRS compared to drug-induced sleep endoscopy.
• There is a substantial false negative rate with awake endoscopy.
• Awake endoscopy may not reliably rule out BOT obstruction in this susceptible population.
• Presumably, the upper airway is more patent during an awake exam due to the increased muscular tone of a crying infant.5
• Drug-induced sleep endoscopy may be indicated as an adjunct in high-risk infants with PRS in order to avoid missing the diagnosis of upper airway obstruction.

References