



Inter-observer reliability of localization of recorded stridor sounds in children

Joost W. Zwartenkot^{a,b}, Hans L.J. Hoeve^a, Johannes Borgstein^{a,*}

^aDepartment of Pediatric Otorhinolaryngology, Erasmus Medical Centre – Sophia Children's Hospital, Dr. Molewaterplein 60 (Sp1417) 3015 GJ Rotterdam, The Netherlands

^bDepartment of Otorhinolaryngology and Head & Neck Surgery, Radboud University Nijmegen Medical Centre, Nijmegen, The Netherlands

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ABSTRACT

Objective: To determine the inter-observer reliability in localization of recorded stridor sounds in children.

Method: The stridor sounds of 28 children programmed for laryngobronchoscopy were recorded with a high quality digital recorder. Nineteen of these recordings with a diagnosis confirmed by endoscopy, were presented to otorhinolaryngology residents, academic specialists and non-academic consultants ($n = 38$) in different situations with and without additional information about the subject. The participants were requested to score the sounds as pharyngeal, supraglottic, glottic, subglottic or tracheal in both situations. The scores were analyzed per group of participants, per location of obstruction and per diagnosis in the different situations.

Results: The performance of the total group was just above chance level with an average score of 29.6%. The total results improved slightly with the additional information present, although not significantly. No significant difference was found between the three categories of participants. The supraglottic sounds were significantly better differentiated from the other locations in both assessment types ($p < 0.001$). The tracheal region ($p < 0.01$) and the supraglottic region ($p < 0.05$) received significantly higher scores when the additional information was present. Laryngomalacia was significantly better differentiated ($p < 0.001$) from the other diagnoses. The improvement in assessment with and without additional information present was significant for laryngomalacia ($p = 0.002$) and tracheomalacia ($p < 0.035$).

Conclusion: The clinical observation of stridor in children shows poor levels of localization. Even though the two most common diagnoses, laryngomalacia and tracheomalacia are localized more correctly, the general performance is not significantly higher than random. No evidence was found in this study for differences in results by the observer's level of experience. We would recommend that an observational policy is only adequate when clear clinical signs are present that indicate laryngomalacia as first differential diagnosis. In other cases rigid and flexible endoscopy is indicated to locate the site and nature of obstruction.

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1. Introduction

Stridor is an important symptom of airway obstruction in children. The Latin word “stridulus” means harsh creaking, whistling or grating [1]. The stridorous sound is caused by turbulence of air beyond the location of the constriction and by vibration of the surrounding tissue [2,3]. Due to the relative narrower airway, stridor is a common and alarming finding in children. Because of the narrow airway, a small constriction can have great consequences for the child's ability to breathe. One millimeter of edema in the glottic mucosa of small children will

reduce the surface area by 65% and even up to 70% in the subglottic surface [4,5].

The causes of stridor can be subdivided by location in the child's airway at the level of the pharynx, larynx, trachea and the bronchia. The most frequent causes of obstruction are congenital diseases, neoplasms, post-intubation injury, neurological and inflammatory diseases [1,6]. In case of acute stridor croup (laryngotracheitis) is the most common cause. Chronic stridor in newborns is mostly caused by laryngomalacia, a combination of anatomical anomalies in the cartilage support in the larynx. It is associated with a long curled epiglottis, short aryepiglottic fold (AEF) and redundant arytenoid mucosa. The precise aetiology is unknown [4,7]. In a large retrospective study presented by Zoumalan et al. in 2007, 77% of the obstructions in stridor were caused by congenital laryngeal diseases, of which 94% proved to be laryngomalacia [5,7].

* Corresponding author. Tel.: +31 10 703 7024; fax: +31 10 703 6472.

E-mail addresses: j.borgstein@erasmusmc.nl, borgstein@gmail.com (J. Borgstein).

Important information is provided by clinical history and physical examination of the stridorous child. The circumstances in which the stridor takes place, improves or progresses provide indications concerning the localization of the obstruction. The stridor can change in a certain body position, during feeding or during sleep. The medical history can provide additional information concerning the chronic character or congenital cause [2,4,8,9].

At the physical examination the child is particularly inspected and auscultated. One should pay attention to retractions (intercostal, subcostal, subjugular, subclavicular or sternal), cyanosis, nasal flaring and the child's preferred body position. Possibly the child suffers from dysphagia or drooling. Also the body is assessed for dermal haemangioma and possible syndromal deformities [3,4,8,10].

It is very important to assess the stridor itself. The stridorous phase, inspiratory, expiratory or biphasic, provides an indication whether the obstruction is localized intra- or extrathoracically. This distinction is most relevant and biphasic stridor can be considered a red flag symptom. Indications for the subdivision in airway location are provided by the frequency itself, either high or low pitched, and by the character of the stridor, either voiced (a pure tone and overtones) or fricative (a noise-like sound). A typical laryngeal stridor is inspiratory, high pitched and voiced, where a pharyngeal obstruction causes a bidirectional, low pitched and fricative sound [4,10,11].

The assessment of the stridorous sound brings difficulties for the diagnostic evaluation of a child with stridor. As described by Papsin et al. in 1999 it is hard to assess the stridor severity by judgment of the stridor in isolation. In their research 38 consultants and 22 trainees participated in an acoustical assessment to score the expected severity of the obstruction by judgment of the stridorous sound. The results showed that the participants did not score any higher than the level of chance. Although their performance increased significantly when the clinical summary of the patients was presented, the results did not differ from the level of chance. No significant difference in performance by the consultants and trainees was found, but the authors suggested that the trend of the results showed that the experience of the participant plays a critical role in the determination of severity of stridor [12].

For this research we were interested whether the results of Papsin et al. would be different when the localization of the obstruction was subject of the assessment rather than the severity of the stridor itself. In the diagnostic process the localization of obstruction can be considered of even importance as the severity of the stridor. We were also interested in the importance of the acoustic evaluation of the stridor in the physical examination of the child.

2. Methods

2.1. Collection of acoustical data

Using a digital wav/mp3-recorder (Edirol R-09HR, Roland Systems Group, Bellingham, WA, USA) we recorded the stridorous sounds of 28 children with audible breathing sounds admitted to or presenting at the outpatients of our ENT clinic in the Sophia children's hospital in Rotterdam, a tertiary referral center for the southern part of the Netherlands.

The subject's medical history, the varying conditions in which the stridor occurred (feeding, body position, wakefulness) and clinical signs like dyspnea, cyanosis and retractions (subcostal, tracheal or intercostal) were noted. We qualified the stridor sound as high or low pitched and voiced (pure sound) or fricative (noise-like sound).

Several breathing cycles were recorded for each child either in a silent audiometry room in the outpatient clinic or in the operating theatre before laryngobronchoscopy. Audio files were created using a high quality digital recording device with build-in microphone (Edirol R-09HR) at 24 bit sample size and 96 kHz sampling rate frequency wave-recording settings. The frequency response for the internal microphone was 20 Hz to 40 kHz. Recordings were made in supine or sitting position and the device held at a distance of between 5 and 10 cm from the subject's mouth. No amplification or filters were applied to the recordings. Nineteen of the stridorous sounds with various diagnoses were carefully selected for the assessment.

2.2. Determination of diagnosis

Only those children with confirmed endoscopic diagnosis were included in the study. The children with an unknown cause of the stridor underwent laryngobronchoscopy (LBS) by flexible and rigid scope under general anesthesia. For the children with a previously known diagnosis, the LBS reports were reassessed and scored for location. The diagnosis and location of obstruction (pharyngeal, supraglottic, glottic, subglottic or tracheal) were noted. When the location of obstruction was unclear or endoscopic evidence of multiple obstructions was present, the patient was excluded from the analysis.

2.3. Study group

The recorded stridorous sounds we selected for the assessment were recorded from 11 male (58%) and 8 female (42%) subjects. Their mean age was 8.5 ± 11 months old (range 0.1–37.8 months). Five of the children had no previous medical history and 4 had been prematures. In 10 children the stridor was present from birth. Seven children had developed a stridor acutely and in 2 children the development was more gradual. Eight children had a history of intubation.

The diagnostic locations of obstruction were 2 pharyngeal, 6 supraglottic, 3 glottic, 4 subglottic and 4 tracheal. An overview of the descriptions, clinical data and diagnosis of the study group can be found in Table 1.

2.4. Presentation of the audio files to the participants

Blank presentations of the stridorous sound without any additional information were individually assessed by 10 residents and 7 specialists of our clinic. The recordings were presented to the participants by a standard computer with a digital media player using desktop speakers. The participants were able to listen repeatedly to the audio files if they required. They were requested to score the presented stridorous sounds as pharyngeal, supraglottic, glottic, subglottic or tracheal. Only one correct answer was accepted. In case of double scoring the answer was considered incorrect.

At a subsequent regional meeting the audio files were presented again, this time with additional information about the subjects' age, medical history, conditions in which the stridor occurred and presenting symptoms. Fifteen trainees of our clinic, of whom 9 had participated in the blank assessment, participated in this assessment, as well as 8 members of staff of our clinic, of whom 6 had participated in the blank assessment. Also 13 regional non-academic otolaryngology-consultants participated in this second assessment.

The audio files were presented by the speaker system of a conference auditorium and were repeated once. The additional information was read to the participants before the audio files were presented, as well as handed out as written information (see

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