OBSTRUCTION OF THE PEDIATRIC AIRWAY

diagnostic and therapeutic aspects

Cover: Rock formation in Mpatamanga Gorge, Malawi.

OBSTRUCTION OF THE PEDIATRIC AIRWAY
diagnostic and therapeutic aspects

LUCHTWEGOBSTRUCTIE BIJ KINDEREN
diagnostische en therapeutische aspecten

PROEFSCHRIFT
ter verkrijging van de graad van Doctor
aan de Erasmus Universiteit Rotterdam
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Prof. Dr. P.W.C. Akkermans M.A.
en volgens besluit van het College voor Promoties.
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door

Lambertus Johan Hoeve

geboren te Leeuwarden

PROMOTIECOMMISSIE:

PROMOTOR:	Prof. Dr. C.D.A. Verwoerd
OVERIGE LEDEN:	Prof. Dr. J.C. Molenaar
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This study is part of the project Airway Stenosis
Supervisor: Dr. H.L. Verwoerd-Verhoef
Institute for Otorhinolaryngology
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Voor Elly, aan wie ik meer heb te danken dan dit proefschrift.

Aan mijn ouders.

VOORWOORD

Dit proefschrift is ontstaan onder leiding van Professor Dr. C.D.A. Verwoerd. Hij heeft, door zijn enthousiasme voor het klinische onderwerp en zijn belangstelling voor de jonge patiënten, mij weten te motiveren het onderzoek te beginnen, het voort te zetten, en het af te ronden. Hij suggereerde onderwerpen voor onderzoek, gaf mij de ruimte en de tijd om eraan te werken, en bestudeerde met zichtbaar genoegen de resultaten ervan. Tijdens vele besprekingen veranderde elk manuscript geleidelijk van een ruwe versie in een afgerond artikel. Ik ben hem zeer erkentelijk dat hij het mij mogelijk heeft gemaakt de opgedane klinische ervaring vorm te geven in dit proefschrift.

Met de onderwerpen die in dit proefschrift worden bestudeerd, de laryngobronchoscopie en de behandeling van stenosen in de luchtweg, zijn de namen van twee mensen verbonden: Cees Bos, die in de zestiger jaren de starre open bronchoscopie, en in de tachtiger jaren de flexibele bronchoscopie in het Sophia Kinderziekenhuis introduceerde, en Rob Berkovits die de behandeling van luchtwegstenosen begon, een behandeling die niet mogelijk is zonder bronchoscopie. Cees Bos bracht mij de kunst van de bronchoscopie bij, terwijl Rob Berkovits mij onderwees in de silicongedachte, de intubatiologie, en de operatietechnieken. Deze diagnostische en therapeutische technieken zijn de fundamenten waarop het onderzoek is gebouwd.

Mevrouw Dr. H.L. Verwoerd-Verhoef heeft een grote bijdrage geleverd aan mijn begrip van het proces van wondgenezing in de larynx, door in een serie dierexperimentele onderzoekingen de morfologische veranderingen van het cricoïd ten gevolge van letsel en groei te beschrijven.

De belangrijkste gegevens van de bronchoscopieën en van de stenosebehandelingen werden verzameld en opgeslagen in gegevensbestanden, ontworpen in samenwerking met Bert van Zanten en Gijs Conijn. Bij het verzamelen, invoeren en bewerken van deze gegevens heb ik onontbeerlijke hulp gehad van Jan Rombout en Orhan Eskici. De statistische bewerking werd uitgevoerd door de heer P.G.H. Mulder van de afdeling Biostatistiek van de Erasmus Universiteit Rotterdam.

De engelse teksten werden bewerkt door Alice Ribbing en Ko Hagoort, de illustraties zijn van de hand van Joop van Dijk werkzaam bij het Audiovisuele Centrum AZR, de vormgeving van het proefschrift is voor een belangrijk deel het werk van Michael Brocaar.

Het totstandkomen van dit proefschrift was ondenkbaar zonder de bijdrage van alle genoemde mensen, die ik op deze wijze mijn grote dankbaarheid betuig.
Verder ben ik dank verschuldigd aan de collega's die, vóór mij en later met mij, bronchoscopieën deden en stenosen behandelden, en meestal zonder het te weten, bouwstenen leverden voor dit proefschrift: Berkovits, Bos, van der Pot, van der Schans, Conijn en van Loosen. Ook de kinderartsen van der Voort, Hazelzet, Joosten, Tibboel en Sauer, en de verpleegkundigen van de drie Intensive Care afdelingen in het SKZ hebben een belangrijke rol gespeeld, omdat succesvolle behandeling van stenosepatiënten alleen mogelijk is wanneer de medische en verpleegkundige zorg van een hoog niveau is. Bronchoscopie kan alleen veilig gebeuren in samenwerking met daarin ervaren anesthesisten en operatie-assistenten. Dat dit invasieve onderzoek zo weinig complicaties heeft gekend, is mede te danken aan hun vakmanschap.
Een groot aantal mensen was en is verantwoordelijk voor een zorgvuldige administratie van de medische gegevens: secretaresses, polikliniekassistentes, het medisch archief, de medische registratie, en de CDAI. Ook hen wil ik graag laten delen in de waardering die ik heb voor de organisatie die deze vorm van patiëntenzorg de ruimte gaf.
De collega's KNO-artsen van het Academisch Ziekenhuis Rotterdam maakten het mij mogelijk veel tijd te besteden aan het onderzoek en het schrijfwerk, waarbij ik in het bijzonder Edwin Eichhorn, John van Loosen, Gijs Conijn, en Anne van Linge wil noemen.
Dankzij het organisatietalent en het secretariële overzicht van Jaqueline Aukes zijn de laatste maanden van de voltooiing van het proefschrift bijna rimpelloos verlopen.

CONTENTS

CHAPTER 1

INTRODUCTION

Pediatric Laryngology in a referral hospital for children such as the Sophia Children's Hospital is to a large extent concerned with patients suffering from airway obstruction, most of whom, owing to the nature of the disorder, are treated in an Intensive Care Unit (ICU). The otorhinolaryngologist, in close cooperation with ICU-pediatricians, is confronted with a great variety of airway pathology: intubation or tracheotomy related problems, post-intubation injury, cicatricial laryngeal stenosis, congenital laryngeal stenosis, subglottic hemangioma, choanal atresia, pharyngeal obstruction, epiglottitis, subglottic laryngitis, tracheomalacia, etc.

Laryngobronchoscopy (LBS) is the most important tool in the diagnosis of such disorders in children. Both rigid and flexible techniques have to be used. Rigid open LBS was introduced in the Sophia Children's Hospital in the 1960s, and flexible LBS in the early 1980s. The technique of rigid open bronchoscopy has been well-established for many years. Excellent equipment is available nowadays, also for very small infants. Disadvantages of the rigid technique are distortion of the laryngeal entrance, necessity of general anesthesia and use of muscle relaxants, which hamper the examination of functional larynx disorders such as laryngomalacia and vocal cord paralysis. Flexible LBS is for that reason indispensable, but not always easy to perform in an uncooperative infant [47,79,108]. In Chapter 2 a method is described to perform flexible LBS under general anesthesia without the use of muscle relaxants, which allows for the larynx to be inspected under optimal physiological circumstances.

Apart from our own department of Otorhinolaryngology, the indications for LBS came from the departments of Intensive Care, Pulmonology, and Pediatrics in the Sophia Children's Hospital, and from other hospitals. The number of LBSs has increased ever since its introduction, now reaching more than 200 annually. To evaluate the indications, diagnoses, and complications of these large numbers of LBSs a data base was created. This data base, its structure, and its contents are discussed in Chapter 3.

The data base proved very useful as an easy access to aspects of LBS, e.g. the circumstances under which complications occurred, which is discussed in Chap-

ter 4. Complications were associated with pre- and post-operative diagnosis, procedures performed during LBS, and other factors present during the LBS. The results were analyzed to detect possible statistically significant relationships. The types of complications and the complication rates were compared with those found in other studies [44,47,48,52,69,74,79,84,85,108].

An important disorder in respect to LBS, both diagnostically and therapeutically, is aspiration of a foreign body. The role of LBS and other diagnostic tools herein is discussed in Chapter 5. The signs and symptoms with which the patients presented, and the results of the preoperative radiographic investigations were studied to assess their diagnostic value [20,24,92,96,98]. In addition a study was made of the elapsed time between aspiration and extraction of the foreign body in two patient groups with a different primary diagnosis [71,86,97].

Preterm infants in a Neonatal Intensive Care Unit are often treated with artificial ventilation and prolonged endotracheal intubation. The presence of a ventilating tube in the larynx frequently results in edema, superficial lesions, ulceration, or granulations in the laryngeal lumen [36,37,43,95]. Symptoms of airway obstruction may occur after artificial ventilation is discontinued and the patient is extubated. This problem of failed extubation may be addressed in various ways: by reintubation with or without endoscopic treatment, or operatively [15,29,32,33]. The results of reintubation as a treatment of difficult extubation in preterm infants is presented in Chapter 6.

Acquired laryngeal or tracheal cicatricial stenosis is reported to occur in 1-10% of the children treated with prolonged intubation [36,37,43,95]. The therapy of severe cicatricial stenosis generally consists of laryngeal surgery followed by intubation or stenting. Many variations of this treatment have been reported [1,23,25,27,38,39, 41,45,46,53,57,72,77,78,87,89,102,104,107,109].

In the Sophia Children's Hospital most children with such a stenosis have been treated with a laryngofissure and a posterior cricoid division (Réthi), followed by stenting with a silicone rubber stent. Laryngofissure without a posterior cricoid split and the castellated anterior incision described by Evans were also performed [41,87]. The details and results of surgical treatment in these children in comparison with other reports are presented in Chapter 7 [8,27,59,70,76].

The factors that contribute to post-intubation injury, and the subsequent process of healing are not yet fully understood. Histologic study of the process of wound

repair, also in relation to growth of the larynx in young rabbits, is the subject of an experimental research program at the Department of Otorhinolaryngology of the Erasmus University Rotterdam. A series of animal experiments revealed that the formation of subglottic stenosis is largely determined by the depth of the laryngeal injury and the age at the time of trauma. A critical factor appears to be the involvement of the perichondrium and cartilage of the cricoid ring. The injured cartilage appears to produce large amounts of cells contributing to the stenosis. Just this type of wound healing of the cricoid ring shows apparent changes with increasing age [2,3,4,5,6,10,11,12,13,14,99,100]. Specimens of the experimentally damaged subglottic area of growing rabbits were compared with biopsies taken from the stenotic area of infants with acquired laryngeal stenosis. The histologic results of this comparative study are presented in Chapter 8.
The final chapter comprises a general discussion.

CHAPTER 2

FIBEROPTIC LARYNGOSCOPY UNDER GENERAL ANESTHESIA IN NEONATES

ABSTRACT

In the Sophia Children's Hospital we perform fiberoptic laryngoscopy in neonates under general anesthesia without the use of muscle relaxants in the diagnostics of functional laryngeal disorders. The necessary diagnostic and anesthetic equipment is described. Special attention is paid to the way in which the fiberscope is introduced into the breathing circuit and into the child's airway. Several advantages over other methods are discussed. The procedure has proved to be easy, safe and informative.

INTRODUCTION

In the Sophia Children's Hospital 150 to 200 laryngobronchoscopies are performed annually for various diagnostic and therapeutic reasons. In all therapeutic and in many diagnostic procedures we prefer to use the rigid bronchoscope with a telescope. The patient is given general anesthesia and is ventilated. The procedure always involves administration of a muscle relaxant.

However, in children presenting with an inspiratory stridor it is necessary to evaluate the function of the larynx. This is not possible if muscle relaxants have been given, so in these cases we have to use a different method.

Direct laryngoscopy in a spontaneously breathing anesthetized child has the disadvantage of distortion of the larynx. Inserting the tip of the laryngoscope blade in the vallecula causes traction of the epiglottis, and consequently impedes examination of the mobility of laryngeal structures. Moreover, during this type of direct inspection inspired oxygen concentration, as well as the concentration of volatile anesthetics (and thereby anesthetic depth), are hard to control because of admixture of ambient air. This might limit the time available for the procedure, especially in the very young child.

Fiberoptic laryngoscopy under topical anesthesia in the awake child [90,91] often leads only to discomfort of patient, parent and physician, while milder abnormalities of the larynx might be masked by active adduction efforts. Some authors advise to perform the procedure under heavy sedation [80].

Since two years we have been performing fiberoptic laryngoscopy under general anesthesia without the use of muscle relaxants in children in whom a functional evaluation of the larynx is indicated. Experience with some 15 patients has shown us that this technique does not have the disadvantages found in direct laryngoscopy under general anesthesia or fiberoptic laryngoscopy in the unanesthetized patient.

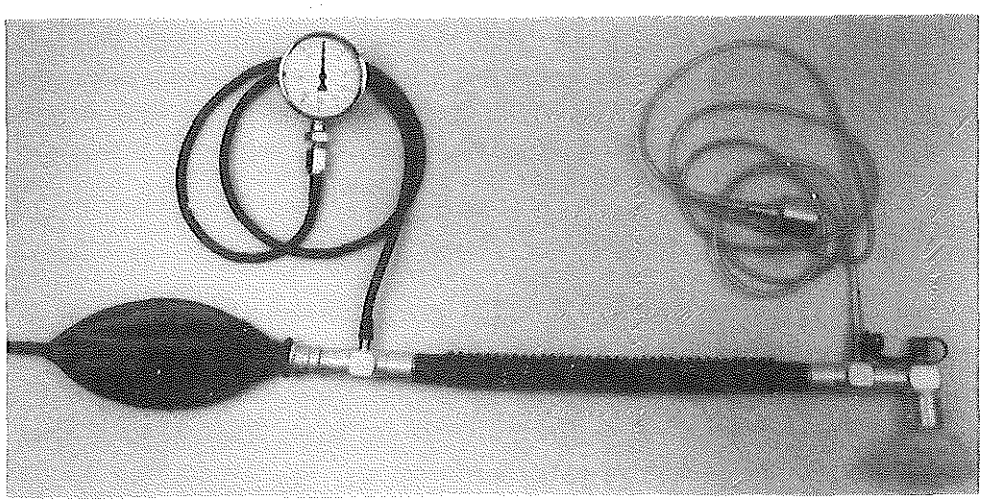

Figure 1: Jackson-Rees system with a manometer and the breathing-circuit elbow.

METHODS

The patient is anesthetized by breathing 60-70% nitrous oxide and halothane in oxygen. When the depth of the anesthesia is sufficient to tolerate the procedure, the mask is taken from the face, and the fiberscope (Olympus BF type 3C10, diameter 3.5 mm.) is introduced through a custom-made breathing-circuit elbow. Figure 1 shows the necessary anesthetic equipment. An air-tight seal is obtained as the fiberscope is passed into the elbowpiece through a perforated rubber sealing cap (Fig. 2).

The tip of the fiberscope is placed in the nose, and the mask is applied to the face again. The fiberscope is then passed through the nose and pharynx, until an excellent view of the larynx is obtained (Fig. 3), with the patient breathing spontaneously.

The instrument is never introduced into the subglottic area or the trachea.

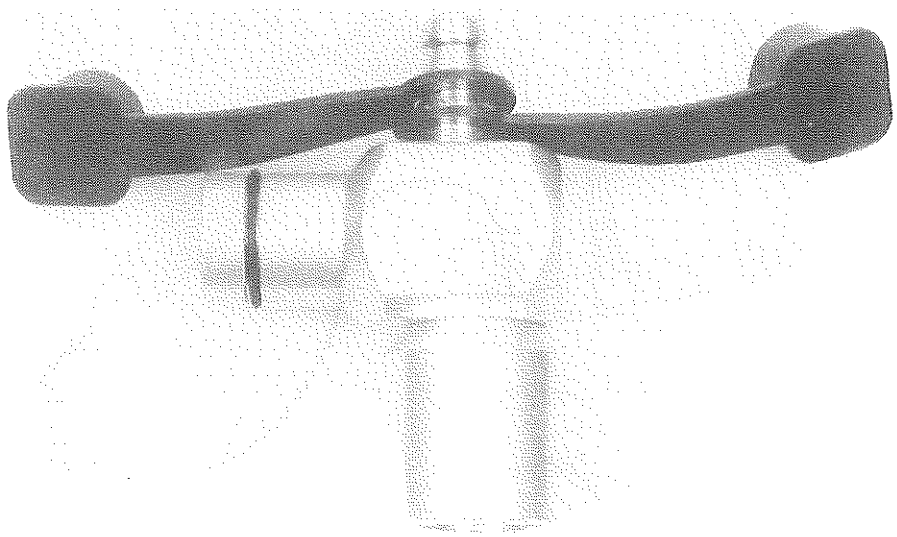

Figure 2: Breathing-circuit elbow with a perforated and an unperforated sealing cap.

DISCUSSION

We think that our method has several advantages over both other methods mentioned in the introduction:

1. Because the larynx is not distorted, and the patient is breathing, the movement of the laryngeal entrance and the vocal cords can be observed. This is indispensable in for example the diagnosis of laryngomalacia and vocal cord paralysis.
2. Photography and video film registration are possible and a second observer can join the examination with the use of the teaching head or the video screen.
3. During the whole procedure inspired concentrations of oxygen and anesthetic gasses can be controlled, since there is no major leak or admixture of ambient air. Therefore the procedure can take longer, and will be more precise.
4. The depth of anesthesia can be adjusted by the anesthetist at any time during the procedure. The influence of the application of various levels of continuous positive airway pressure (CPAP) on the functional abnormality can be studied at the same time.

Passing the fiberscope through the vocal cords to examine the subglottic space and the trachea has to be condemned on account of laryngospasm (except when topical

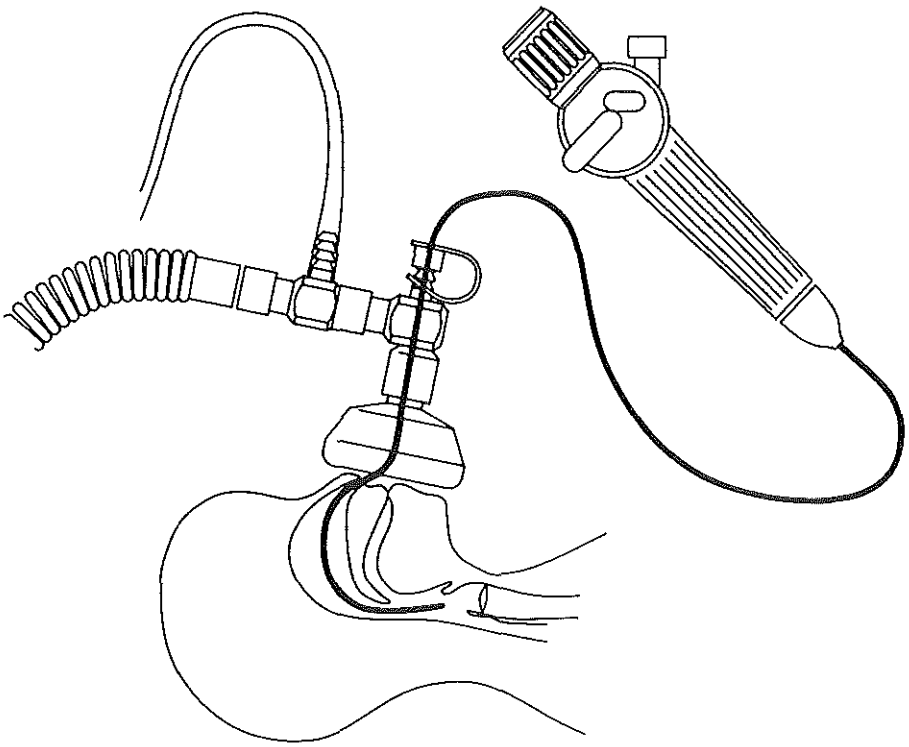

Figure 3: Schematic representation of the anesthetic equipment and the fiberscope introduced into the laryngeal entrance.

anesthesia is used) and obstruction of the airway, especially in the neonate. To examine these structures a rigid bronchoscope is introduced after a muscle relaxant has been given and the child is ventilated by hand.

In conclusion, anatomical disorders of the larynx, trachea and bronchial tree can be diagnosed perfectly well with the rigid bronchoscope. Fiberoptic laryngoscopy however is very useful in demonstrating functional disorders of the larynx. The procedure described has proved to be very informative, safe, and easy.

CHAPTER 3

PEDIATRIC LARYNGOBRONCHOSCOPY: 1332 PROCEDURES STORED IN A DATA BASE

ABSTRACT

Laryngobronchoscopy (LBS), using both rigid and flexible bronchoscopes, has become a frequently performed operation in children. A data base was established to enable retrospective evaluation of a large number of LBSs carried out in a pediatric center. Experience with 1332 cases of LBS involving 808 patients over an eight year period is presented. The main indications for LBS were inspiratory stridor, atelectasis, and suspected foreign body aspiration. Most frequent diagnoses at LBS were bronchopneumonia, intubation trauma, tracheomalacia, laryngomalacia, and foreign body aspiration. Only 25 complications occurred (1.9%) including two cases of xylometazoline intoxication.

INTRODUCTION

The increased survival rate of prematurely born infants due to the advancement of neonatal intensive care, has increased the incidence of both congenital and acquired respiratory anomalies. Laryngobronchoscopy (LBS) is an important tool in the diagnosis and treatment of disorders of the respiratory tract in children. Diagnostically, the combined use of flexible and rigid bronchoscopes renders this procedure superior to even the most modern radiographic examination, such as CT-scan and Magnetic Resonance Imaging (MRI). The improvement of optical instruments used with rigid bronchoscopes has increased the therapeutic potential. Finally, the advance of modern pediatric anesthesiology has made LBS a safe and frequently used procedure in children.

Between 150 and 200 LBSs are performed annually in a Dutch referral center for children with airway disorders. To enable evaluation of this procedure, LBS data for a period of eight years were stored in a data base. Subsequently, the indications, diagnoses, and complications were analyzed statistically.

PATIENTS/MATERIALS AND METHODS

From 1982 to 1990, a total of 1332 LBSs were performed at the department of Otorhinolaryngology of Sophia Children's Hospital in Rotterdam, The Netherlands. The operation was generally carried out by one of a group of four ENT specialists

Table I: Code table for laryngobronchoscopy: indications, diagnoses, procedures and complications.

INDICATIONS AND DIAGNOSES

ORAL CAVITY AND PHARYNX 00
01 acquired pharyngeal stenosis
02 dysphagia
03 micro-/retrognathia
04 congenital pharyngeal stenosis
05 vallecular cyst
06 lymphangioma
09 other oral cavity, pharynx

LARYNX 10
11 epiglottitis
12 laryngitis
13 subglottic laryngitis
14 edema/fibrin larynx
15 granulations/ulcerations larynx
16 acquired laryngeal stenosis
17 intubation trauma
18 foreign body larynx
20 laryngomalacia
21 vocal cord paralysis
22 congenital laryngeal stenosis
23 congenital subglottic stenosis
24 papillomatosis
25 hemangioma
26 laryngofissure postoperatively
19 other larynx

TRACHEA 30
31 tracheitis
32 granulations trachea
33 acquired stenosis trachea (web)
34 foreign body trachea
35 tracheomalacia
36 congenital stenosis trachea
37 external compression trachea
38 tracheal fistula/sinus
39 other trachea
07 tracheostomy

BRONCHIAL TREE 40
41 bronchopneumonia/infiltrate
42 atelectasis
43 mucous plug
44 granulations bronchus
45 stenosis bronchus
46 hemoptysis
47 foreign body bronchi
48 bronchomalacia
50 hypoplastic lung, bronchial tree
51 bronchopulmonary dysplasia
49 other bronchi

60 no abnormalities

OTHER INDICATIONS
61 inspiratory stridor
62 expiratory/biphasic stridor
63 dyspnea/ventilation problems
64 apnea, sleep apnea
65 aspiration/reflux
66 hoarseness
67 cough
68 tuberculosis
69 other indications (not localized)

PROCEDURES

70 no procedures
71 in-/excision cyst
72 granulations/polyps extraction
73 steroids injection
74 dilation stenosis
75 laryngeal stent insertion or removal
76 (re)intubation
77 (re)cannulation
78 decannulation
80 tracheotomy
81 laser surgery
82 foreign body extraction
83 bronchial lavage
84 bronchoalveolar lavage (BAL)
85 bacterial sampling
86 xylometazoline application
87 biopsy
79 other procedures

COMPLICATIONS

90 no complications
88 inspiratory stridor
89 stridor and intubation
91 hemorrhage
92 bradycardia
93 laryngospasm
94 bronchospasm
95 pneumothorax
96 anesthesia
97 intoxication
98 death
99 other complications

using rigid or flexible bronchoscopes. Rigid laryngobronchoscopy was performed under general anesthesia using a rigid, pediatric, Storz bronchoscope with a diameter ranging from 2.5 to 6 mm and a Hopkins telescope. Alternatively, a flexible, fiberoptic pediatric bronchoscope with a diameter of 3.6 mm (Olympus BF type 3C20) was used to assess laryngeal dynamics in a spontaneously breathing patient under local or general anesthesia without the use of muscle relaxants [60].
Pertinent data concerning these 1332 LBSs were collected, stored in a data base (S1032), and then analyzed statistically using SPSS-X. These data consisted of name, date of birth, and sex of patient; primary and secondary indication for laryngobronchoscopy; date of performance; type of technique; diameter of bronchoscope; name of laryngoscopist; primary and subsequent diagnoses; procedures during laryngobronchoscopy; any complications. Table I gives the code used for indications, diagnoses, procedures, and complications.

Number of LBSs per patient	Number of patients
1	603
2	108
3	38
4	20
5	14
6	8
7	1
8	4
9	2
10	3
13	2
17	1
18	2
22	1
28	1
Total	808

Table II: Number of laryngobronchoscopies per patient.

RESULTS

The 1332 LBSs were performed in 808 children, 486 boys and 322 girls. Table II gives the number per patient. The average was 1.6 LBS per patient with 25 undergoing more than five procedures, while one patient required LBS 28 times.

The age at LBS ranged from one day to 18 years, with a mean of 2.0 years. Figure 1 represents the age distribution.

The rigid technique was used in 1250 cases, the flexible technique in 19, and a combination of both techniques in 54. In the remaining 9 cases the technique was not recorded.

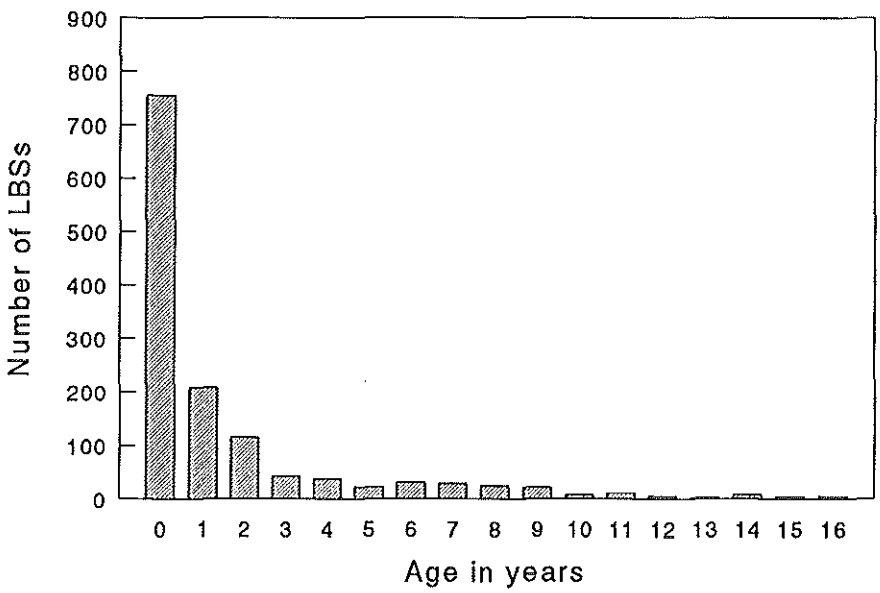

Figure 1: Number of laryngobronchoscopies per age group.

Table III lists the indications for LBS, the most frequent being inspiratory stridor, atelectasis, bronchopneumonia, and foreign body aspiration. The findings at LBS are given in table IV, with the most frequent diagnosis being bronchopneumonia, followed by tracheomalacia and intubation trauma. No abnormality was found in 173 cases (13.0%). The indications and diagnoses lumped together under 'miscellaneous' in tables III and IV include hamartoma, laryngeal cleft, lymphangioma, and respiratory papillomatosis.

The findings in patients with the two most frequent indications for LBS are presented separately in tables Va and Vb. The most frequent cause of inspiratory stridor appeared to be laryngomalacia, followed by intubation trauma, subglottic laryngitis and laryngeal edema. Regarding atelectasis, LBS generally confirmed the anticipated diagnosis and also served as a therapeutic procedure.

Table III: Primary and concomittant indications for 1332 laryngobronchoscopies.

Indication	Primary	Concomittant
Inspiratory stridor	264	287
Atelectasis	205	221
Bronchopneumonia	144	166
Foreign body aspiration	90	100
Intubation trauma	67	75
Dyspnea, ventilation problems	49	64
Congenital tracheal stenosis	46	59
Aspiration, GE reflux	46	54
Subglottic hemangioma	43	49
Tracheomalacia	28	49
Laryngeal stent	27	31
Tracheal fistula/sinus	24	37
Expiratory stridor	22	24
Laryngomalacia	19	23
Tuberculosis	18	18
Epiglottitis	17	18
Decannulation	16	19
Apnea, sleep apnea	15	21
Intubation	14	17
Acquired laryngeal stenosis	11	18
Congenital laryngeal stenosis	10	11
Hemoptysis	10	11
Congenital subglottic stenosis	9	11
Cough	8	23
Subglottic laryngitis	8	13
Hoarseness	6	6
Miscellaneous	116	161
Total	1332	1586

Table VI lists the procedures that were carried out during LBS for therapeutic or other purposes. The most frequent procedure was bronchial lavage, followed by intubation, bacterial sampling, localized injection of steroids, foreign body extraction, and biopsy.

As shown in table VII, complications occurred in 25 cases (1.9%), the most serious being bradycardia or asystole, pneumothorax, and intoxication. Other complications were hemorrhage and inspiratory stridor. Regarding the two cases of intoxication, in one case it was confirmed and the other suspected that this was due to overdosage of xylometazoline hydrochloride, a sympathomimetic drug applied intrabronchially to reduce mucosal swelling. The complications related to anesthesia were hyperthermia, and trismus following succinylcholine chloride administra-

Table IV: Primary and concomittant diagnoses at 1332 laryngobronchoscopies.

Indication	Primary	Concomittant
Bronchopneumonia/infiltrate	227	307
Tracheomalacia	68	113
Intubation trauma	55	109
Tracheal granulations	36	102
Laryngomalacia	72	79
Foreign body aspiration	73	75
Granulations larynx	57	71
Edema/fibrin larynx	50	70
Congenital tracheal stenosis	56	64
Bronchial stenosis	44	61
Hemangioma larynx	47	50
Tracheal fistula/sinus	19	49
Tracheitis	21	43
Subglottic laryngitis	37	39
Acquired laryngeal stenosis	25	32
Bronchial granulations	20	31
Congenital laryngeal stenosis	20	29
Palsy vocal cords	21	25
Laryngitis	18	23
External compression trachea	18	22
Epiglottitis	21	21
Mucous plug bronchi	12	19
Congenital subglottic stenosis	9	13
Micro-/retrognathia	8	9
Hemoptysis	9	9
Tuberculosis	6	6
Bronchomalacia	2	3
Vallecular cyst	3	3
Hypoplasia bronchial tree	3	3
Miscellaneous	102	193
No abnormalities	173	173
Total	1332	1846

tion. Emergency thoracotomy was never required and there was no mortality.

Analysis of the findings showed a male predominance in cases of bronchopneumonia, foreign body aspiration, tracheal granulations, subglottic laryngitis, and bronchial granulations, while subglottic hemangioma and bronchial stenosis was mainly found in girls (Table VIII).

The numbers for indications and diagnoses refer to LBS, not to individual patients. Consequently, a high figure in tables III, IV, V, or VIII may imply either a large number of patients or many LBSs per patient.

Table V: Diagnoses at laryngobronchoscopy for the two most frequent indications.

a: LBS for inspiratory stridor (n = 287)

Diagnosis	Number
Laryngomalacia	66
Intubation trauma larynx	41
Subglottic laryngitis	33
Laryngeal edema/fibrin	28
Subglottic hemangioma	25
Tracheomalacia	20
Granulations larynx	19
Tracheitis	19
Vocal cord palsy	18
Laryngitis	15
Congenital laryngeal stenosis	15
Acquired laryngeal stenosis	12
Congenital tracheal stenosis	8
Congenital subglottic stenosis	7
External tracheal compression	7
Granulations trachea	6
Epiglottitis	5
Bronchial stenosis	3
Vallecular cyst	3
Micro-/retrognathia	3
Laryngeal foreign body	1
Miscellaneous	46
No abnormalities	12
Total	412

b: LBS for atelectasis (n = 221)

Diagnosis	Number
Bronchopneumonia	120
Bronchial stenosis	35
Granulations bronchi	11
Mucous plug	10
Tracheomalacia	10
Tracheal fistula/sinus	6
Congenital tracheal stenosis	4
Granulations trachea	3
Bronchomalacia	2
Foreign body aspiration	2
External compression trachea	2
Hypoplasia bronchial tree	2
Hemoptysis	1
Miscellaneous	29
No abnormalities	36
Total	273

DISCUSSION

The Otorhinolaryngology Department of Sophia Children's Hospital has a tradition of focusing on upper airway obstruction. Clinical studies of intubation trauma [16], congenital cricoid stenosis [17], subglottic hemangioma [73], nasal stenosis [65], and experimental research [2,5] illustrate the engagement of this department with airway stenosis. The interest is also reflected in the type of referrals and e.g. the prevalence of patients with subglottic hemangioma. Likewise, the number of indications and procedures related to obstruction of the upper airway is high in comparison with other centers [48,84,108]. Referrals include patients with intubation trauma, granulations, tracheal or bronchial stenosis, or subglottic hemangioma, who repeatedly require intubation, localized injection of steroids, removal of granulations, insertion of a laryngeal stent, and dilations. These procedures all require LBS. Another group of referrals requiring LBS concern pulmonary

Table VI: Procedures during 1332 laryngo-bronchoscopies.

Diagnosis	Number
Bronchial lavage	510
Intubation	386
Bacterial sampling	355
Steroids injection	102
Foreign body extraction	75
Biopsy	64
Extraction of granulations	29
Laryngeal stent	26
Bronchoalveolar lavage	26
Tracheotomy	21
Dilation	17
Decannulation	10
Excision of cyst	4
Miscellaneous	13
Total	1642

Table VII: Complications of 1332 laryngobronchoscopies.

Diagnosis	Number
Bradycardia, asystole	6
Hemorrhage	5
Inspiratory stridor	5
Pneumothorax	2
Intoxication	2
Anesthesia related	2
Bronchospasm	2
Respiratory insufficiency	1
Total	25
No complications	1307

anomalies. LBS is often performed at the request of neonatologists for atelectasis, ventilation problems, and postextubation stridor in prematurely born infants, or at the request of pediatric pulmonologists for bronchopneumonia of unknown origin, and foreign body aspiration. The procedures carried out at LBS then include bronchial and bronchoalveolar lavage (BAL), bacterial sampling, foreign body extraction, and biopsy. Bronchoalveolar lavage may be requested by gastroenterologists as well as pediatric pulmonologists, as this procedure serves to collect bronchial and alveolar secretions and cells for laboratory investigation in case of gastroesophageal reflux or pulmonary disorder [64].

The rare airway disorders found in this series include laryngeal cleft, hamartoma, and lymphangioma. Respiratory papillomatosis, which was frequently observed in the seventies, was diagnosed in only two cases.

The male predominance found in this population is in contrast with the findings of Friedman [48], but agrees with two studies of Puhakka [84,85].

Our study population had a wide age range with a mean of 2.0, but the majority was under one year of age. The predominance of certain anomalies accounts for this predominance of young infants. Bronchopneumonia frequently presents secondary to artificial ventilation of neonates, particularly those born prematurely. Tracheomalacia and laryngomalacia, which were also prevalent in this series, are congenital disorders likely to cause symptoms soon after birth. Another major

Table VIII: Primary diagnosis at laryngobronchoscopy, male/female ratio.

Diagnosis at LBS	Boys	Girls
Male predominance		
Bronchopneumonia	152	75
Foreign body aspiration	57	16
Tracheal granulations	27	9
Subglottic laryngitis	26	11
Bronchial granulations	17	3
Female predominance		
Subglottic hemangioma	13	34
Bronchial stenosis	14	30
Others		
Other diagnoses	361	314
No abnormalities	97	76
Total number LBS	764	568

finding, foreign body aspiration, appears to occur mainly in one to three year olds. The complication rate of 1.9% in this series is comparable to that of other workers. Puhakka reported 2% in a series of 386 bronchoscopies [85], Friedman 1.6% in 616 [48], and Godfrey 2.5% in 364 [52]. In this series, LBS was generally carried out by one of four ENT specialists. However, in our hospital LBS is a routine operation for all ENT specialists because this is an important aspect of the teaching program for ENT residents. The occurrence of complications depends on preoperative planning, the skills and cooperation of the ENT specialist, anesthetist, and operating room staff, as well as availability of the necessary equipment [63,84]. Xylometazoline hydrochloride intoxication was caused by overdosage. No such complications have occurred since, as we now administer only a few drops on cotton wool to produce localized vasoconstriction.

To establish and maintain a data base for LBS is very useful, because it provides easy access to indications, diagnoses, procedures and complications. This gives the ENT specialists an objective point of reference, an additional memory that is neither biased nor selective in what it chooses to remember. A data base also paves the way for clinical studies and statistical analyses of results.

CHAPTER 4

COMPLICATIONS OF RIGID LARYNGOBRONCHOSCOPY IN CHILDREN

ABSTRACT

Twenty-five complications (of which 2 were lethal) occurred in 1332 rigid laryngobronchoscopies performed under general anesthesia in the Sophia Children's Hospital during an eight year period (1982-1990). The nature and circumstances of these complications were studied retrospectively. Important intraoperative complications were hemorrhage and cardiac arrhythmia; postoperative complications were intoxication and respiratory complications. Three risk factors: tetralogy of Fallot, biopsy/drainage, and extraction of an aspirated foreign body appeared to be significantly associated with complications. The results of this study were compared with those reported in studies of rigid and flexible laryngobronchoscopies in children.

INTRODUCTION

The pediatric rigid laryngobronchoscopy (LBS) has become a well-established operation in many institutions. Its safety depends on several factors: anesthesia technique and monitoring of the patient, the procedures performed during the endoscopy, adequate equipment, the expertise of the staff involved, and the condition of the patient [63]. The complication rate of rigid LBS in children ranges from 2-4% (Table I) [48,52,69,85]. Minnigerode found a complication rate of 4% in 319 LBSs in patients younger than 3 months [74]. In these studies the complications are mentioned, but they have not been statistically analyzed.

A 1.9% complication rate was found in 1332 LBSs performed in 808 children in the Sophia Children's Hospital from 1982 to 1990 [61]. The records of these children were studied in order to analyze the causes and advance prevention of complications. The results of this study were compared with other reports of rigid and flexible bronchoscopy.

MATERIALS AND METHODS

Anesthesia with rigid LBS in our hospital comprises inhalation anesthesia, establishing an intravenous route, and administering a muscle relaxant. The patient is first ventilated with bag and mask, then via the bronchoscope introduced by the otolaryngologist. Standard monitoring consists of pulse rate, temperature, blood

Table 1: Review of rigid pediatric laryngobronchoscopy reported in the literature.

	Puhakka [85]	Friedman [48]	Godfrey [52]	Levy [69]	Minnigerode [74]
Anesthesia	general, muscle relax.	general	general, muscle relax.	general, muscle relax.	ketamin, diazepam
Number of LBS	386	616 (in 429 pts.)	364	110	319
Age	mean 2 3/12 yrs	23.5% < 1 yr 45.9 % < 3 yrs	35% < 3 yrs	6 m - 12 yrs	< 3 m
Sex	235 m, 151 f	214 m, 215 f	?	?	204 m, 115 f
Complications	8 (2%) laryngospasm 1 intubation 5 anesthesia 2 mortality 0	12 (1.9%) tear vocal cord 1 card. arrhythmia sev. subgl. edema 6 pneumonia 2 pneumothorax 1 mortality 2	9 (2.5%) hemorrhage 3 subgl. edema 3 hoarseness. occ. mortality 3	4 (3.6%) pneumothorax 1 morbidity minim. mortality 3	12 (3.8%) card. arrhythm. 2 hypothermia 1 burn injury 1 subgl. edema 4 pneumothorax 1 tracheotomy 3 mortality 3

sev. = several
occ. = occasionally
minim. = minimal

In these cases no exact data are reported.

pressure, capnography, ECG, and pulse oximetry (the latter since 1987). After completion of the LBS, the patient is usually intubated endotracheally and then allowed to wake up. Once normal respiration is restored, the endotracheal tube is removed.

Procedures performed during LBS were removal of granulations, crusts, or foreign bodies, biopsy, drainage of a tuberculous abscess, dilation of stenoses, local administration of steroids or other drugs, insertion of an endotracheal tube, cannula or stent, and bronchial lavage. All LBSs were performed or supervised by an experienced pediatric otolaryngologist. The condition of the patients at the time of LBS ranged widely, from severely ill to relatively healthy.

Complications were defined as all adverse effects resulting from the LBS.

Statistical analysis was performed to demonstrate the association between risk factors and complications. The dependency of complications on risk factors was specified by means of a logistic regression analysis. Because the data set contained repeated LBSs per patient, a random effect in the logistic regression model was included. The coefficients in this model can be interpreted as natural logarithms of odds ratios. These coefficients are estimated by a maximum likelihood method using the EGRET package [7]. The differences were considered to be significant if $p < 0,05$.

RESULTS

The age and sex of the patients undergoing LBS are shown in figure 1. More than half of the LBSs were performed in patients younger than one year, and 30% in patients younger than 3 months. Complications were observed in 25 out of 1332 LBSs, resulting in a complication rate of 1,9%. Age and sex of patients with a complicated LBS are shown in figure 2. We distinguished between intraoperative and postoperative complications (Table II).

INTRAOPERATIVE COMPLICATIONS (N = 13)

Hemorrhage occurred in five patients, three following drainage of a tuberculous abscess, one after biopsy, and one after extraction of a foreign body. This complication was controlled by suction, local application of adrenalin, or intravenous administration of vasopressin.

Figure 1: Age and sex of patients in 1332 laryngobronchoscopies.

Cardiac arrhythmia complicated LBS in six patients, who suffered from tracheal stenosis, diminished pulmonary function, or tetralogy of Fallot. In four patients the arrhythmia consisted of bradycardia. They recovered after ventilation with 100% oxygen, administration of atropine, or termination of the endoscopy. However, one of them died of cardiac failure three days later. One of the cases of bradycardia occurred during a failed attempt to dilate and pass a tracheal stenosis with the bronchoscope. In two other patients the arrhythmia consisted of asystole. Both were resuscitated, but one (with tetralogy of Fallot previously treated with a Waterston shunt) died six days after the LBS of repeated attacks of ventricular fibrillation. In the other hypoxemia resulted in permanent cerebral damage.

Trismus and malignant hyperthermia were complications related to anesthesia in two patients. One demonstrated an adverse reaction to succinyl choline resulting in a trismus, making intubation very difficult. The other developed malignant hyperthermia immediately after an uneventful LBS. After reversal with atropine and neostigmine the patient's temperature rose quickly from 36.2^0 C till above 40^0 C, the endotracheal CO_2 went up dramatically, and the patient looked mottled. He was successfully treated with Dantrolene i.v., and active cooling. The analysis of

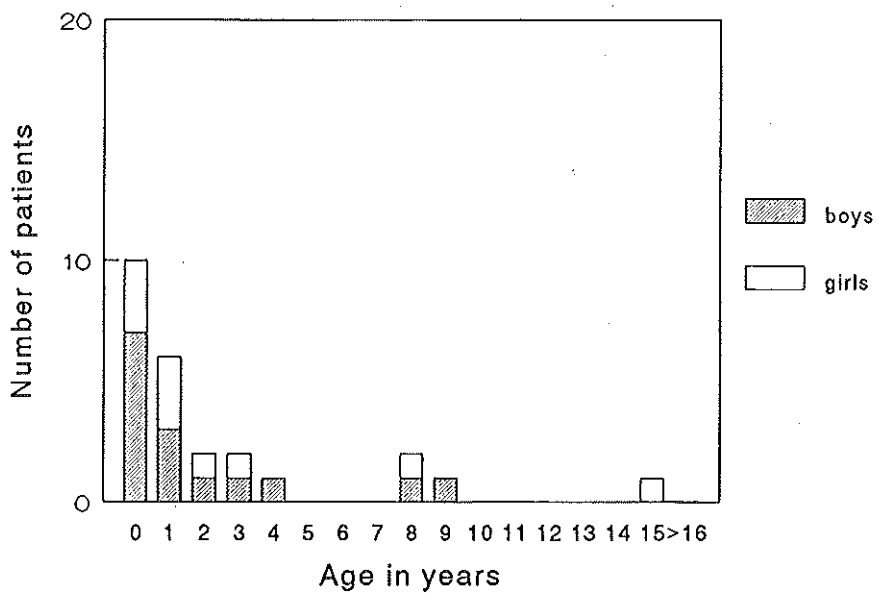

Figure 2: Age and sex of 25 patients with complications.

his parents did not demonstrate positive signs for malignant hyperthermia.

POSTOPERATIVE COMPLICATIONS (N = 12)

Subglottic edema occurred five times; in three patients after extraction of a foreign body, in one after bronchoalveolar lavage in combination with adenotonsillectomy, and in one patient with a pre-existing subglottic stenosis. In two patients the subglottic edema necessitated endotracheal intubation for 24 hours, the others were treated with intravenous dexamethasone.

Bronchospasm occurred postoperatively in two cases, and was treated successfully with bronchodilators.

Pneumothorax was discovered postoperatively in two patients. One underwent LBS for bacterial tracheitis and pneumonia, and the other had an LBS followed by a tracheotomy. Both complications were treated successfully with aspiration and drainage of the pleural space.

Postoperative respiratory insufficiency was observed in a patient with tetralogy of Fallot, cystic fibrosis and a pneumonia. Intubation and artificial ventilation were necessary after the bronchoscopy. Extubation took place after one day.

Table II: Review of 25 complications and patient data of 1332 rigid laryngobronchoscopies.

Complications	Patients	Outcome
Intraoperative (n = 13)		
Hemorrhage	Foreign body	Suction, recovered
	Tuberculous abscess	Vasopressin, recovered
	Tuberculous abscess	Vasopressin, recovered
	Biopsy	Adrenalin, recovered
	Tuberculous abscess	Suction, recovered
Cardiac arrhythmia		
Bradycardia	Fallot, pulm. infiltrate (1982)	Recovered, died after 3 d.
	Fallot, bronchopneumonia (1983)	LBS terminated, recovered
	Tracheal stenosis (1985)	Recovered
	Tracheal stenosis (1986)	Recovered
Asystole	M. Hodgkin, pulm. infiltrate (1982)	Resuscitation, cerebral damage
	Fallot (1983)	Resuscitation, died after 6 d.
Anaesthesia related	Trismus, succinylcholine	Recovered
	Malignant hyperthermia	Recovered
Postoperative (n = 12)		
Subglottic edema	Foreign body	Steroids, recovered
	Foreign body	Recovered
	Foreign body	Intubation, recovered
	Subglottic stenosis	Recovered
	ATE, BAL	Intubation, recovered
Bronchospasm	4q syndrome	Salbutamol, recovered
	COPD, BAL, biopsy	Salbutamol, recovered
Pneumothorax	Tracheitis, pneumonia	Drainage, recovered
	Laryngeal stenosis, tracheostomy	Drainage, recovered
Resp. insufficiency	Fallot, CF, atelectasis	Intubation, ventilation, recovered
Intoxication	Foreign body	Coma, recovered
	COPD, bronchial granulations	Coma, convulsion, recovered

Intoxication with xylometazoline was observed in two cases. One patient did not wake up after the bronchoscopy, due to a coma lasting several hours. The other patient also had a coma, but in addition a convulsion six hours after the bronchoscopy. Extensive neurologic examination of both patients revealed no cerebral damage, nor pre-existing neurological disease. Both patients recovered completely within 24 hours. In retrospect these complications could be attributed to an overdose of intrabronchially applied xylometazoline.

Table III: Presence of risk factors in 1332 rigid laryngobronchoscopies and in the laryngobronchoscopies with complications.

Risk factors	LBS n=1332	LBS with complications n=25 (1.9%)	
Fallot	14	4	(29%)
Biopsy/drainage	64	5	(8%)
Foreign body extraction	75	5	(7%)
Tracheal stenosis	64	2	(3%)
Male sex	764	15	(2.0%)
Female sex	568	10	(1.8%)
Prematures (NICU patients)	179	0	(0%)
Age <3 months	394	2	(0.5%)
Age 3-12 months	361	8	(2.2%)
Age 12-36 months	322	8	(2.5%)
Age >36 months	255	7	(2.7%)

ANALYSIS

Complications occurred at the same rate in boys and girls, and in the age categories older than 3 months (Table III). A complication rate of 0.5% was found in patients younger than 3 months. No complications were observed in premature patients. Tetralogy of Fallot, biopsy or drainage, foreign body extraction, and tracheal stenosis seemed to be related to complications and were preliminarily considered risk factors. The results of the statistical analysis of complications and risk factors are given in table IV. Because many patients had more than one LBS a random effect was included. In the statistical analysis the random effect was demonstrated to be not significant ($p=0.284$). Because the standard errors of the estimated coefficients increased by taking the random effect into account, the random effect was kept in the model. The odds ratio in tetralogy of Fallot was 64.9 ($p<0.001$), in biopsy/drainage 5.7 ($p=0.006$), and in foreign body extraction 7.8 ($p<0.001$). However, the odds ratio in tracheal stenosis was 3.0 ($p=0.211$), and therefore not significantly increased.

Table IV: Statistical analysis of risk factors and complications of 1332 rigid laryngobronchoscopies.
(Logistic regression with random effects)

Risk factors	Coeff.	SE	p	OR	95% CI
Constant	-5.092				
Fallot	4.172	1.03	<0.001	64.9	8.3 - 508.8
Biopsy/drainage	1.744	0.630	0.006	5.7	1.6 - 20.2
Foreign body extr.	2.055	0.593	<0.001	7.8	2.2 - 28.2
Tracheal stenosis	1.104	0.882	0.211	3.0	0.5 - 17.6

SE = Standard error of the coefficient
OR = Odds ratio
CI = Confidence interval of odds ratio

DISCUSSION

Five other studies have reported complications of rigid laryngobronchoscopy in pediatric patients [48,52,69,74,85]. The number of LBSs in our study (1332) is larger than in those studies (110-616). Our patients are younger than those in studies by Levy [69], Godfrey [52], and Friedman [48], older than those in the study by Minnigerode [74], and comparable in age to those in Puhakka's study [85]. In our study more LBSs were performed in boys (764) than in girls (568), which reflects two studies [74,85], but is in contrast with Friedman's report [48].
The number and nature of the complications in our series were not different from those reported in literature. Percentages of complications in subgroups cannot be compared with the literature, because they are not given. There is only one exception: in our study a 0.5% complication rate was found in patients younger than 3 months (394 LBSs). Minnigerode mentions a 4% complication rate in the same age group (319 LBSs) [74].
Our study differed from the others in that an analysis was made of circumstances and causes of complications. This analysis did not show any preference for sex, nor for age groups older than 3 months. Few complications were observed in the age group <3 months (0.5%) and no complications occurred in premature babies. The serious pathology very often encountered in these infants was always related to prematurity and not to the LBS. Three risk factors in our study were significantly associated with complications: tetralogy of Fallot, biopsy/drainage, and foreign body extraction. Such a relation could not be demonstrated for the fourth factor, tracheal stenosis (Table IV). Patients with tetralogy of Fallot (and thus pulmonary

artery stenosis) suffer from a diminished pulmonary circulation. Artificial ventilation during LBS causes an increase of intrathoracic pressure resulting in a further diminution of the pulmonary blood flow. This may result in more serious hypoxemia and subsequent cardiac arrhythmia. Non-invasive pulse oximetry is used with LBS since 1987. All cardiac arrhythmias are from an earlier date (Table II). This observation leads to the conclusion that pulse oximetry is indispensable with laryngobronchoscopy. Obviously instrumentation during LBS such as biopsy, transbronchial drainage of a tuberculous abscess, and extraction of a foreign body can be complicated by hemorrhage or subglottic edema. Using the rigid bronchoscope to dilate a distal tracheal stenosis easily leads to hypoxemia which causes severe cardiac arrhythmia. A safer procedure is the use of Hegar dilators via a tracheostomy, with the bronchoscope introduced through the larynx into the proximal trachea to maintain ventilation, and oxygenation, and to observe the procedure (Fig. 3).

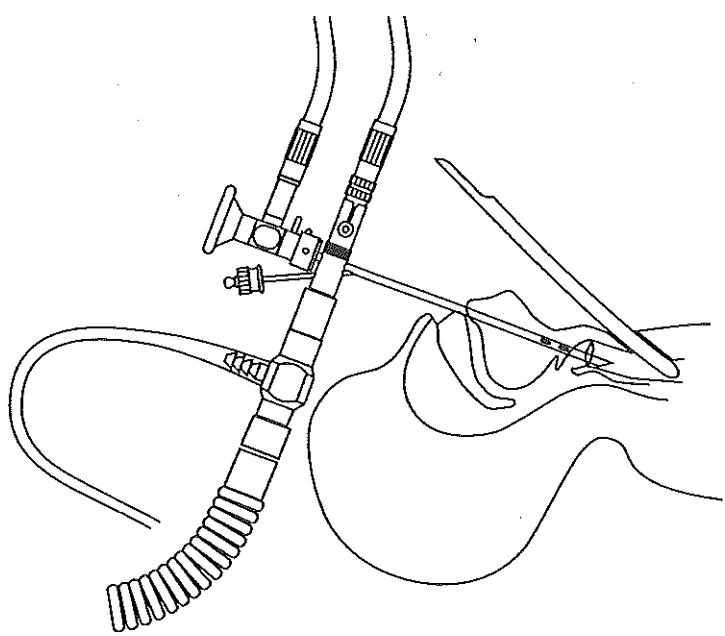

Figure 3: Dilation of a distal tracheal stenosis via a tracheostomy.

The measures to be taken in case of severe hemorrhage are described by Stradling [94]. It is very important to leave the bronchoscope at the site of the bleeding. Only then pressure can be applied at the right place to stop the bleeding, or to

prevent the blood from entering other parts of the lungs. The greatest danger of pulmonary hemorrhage is not blood loss, but the patient drowning in his own blood.

Xylometazoline was applied intrabronchially to improve visibility of an impacted foreign body in the presence of edema and granulations. Apparently an effective pulmonary resorption takes place, resulting in neurological symptoms. The effects of xylometazoline have been described in the literature [93,105]. Use of the drug was not abandoned, but care was taken to administer intrabronchially only a few drops on a swab.

Flexible bronchoscopy in children, introduced in the 1970s by Wood, is becoming increasingly popular especially with pediatric pulmonologists [44,47,79,108]. Its main advantage is the use of topical anesthesia and sedation, while in rigid LBS general anesthesia is always needed. The disadvantages of flexible bronchoscopy are the obstruction of a large proportion of the airway in small infants, the lower quality of the optics, and the difficulties with foreign body extraction and other procedures. The complication rates of flexible and rigid bronchoscopy are difficult to compare, because of differences in anesthesia technique and patient selection. The indications for flexible bronchoscopy are more often of pulmonary origin, whereas the (relative) contraindications are hemorrhagic diathesis, cardiac arrhythmia, hypoxemia, foreign body aspiration, and respiratory distress [79,108]. These conditions increase the risk of complications, and may result in referral for rigid bronchoscopy. The nature of the complications of flexible bronchoscopy also shows some differences: bradycardia, epistaxis, anesthetic problems, laryngospasm, pneumothorax, hemorrhage, and no subglottic edema [47,108]. The complication rate of flexible bronchoscopy ranges from 2.2 to 8% [44,47,79,108]. Wood's [108] large series of 1095 flexible bronchoscopies does not differ much from our series of rigid LBSs in ages and indications. An important difference, however, is the absence of foreign body extractions in flexible bronchoscopies. Wood's complication rate is 2.9%, with no mortality. In our series of rigid LBSs the complication rate was 1.9%, and two patients died. A causal relationship between the LBS and these patients' death after several days is questionable. A flexible bronchoscopy in these critically ill patients was possibly contraindicated. We therefore do not agree with Wood's statement that flexible procedures are associated with less risk of complication [108].

CONCLUSIONS

The 1.9% complication rate of rigid LBS in our patients was not influenced by sex or age, with exception of patients younger than 3 months of age, especially premature babies, in which the complication rate was much lower than the mean.

The complications observed were hemorrhage, cardiac arrhythmia, trismus, malignant hyperthermia, subglottic edema, respiratory disorders, and intoxication.

Risk factors were tetralogy of Fallot, biopsy/drainage, and foreign body extraction.

Pulse oximetry is mandatory to detect hypoxemia and to prevent cardiac arrhythmia.

Administration of high doses xylometazoline in the bronchial tree may result in coma and convulsions.

The complication rate of rigid laryngobronchoscopy is not higher than that of flexible LBS.

CHAPTER 5

FOREIGN BODY ASPIRATION IN CHILDREN: THE DIAGNOSTIC VALUE OF SIGNS, SYMPTOMS AND PREOPERATIVE EXAMINATION

ABSTRACT

One hundred and fifteen patients, between six months and twelve years of age had bronchoscopy on suspicion of foreign body aspiration. They were referred under the initial diagnoses foreign body aspiration (80), pneumonia (34), and subglottic laryngitis (1). In 85 infants a foreign body could be extracted. In the other 30 patients no foreign body was identified; they suffered from pneumonia or other respiratory tract infections. The initial diagnosis foreign body aspiration was correct in 72% of the cases. In these cases the period between aspiration and extraction of the foreign body was on average 6 days, compared with 55 days if the initial diagnosis was pneumonia or subglottic laryngitis.

The sensitivity of the symptoms choking and cough was fairly high (81 and 78% respectively), but the specificity was poor. The sensitivity of a chest X-ray and of in- and expiratory X-rays was 82 and 80% respectively, and the specificity 44 and 55%.

It is concluded that the diagnosis foreign body aspiration is too often missed, and that apart from bronchoscopy most diagnostic tools are of little value.

INTRODUCTION

Aspiration of a foreign body occurs mostly in infants aged between 1 and 3 years [19,66,71,98,103]. In the United States aspiration is even the most important cause of accidental death in infants less than one year of age [75]. In 1987 in the Netherlands, with a population of 15 million people, aspiration of a foreign body was diagnosed in 140 children under the age of 15 years (103 male and 37 female). Nineteen patients (11 boys and 8 girls) died as an immediate result of aspiration [22]. If the infant survived the acute episode, and the foreign body remained in the respiratory tract, the patient presented with a chronic respiratory tract infection.

The possible diagnostics of a foreign body aspiration concern the chest X-ray, in- and expiratory chest X-rays, or fluoroscopy [24,92,96]. More controversial procedures are ventilation-perfusion scan, CT-scan, magnetic resonance imaging, and xero-radiography [20,24,98]. The abnormalities found with the last mentioned methods are usually not specific to a foreign body. If the diagnosis is not considered, extraction of a foreign body can be delayed for weeks up to several years. In

the various reports the average interval between aspiration and removal varies from 14 to 28 days [71,86,97]. Only bronchoscopy will give certainty about the diagnosis of a foreign body [96].

A total of 115 infants underwent bronchoscopy for suspected foreign body aspiration in Sophia Children Hospital over a period of ten years. The records of these patients were studied to examine the value of signs and symptoms, and of preoperative examinations, using the findings at bronchoscopy as a point of reference.

MATERIALS AND METHODS

All infants, who underwent bronchoscopy for suspected foreign body aspiration in Sophia Children's Hospital from 1980 to 1990, entered the study. Their history, signs and symptoms, the initial and final diagnosis, the time between these diagnoses, and the findings at preoperative examination were recorded. Examination of these patients included chest X-rays on in- and expiration, or fluoroscopy. The specificity and sensitivity of a history of choking, cough, auscultation, and roentgenographic examination was measured.

The initial diagnosis was defined as the diagnosis made at the patient's first presentation usually by a general practitioner or a pediatrician. All patients with an initial diagnosis foreign body aspiration underwent bronchoscopy. Patients with another initial diagnosis were selected for bronchoscopy by the otolaryngologist because of a typical history of choking, or persistent pneumonia, often with an acute onset in a previously healthy child.

The final diagnosis was the diagnosis after bronchoscopy. The findings at bronchoscopy were considered to be decisive for the diagnosis foreign body aspiration.

Table I: Initial and final diagnosis in 115 patients who underwent bronchoscopy for suspected aspiration of a foreign body.

Initial diagnosis	Final diagnosis	
Foreign body	Foreign body	61
Pneumonia	Foreign body	23
Subglottic laryngitis	Foreign body	1
Pneumonia	No foreign body	11
Foreign body	No foreign body	19

RESULTS

A total of 115 patients entered the study. The initial and the final diagnoses are presented in table I. The signs, symptoms, and results of examination of all patients are listed in table II. The same data of patients, divided in subgroups according to initial and final diagnosis, are given in table III.

A foreign body was found in 85 patients of which 69 (81%) presented with a history of choking. Choking was also present in 20 patients of the group of 30 in whom no foreign body was found. In our study the sensitivity of a choking spell was 81%, the specificity 33%. Further symptoms were cough and abnormal auscultatory findings. The sensitivity of both findings was 78%, the specificity 37% and 50%, respectively.

Table II: History and examination in 115 patients with suspected foreign body aspiration.

Signs and symptoms	Foreign body (n=85)		No foreign body (n=30)	
choking	69	(81%)	20	(67%)
cough	66	(78%)	19	(63%)
abnormal auscultation	66	(78%)	15	(50%)
stridor (wheezing)	51	(60%)	18	(60%)
abnormal chest X-ray	45/55	(82%)	9/16	(56%)
abnormal in/expir. X-ray	19/24	(79%)	5/11	(45%)

Patients with an incorrect initial diagnosis pneumonia and a final diagnosis foreign body had a history of choking in 61% of the cases (Table III).

Examination of the 115 patients included in 71 cases a conventional chest X-ray. These chest X-rays demonstrated disorders in 54 patients. The disorders were hyperinflation, infiltrates and atelectasis. The sensitivity of a chest X-ray in this study was 82%, the specificity 44%.

In 35 patients a chest X-ray at in- and expiration or fluoroscopy was made. Disorders were present in 24 cases. The sensitivity was 79%, the specificity was 55% (Table II).

In 9 patients no X-rays were made.

In 61 (72%) of the 85 patients with a final diagnosis of foreign body aspiration the initial diagnosis was correct. Twenty-three patients (27%) were initially incorrectly diagnosed as pneumonia, and one as subglottic laryngitis. (Table I). The time that

Table III: History and examination in patients subgroups.

Both initial and final diagnosis foreign body. (n = 61)		
choking	54	(89%)
cough	42	(69%)
abnormal auscultation	43	(70%)
stridor	42	(69%)
abnormal chest X-ray	30/37	(81%)
abnormal in-/expiratory X-ray	15/18	(83%)
Initial diagnosis pneumonia, final diagnosis foreign body. (n = 23)		
choking	14	(61%)
cough	23	(100%)
abnormal auscultation	22	(96%)
stridor	8	(35%)
abnormal chest X-ray	15/17	(88%)
abnormal in-/expiratory X-ray	4/6	(67%)
Initial diagnosis pneumonia, final diagnosis no foreign body. (n = 11)		
choking	6	(55%)
cough	9	(82%)
abnormal auscultation	3	(27%)
stridor	4	(36%)
abnormal chest X-ray	6/6	(100%)
abnormal in-/expiratory X-ray	1/2	(50%)
Initial diagnosis foreign body, final diagnosis no foreign body. (n = 19)		
choking	14	(74%)
cough	10	(53%)
abnormal auscultation	12	(63%)
stridor	14	(74%)
abnormal chest X-ray	3/10	(30%)
abnormal in-/expiratory X-ray	4/9	(44%)

elapsed between aspiration and removal of the foreign body varied from a few hours to 455 days, the average being 19 days. If the incorrect initial diagnosis was pneumonia the average delay was 55 days (2-455), and only 6 days (0-90) when a foreign body was diagnosed immediately.

DISCUSSION

Most of the symptoms and signs of a foreign body merely represent the resulting respiratory tract infection, and are therefore not characteristic.

A history of choking appeared to be rather sensitive, but was not very specific. Possibly some aspirated foreign bodies were expectorated before bronchoscopy. Esclamado and Banerjee have reported a better sensitivity of about 90%, but do not mention specificity [9,42].

Cough in our patients was a less sensitive symptom (sensitivity of 78%), and little specific (37%).

In literature a chest X-ray is considered to have a sensitivity of 60-90% [20,86,96, 106], which is in accordance with the 82% of this study.

The combination of in- and expiratory chest X-rays are reported to have a sensitivity of 68% and a specificity of 67% [96]. In our study the sensitivity was somewhat better (80%), but the specificity appeared to be less (55%).

This type of roentgenographic examination is supposed to be helpful in finding more characteristic abnormalities, such as an area with permanent hyperinflation or a mediastinal shift. However the figures in this study as well as in literature demonstrate a rather poor specificity.

In our patients the average interval between aspiration and removal of the foreign body was 19 days, which is not different from literature. No publication has made a distinction between the intervals after a correct and an incorrect initial diagnosis. Aspiration of a foreign body was incorrectly diagnosed and treated as pneumonia in 27% of our patients. The consequence for these children was serious: the interval between aspiration and removal was on average 49 days longer than in patients with a correct initial diagnosis. The reasons for prolongation of the interval were that the possibility of a foreign body as a cause of pneumonia was at first not considered, even though a history of choking often was present. The chest X-rays often showed abnormalities, but these were not specific for a foreign body.

CONCLUSIONS

Aspiration of a foreign body must be considered if a history of choking exists, or if a pneumonia does not respond well to the usual treatment, especially if the child was previously in good health.

Roentgenographic examinations are of little value for excluding the diagnosis. They are however important to localize a foreign body, and to increase the probability of the diagnosis.

If aspiration of a foreign body is considered, rigid bronchoscopy is mandatory.

CHAPTER 6

THERAPEUTIC REINTUBATION FOR POST-INTUBATION LARYNGOTRACHEAL INJURY IN PRETERM INFANTS

ABSTRACT

The failure to extubate a preterm infant after prolonged intubation is often caused by laryngotracheal injury. This condition is treated by tracheotomy, anterior cricoid split, or, often, by reintubation and subsequent extubation attempts in a later stage.

To assess the value of reintubation as treatment of post-intubation injury, we retrospectively studied a group of preterm infants from the neonatal intensive care unit in the Sophia Children's Hospital. Three categories of injury were distinguished according to the findings at laryngobronchoscopy: a. edema or superficial lesions, b. ulcerations and edema, and c. granulations.

Twenty-three infants were therapeutically reintubated after post-intubation injury was diagnosed, for a mean period of 17 days. The therapy was successful in 22 patients, and a failure in one. The follow-up period was a mean 34 months.

The result and the duration of the treatment vary with the category of the injury and the condition of the patient. Therapeutic reintubation is compared with alternatives such as anterior cricoid split and tracheotomy. We conclude that reintubation is a valuable therapy that should precede the decision for surgery.

INTRODUCTION

Prolonged intubation and artificial ventilation play an important part in present neonatal intensive care. Pathological changes in the larynx or trachea following prolonged intubation have been reported in 74-100% of the autopsied neonates. In surviving neonates severe airway injury, including ulceration and granulation, occurs in 44-47%, and subglottic stenosis in 0.7-9% [36,37,43,95]. These injuries may result in failure to extubate the patient. The condition can be diagnosed and categorized by rigid laryngobronchoscopy, and then treated by reintubation, tracheotomy, or anterior cricoid split [15,29,32,33].

The present retrospective study concerns the efficacy of therapeutic reintubation in a group of preterm infants in whom extubation had failed, as a result of endoscopically confirmed post-intubation injury of larynx or trachea.

Table I: Duration of intubation and artificial ventilation preceding the diagnosis of post-intubation injury at laryngobronchoscopy.

	n	Intubation days	Ventilation days
All patients	23	34 (3-114)	25 (1-102)
- Edema (a)	9	24 (3-102)	23 (1-102)
- Ulcerations (b)	3	22 (16-30)	22 (16-29)
- Granulations (c)	11	45 (7-114)	28 (2-55)

PATIENTS AND METHODS

The study group consisted of 23 consecutive preterm infants (15 male, 8 female), admitted to the Neonatal Intensive Care Unit (NICU) of the Sophia Children's Hospital, Rotterdam, the Netherlands, between 1982 and 1990 [61]. The selection criteria were: prolonged intubation and artificial ventilation resulting in an intubation injury, confirmed and categorized by laryngobronchoscopy with rigid telescopes under general anesthesia in the operating theater [15]. All patients were treated by reintubation.

The mean postconceptual age of the patients at birth was 30 weeks (25-36). The patients had been intubated and artificially ventilated for respiratory insufficiency, hyaline membrane syndrome, diaphragmatic hernia, patent ductus arteriosus, or cyanotic spells. Intubation had lasted a mean 34 days (3-114), and artificial ventilation a mean 25 days (1-102) (Table I). When they could breath spontaneously, ventilation was stopped and extubation was attempted, but proved to be unsuccessful (Fig. 1). Subsequent laryngobronchoscopy demonstrated the cause of this failure to be laryngeal or tracheal injury related to prolonged intubation. The lesions narrowing the airway were classified into one of the following categories: (a) edema and/or superficial lesions of the (sub)mucosa, (b) deep ulceration of perichondrium and cartilage, and edema, or (c) granulation tissue. The laryngotracheal injury was subsequently treated by nasotracheal reintubation, without artificial ventilation. A PVC (polyvinylchloride) tube was used in 22 patients, and a siliconized silicone rubber tube in the remaining one [18]. The size of the tube was 'loose fit', permitting leakage of air at the end of an insufflation. After some time (Fig. 1) extubation was reattempted. Every attempt of extubation was preceded and followed by intravenous administration of dexamethasone.

Subsequently the category of pathology determined by laryngobronchoscopy, and the outcome of therapeutic reintubation were studied.

RESULTS

The mean age of the 23 infants at laryngobronchoscopy was 37 days (3-115).

Edema or superficial lesions (category a) of the glottis and the subglottic area were found in 9 patients.

Ulcerations and edema (category b) were observed in 3 patients at the glottic and subglottic level. Posterior parts of the cricoid cartilage were not covered by mucosa or perichondrium and exposed to the airway.

Granulation tissue (category c) was present in 11 patients. The granulations involved the glottis in 4 patients, the subglottis in 3, both glottis and subglottis in 2, and the trachea in 2 patients. In 10 of these 11 patients extensive granulation tissue completely obstructed the lumen due to a circular localization. The other patient had a left sided subglottic cyst and granulations.

Table II: Results and duration of therapeutic reintubation in preterm infants with post-intubation laryngeal or tracheal injury.

	n	Result	Days intubation
All patients	23	Cured 22	17 (1-66)
		Failed 1	35
- Edema (a)	9	Cured 9	8 (2-30)
- Ulcerations (b)	3	Cured 3	13 (5-23)
- Granulations (c)	11	Cured 10	27 (1-66)
		Failed 1	35

THERAPEUTIC REINTUBATION AND FINAL EXTUBATION

In 17 patients the first extubation attempt was successful, in 5 others two or more attempts were necessary. Eventually 22 infants were successfully extubated, after a mean reintubation period of 17 days (1-66) (Table II). One patient needed a tracheotomy when reintubation apparently failed as a result of extensive circular subglottic granulations. He could be decannulated four months later without any further treatment.

All nine patients with **edema or superficial lesions (category a)** could be extubated, after a mean reintubation period of 8 days (2-30), the three with **ulcerations (category b)** after a mean 13 days (5-23). In the group **granulation tissue (category c)** 10 out of 11 patients were finally extubated after a mean 27 days (1-66). The only remaining patient had a tracheotomy (Table II).

COMPLICATIONS

Complications of therapeutic reintubation occurred in 10 out of 23 patients. The complications were: pulmonary infection in 8, accidental extubation in 8, laryngeal granulations in 2, aspiration in 2, tube obstruction in 1, and feeding problems in 1 patient. Three patients needed artificial ventilation for a few days because of a pulmonary infection.

Table III: Patients with complaints during the period of follow-up.
(LBS = laryngobronchoscopy; pseudocroup is defined as a respiratory tract infection with inspiratory stridor, and no need for intubation)

Pat	Cat	Follow-up disorders	Follow-up duration (m)	LBS
13	c	stricture post. comm.	60	+
15	c	pseudocroup 1x	30	-
17	c	pseudocroup ± 3x	26	-
18	c	audible insp. on exertion	65	-
23	c	audible insp. on exertion, hoarseness	36	-

LATE RESULTS

The mean follow-up period of the 22 successfully treated patients is 34 months (4-68). Seventeen patients showed no signs of airway disorder. Five patients, all originally categorized in the granulations group (c), had minor complaints that could point to laryngeal dysfunction (Table III). One patient (no. 18) still demonstrates a slightly audible inspiration on exertion only after 65 months, a second (no. 23) has an audible inspiration on exertion and hoarseness after 36 months, the third (no. 15) had one minor episode of pseudocroup (a respiratory tract infection with inspiratory stridor, but no need for intubation) one year after extubation, but no complaints 18 months since then, the fourth (no. 17) had a few episodes of pseudocroup during the follow-up period of 26 months after extubation, but has

been lost to follow-up for five years now. The problems of these four patients did not necessitate another laryngoscopy. The fifth patient (no. 13), demonstrated a slowly progressive inspiratory stridor for many years. Laryngobronchoscopy five years after extubation revealed a stricture in the posterior commissure of the glottis/subglottis, leaving a lumen of 4 mm. Further treatment will probably be necessary in this case.

DISCUSSION

Edema or superficial lesions (category a) in infants are generally treated by intubation or tracheotomy [25,68]. There is no consensus about the optimal treatment of ulcerations (category b) and granulations (category c). Besides tracheotomy, intubation [40], removal of granulations with a forceps or laser surgery [62], or anterior cricoid split [25], have been advocated.

The present study demonstrates that the majority of preterm infants with post-intubation edema, (sub)mucosal lesions, ulcerations, and granulations can be cured with therapeutic reintubation, thus avoiding a tracheotomy and further surgery.

Extubation was successful in the 12 patients with category a and b lesions (superficial lesions, edema, and ulcerations) after a relatively short period of reintubation (means of 8 and 13 days respectively). Interestingly ulcers appeared to heal within a few weeks even with an endotracheal tube in place, which confirms previous observations [37,54].

Nearly half of the patients with category a injury were successfully extubated within three days (Fig. 1). Therefore it seems advisable to attempt a first extubation in this group of patients after two or three days. If the extubation does not succeed, the next attempts should be spaced according to the findings at laryngobronchoscopy, and the observation of the respiration between extubation and reintubation.

In case of ulceration and associated edema (category b) extubation can be attempted after 3-7 days, depending on the severity of the injury. Attempts may be repeated at weekly intervals and should be successful within 4 weeks. If the treatment is not successful in that period tracheotomy should be performed.

For granulations (category c) the success rate was less, 10 out of 11 patients could be extubated, while the period of reintubation was longer (27 days). Secondary stenosis five years after the final extubation was observed in a patient from

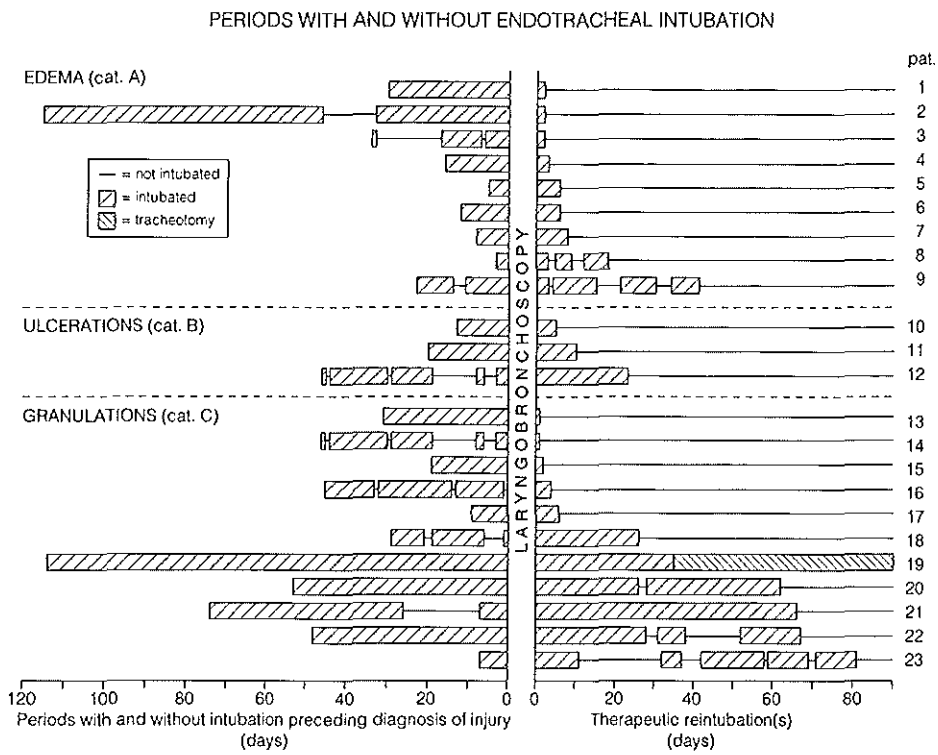

Figure 1: Periods with and without intubation since birth, before and after the diagnosis of intubation injury. The patients have been divided into three categories of injury: a. edema or superficial lesions, b. ulcerations and edema and c. granulations. (The periods without intubation are only shown if they lasted more than 12 hours).

category c. Five of the 10 successfully treated patients with granulations (category c) were extubated within 10 days. The other five needed a longer period of intubation (Fig. 1). We suggest to attempt extubation after about 10 days of therapeutic reintubation, and repeat this at intervals of the same duration. The maximum period of therapeutic intubation in category c patients depends on factors as respiratory infections, feeding difficulties, and the involution of the granulations. Alternatives for therapeutic reintubation in category b and c are tracheotomy and anterior cricoid split.

A tracheotomy is less unpleasant to the infant than an endotracheal intubation. However the complication rate (60%) of a tracheotomy in preterm infants is higher

than in full-term infants (40%) [50], while the mortality in infants is 0-5% [49,50]. Reported complications are pneumothorax, accidental decannulation, tube obstruction, granulation tissue, tracheomalacia, tracheal stenosis, and hemorrhage [34,49,50,51]. The complication rate in our patients was 43% with no mortality. In our view tracheotomy is indicated if therapeutic intubation appears to fail.
The anterior cricoid split is not indicated in patients with minor laryngeal injury or ulcerations [29,33,25]. Anterior cricoid split appears in our experience less successful if fresh granulation tissue is present. The operation induces further granulation formation and delays extubation. The operation seems useful in intubated infants with 'old' granulations or mild subglottic stenosis, in the absence of respiratory tract infection. Therefore we prefer intubation to anterior cricoid split in patients with granulations.

CONCLUSIONS

Therapeutic reintubation is the treatment of choice in preterm infants with edema, superficial lesions, or ulceration (categories a and b) of the larynx or trachea. Therapeutic reintubation is less successful in patients with granulations, but the alternatives tracheotomy and anterior cricoid split have important disadvantages.
Intubation injury in preterm infants is therefore preferably treated with reintubation. If that treatment fails, or if the infant's tolerance does not permit continuation of the intubation, tracheotomy is indicated. Anterior cricoid split is only considered in case of old granulations.

CHAPTER 7

ACQUIRED LARYNGEAL STENOSIS IN INFANTS AND CHILDREN TREATED BY LARYNGOFISSURE AND STENTING

ABSTRACT

Thirty young patients with acquired laryngeal stenosis were treated by means of a laryngofissure. The age distribution suggested two subgroups: infants aged 0-2 years (n=24), and children aged 6-16 years (n=6). The causes of the stenosis and the treatment results were different in these subgroups. Treatment resulted in successful decannulation in 21 of 24 infants, and in 4 of 6 children. In 3 patients the therapy failed, and 2 patients died after fatal complications. Decannulation rate, duration of the tracheotomy, and the number of reoperations compare favorably to the results of other centers. The longer duration of stenting, and the high mortality rate may be considered disadvantages.

INTRODUCTION

Acquired laryngeal stenosis in children is usually caused by prolonged intubation, and seldom by acute endolaryngeal injury. Its treatment has varied from a tracheotomy and wait-and-see in the past, to extensive surgery in combination with endoscopic procedures nowadays [23,25,27,29,38,39,41,45,46,57,72,76,87,102, 104,107]. After surgery the laryngeal lumen is usually 'stented' for some weeks or even months [1,27,41,53,67,77,78,89,109,110].

Three types of current surgical treatment may be distinguished: laryngofissure, laryngofissure and interpositioning of a graft, and resection of the stenotic area.

The laryngofissure is based on the concept of augmenting the laryngeal lumen by enlarging the cartilaginous framework rather than by excision of scar tissue, as introduced by Réthi, and later supported by Grahne, Evans, Cotton, and Fearon [27,41,45,56,87].

An anterior incision of a child's cricoid cartilage results in a gap of a few millimeters [29], which could be explained by the release of interlocked stresses in the cartilaginous ring [4,10]. Additional division of the posterior cricoid lamina creates posteriorly a 'hinge-joint' which permits the cricoid halves to rotate outwardly by the contractional force of the cricopharyngeus muscle [10]. This outward rotation could be counteracted by scar tissue formation in the anterior gap. Retraction of

scar tissue can be prevented by long-term stenting or by interpositioning of a graft. A recent development is resection of the stenotic area including a part of the deformed cricoid and primary anastomosis larynx and trachea [76].

Another approach is endoscopic laser surgery, aiming at expanding the stenotic lumen by evaporation of scar tissue [58].

The results of the different treatment modalities are usually compared on the basis of decannulation rates. Other aspects, however, play an important role in the outcome as well: selection of patients, duration of treatment, and complication rate. The present study aims to describe the experience with laryngofissure in the Sophia Children's Hospital (Erasmus University Rotterdam) and compare the results with those in other centers.

Table Ia: Endotracheal tube sizes (outer diameter, O.D.) according to age (after Bos) [21].

Age (months)	Diameter (mm O.D.)	Age (years)	Diameter (mm O.D.)
0	4	2	7.3
1	4.7	4	8
3	5.3	6	8.7
6	6	8	9.3
12	6.7	10	10
		12	10.7
		16	11.3

Table Ib: Cotton classification of stenosis [27].

Grade 1 = <70% obstruction
Grade 2 = 70-90% obstruction
Grade 3 = >90% obstruction, or obliteration not including the glottis
Grade 4 = obliteration including the glottis

METHODS: DIAGNOSIS AND TREATMENT

Both flexible and open rigid laryngoscopy under general anesthesia were part of the preoperative evaluation procedure. Patients with immobility of the laryngeal entrance, or with severe tracheomalacia, were not included in this series. Open rigid (video)laryngoscopy was performed to record the exact site and length of the stenosis. The consistency and the diameter of the stenosis were examined with Hegar dilators (Fig. 1). The measured diameter was compared with the expected

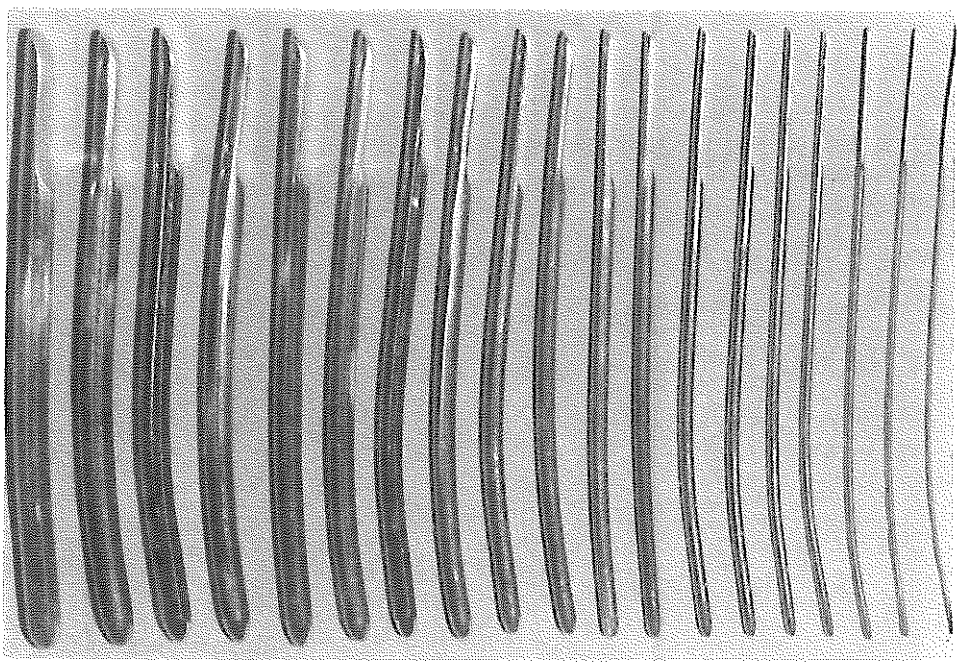

Figure 1: *Hegar dilators used to palpate and calibrate the stenosis.*

diameter, taken from a table with age-dependent endotracheal tube sizes (Table Ia) [21]. Subsequently the stenosis was classified according to Cotton (grade 1-4) (Table Ib) [27].

Preoperative evaluation also included esophageal pH measurement. If gastro-esophageal reflux was noted, treatment was started well before the stenosis treatment.

Because all patients required intubation or a tracheotomy, preoperative pulmonary function tests and speech tests were usually not possible.

A laryngofissure was not performed until the infant had reached a weight of 7 kg. Two types of operations, both without interpositioning a graft, were performed:

(1) Anterior laryngofissure: a midline incision, or a castellated incision as described by Evans [41], through thyroid, cricoid and tracheal rings without division of the posterior cricoid lamina.

(2) Anterior and posterior laryngofissure: an anterior incision of the thyroid cartilage, the cricoid, and the proximal tracheal rings, and posteriorly a midline incision of the posterior cricoid lamina [87]. Postoperatively all patients were

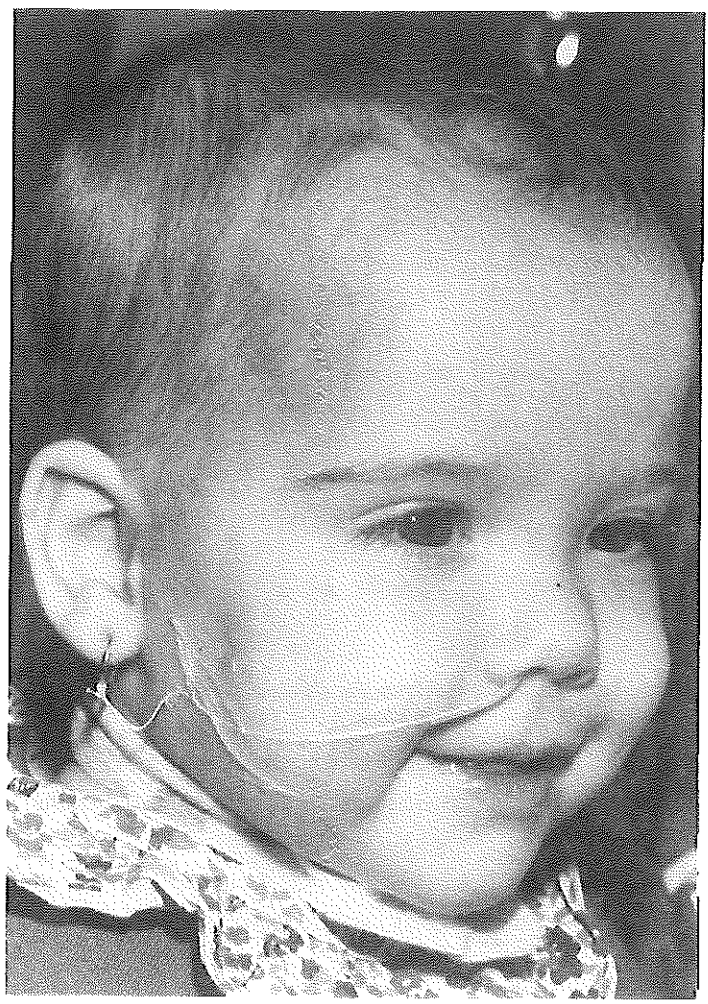

Figure 2: The fixation of a laryngeal stent to the earlobe.

allowed to wake up and breathe spontaneously.

Postoperatively the laryngeal lumen was splinted by either a siliconized silicone rubber stent or a nasotracheal tube of the same material. The stents and tubes were designed by one of the authors (RNPB) [17]. The cylindrical stent is available in all desired lengths and diameters. The caudal end is sutured to a metal tracheotomy tube with an inner cannula. By means of a suture through pharynx and nose the

cranial end of the stent is anchored to a small ring (from a jeweler) pierced through an earlobe (Fig. 2).

The stent or tube had to be replaced by a new one every three to four weeks because it was colonized by micro-organisms. Granulations were then removed or injected with corticosteroids. Usually this procedure lasted approximately 10 minutes under general anesthesia. Most patients were discharged a few weeks after the laryngofissure, and were nursed by the parents. The stenting was discontinued as soon as granulations had disappeared, and the mucosa had healed. Subsequently the patient was decannulated after a few months of observation. Laryngobronchoscopy, pulmonary function tests, and speech tests were not routinely performed during the follow-up period.

Figure 3: Number of patients and grade of stenosis per age group.

RESULTS

PREOPERATIVE EVALUATION

The reported series consists of a consecutive group of 30 young patients (19 boys and 11 girls) presenting with an acquired laryngeal or proximal tracheal stenosis between 1976 and 1993. Two subgroups can be distinguished: 24 infants (14 boys,

Table II: Laryngofissure patients in the Rotterdam Sophia Children's Hospital.

Patients			LBS			Therapy			Decan /extub	Result	FU
Subgroup infants (n = 24)											
1	f	3y9m	○○●○	-g	TT	AP	S	99	176	+	0y9m
2	f	2y5m	○●●○	-g	TT	AP	S	18	-	D	-
3	m	1y2m	○●●○	-g	NT	AP	S	84	97	+	0y9m
4	m	1y1m	○○●○	2s	TT	AP	S	92	164	+	0y4m
5	m	1y5m	○○●○	2sc	TT	AP	S	82	224	+	7y8m
6	m	1y4m	○○●○	2sc	TT	AP	S	204	218	+	2y0m
7	f	1y2m	○○●○	2sc	TT	AP	S	87	94	+	0y5m
8	f	1y1m	○○●○	2t	TT	A	S	49	63	+	5y9m
9	f	0y5m	○●●○	2s	NT	AP	S	35	43	+	5y9m
10	m	0y10m	○●●○	2s	TT	A	S	153	155	+	0y1m
11	m	1y8m	○○●●	2s	TT	AP	S	170	363	+	2y2m
12	f	1y4m	○○●●	2sc	TT	A	S	61	-	D	-
13	m	1y9m	○○●○	3s	TT	AP	S	273	397	+	6y5m
14	m	1y1m	○○●○	3s	TT	AP	S	125	132	+	0y6m
15	m	2y3m	○○●○	3s	TT	AP	S	43	155	+	1y6m
16	f	1y1m	○○●○	3s	TT	AP	S	113	154	+	1y2m
17	m	1y1m	○○●○	3s	TT	AP	S	183	226	+	0y10m
18	f	0y10m	○○●○	3s	TT	AP	S	140	TT	-	Reop. +
19	m	1y0m	○○●○	3s	TT	AP	S	42	49	+	0y4m
20	f	1y6m	○○●○	3sc	TT	A	NT	102	102	+	13y6m
21	f	0y9m	○●●○	3s	TT	AP	S	113	205	+	0y4m
22	m	0y6m	○●●○	3sc	TT	A	NT	66	66	+	9y5m
23	m	1y1m	●●●●	3s	TT	AP	S	285	385	+	2y7m
24	m	1y7m	●●●○	4s	TT	A	S	107	449	+	0y3m
Subgroup children (n = 6)											
25	m	6y10m	○○●○	3s	NT	Ac	NT	48	48	+	11y0m
26	m	15y4m	○○●○	3s	TT	A	S	181	258	+	1y9m
27	m	9y4m	○●●○	3s	NT	AP	S	90	97	+	2y0m
28	f	6y1m	●●●○	3st	TT	AP	S	189	343	+	4y5m
29	m	8y3m	○●●○	4s	TT	AP	S	350	TT	-	Lost
30	m	12y0m	○●●○	4s	TT	AP	S	230	TT	-	Reop. +

10 girls), most of whom were operated on in their second year of life, and 6 children (5 boys, 1 girl) operated on between 6 and 16 years of age (Table II).

In the **subgroup infants** the causes of the stenoses were: prolonged intubation in relation to prematurity (9 cases), traumatic intubation (5), repeated episodes of subglottic laryngitis (5), prolonged intubation in relation to surgery (4), and endolaryngeal tumor (1). The stenoses had been present between 3 months and 2 years at the time of the operation. Three infants had subglottic granulations completely obstructing the lumen, nine had grade 2 stenoses, eleven grade 3, and one grade 4 (Fig. 3). The indication for surgical treatment was a persistent need of

Table II: Legends.

Serial number of patient, sex, and age at operation; localization, grade and consistency of the stenosis, tracheotomy or intubation; type of operation, stent or tube and duration; days from operation to decannulation (or extubation); result of the treatment; follow-up.

Laryngobronchoscopy (LBS):

○○○○/●●●●	= supraglottis / glottis / subglottis / proximal trachea (○ = no stenosis, ● = stenosis)
1,2,3,4	= stenosis grade (Cotton)
g	= granulation tissue
s	= scar tissue
c	= cartilaginous stenosis
t	= tumor
NT	= intubation
TT	= tracheotomy

Surgical treatment:

A	= anterior incision
Ac	= anterior castellated incision
P	= posterior cricoid incision

Further treatment:

S	= laryngeal stent
NT	= nasotracheal intubation

Result:

+	= decannulation or extubation
-	= no decannulation or extubation
D	= died

Follow-up (FU):

Reop. +	= successfully reoperated
Lost	= lost from follow-up

a tracheotomy (22 cases), or repeated unsuccessful attempts to extubate the patients in spite of conservative treatment (2 cases).

In the **subgroup children** the causes were: prolonged intubation after a traffic accident (2), prolonged intubation in relation to surgery (2), prolonged intubation for M. Guillain Barré (1), and endolaryngeal tumor (1). These patients were operated on between 4 months and 10 years after the endolaryngeal injury. Four grade 3, and two grade 4 stenoses were observed (Fig. 3). Four children had a tracheotomy, two had been treated previously with intubation, but without success.

More data concerning the individual patients, and the observed stenosis, are listed in table II. No patients died in the period between presentation and surgery.

SURGICAL TREATMENT AND RESULTS

In the **subgroup infants** the mean age at the time of surgery was 17 months (5-45 m.). Six anterior laryngofissures, and 18 anterior and posterior laryngofissures were performed. Twenty-two infants were stented, two intubated after surgery. Stenting lasted a mean 123 days (35-285), intubation 66 and 102 days, respectively, and decannulation took place a mean 187 days (43-449) after the operation in the successfully treated patients. In most infants the number of procedures under general anesthesia (laryngofissure, stent replacement, and laryngoscopies) was between 6 and 10.

Twenty-one of 24 infants could be decannulated or extubated after one operation. In one case (18) the therapy failed. This infant was later re-operated (laryngofissure with interpositioning of a costal cartilage graft), and subsequently decannulated. Two patients died during the treatment (Table II). The patients decannulation rate in the subgroup infants was 92%.

The mean follow-up period after decannulation of the 21 cured infants was 2 years and 11 months. One infant (9) initially had signs of hoarseness and aspiration of food, and later she had slight complaints of inspiratory stridor on exertion, which did not warrant laryngoscopy. The tracheotomy openings spontaneously closed after decannulation in most infants. In five cases a tracheocutaneous fistula had to be closed surgically.

In the **subgroup children** one child underwent an anterior laryngofissure, another an anterior laryngofissure with a castellated incision, and four a combined anterior and posterior laryngofissure. One child was intubated, five children were stented after surgery. The intubation lasted 48 days, and the stenting 90, 181, and 189 days, respectively, in the 4 successfully treated patients. The extubation or decannulation took place 48, 97, 258, and 343 days, respectively, after the operation. Between 3 and 12 procedures in general anesthesia were needed.

Four children could be decannulated or extubated. In the other two (29, 30) the therapy failed; they both had a grade 4 glottic and subglottic stenosis. The stent was removed after 350 and 230 days, respectively, when the airway was completely covered with epithelium without any granulations. One of these children (29) is

lost from follow-up, the other (30) has been re-operated recently (laryngofissure with interpositioning of a costal cartilage graft), and has now been decannulated (Table II).

The 4 successfully treated children were followed between almost 2 and 11 years. One child (28) had dysphonia probably as a result from scarring of the vocal cords, and a slight, permanent inspiratory stridor. A second child (26) had inspiratory stridor on exertion. The severity of the complaints did not warrant laryngoscopy.

Table III: Complications in the subgroups infants and children.
(The total numbers of patients in which the various complications occurred are given.)

	Infants	Children	Total
Surgery related:			
Wound infection	1	0	1
Subcutaneous emphysema	0	1	1
Pneumothorax	1	0	1
Related to the postoperative period:			
Granulations in larynx or trachea	13	4	17
Respiratory tract infection	13	4	17
Dysphagia, aspiration	8	0	8
Obstruction of tube	4	0	4
Accidental decannulation or extubation	2	0	2
Fatal cardiac arrest	2	0	2
Stent disconnection	1	0	1
No complications	4	0	4

COMPLICATIONS

The complications, summarized in table III, are either related to surgery or to the postoperative period. They range from frequently-occurring, relatively minor events, such as respiratory tract infections, to life-threatening events, such as accidental extubation or tube obstruction.

In the **subgroup infants** complications occurred in 20 of the 24 patients (Table III). Laryngeal or tracheal granulations were observed in 13 infants. These gradually disappearing granulations were the main factor responsible for prolongation of the stenting. More than half of the infants had at least one episode of respiratory tract infection, which always responded to antibiotics. Dysphagia and

aspiration, probably caused by the presence of the stent and the cannula, necessitated nasogastric tube feeding in eight infants. These symptoms, which were only observed in the infants, and not in the older children, disappeared only gradually after decannulation. Other complications were obstruction of the endotracheal or tracheotomy tube, accidental extubation or decannulation, and stent disconnection from the tracheotomy cannula. These potentially life-threatening events, only observed in four infants, could be adequately managed without cerebral or other damage. Two infants discharged to other institutions, both with a stent in place, died of cardiac arrest. One (12) had cardiac and other congenital anomalies, the other (2) was healthy except for the laryngeal stenosis. It was reported that neither inspection of the tracheotomy tube and the stent, nor autopsy revealed the cause of these fatal events.

In the **subgroup children** no life-threatening events were recorded. Frequent complications were again granulations and respiratory tract infections.

DISCUSSION

The reported group of patients could be divided into two subgroups (infants and children) that differed in number, in sex ratio, cause and severity of the stenosis, and in treatment results.

Most of these patients were infants, which may be explained by two reasons. First, it is particularly preterm infants, who are treated with prolonged intubation and ventilation, a treatment which may cause laryngeal stenosis [43]. Secondly, in young infants the subglottic diameter is proportionally small, and increases rapidly (Table Ia). Selecting the correct size of the endotracheal tube is therefore a critical procedure with the risk of intubation with too large a tube.

The majority of the patients were boys, and in the subgroup children male predominance was even more pronounced than in the infants. In both subgroups most stenoses occurred after prolonged intubation. In the infants the necessity of intubation was often related to prematurity.

Similar differences in age, number of patients per age group, and cause of the stenosis are found in the London series [81]. The patients in Cincinnati are more evenly distributed over the age groups [28]. Differences in grade of stenosis per age group are not reported by other centers, but may be an important factor in the outcome of treatment.

The complications in the subgroup infants appeared to be more serious and more frequent than those in the subgroup children. Life-threatening complications occurred only in infants. Two infants died under unclear circumstances after discharge to other hospitals, both had a stent connected to a tracheotomy cannula. In this respect it may be mentioned that the mortality rate of a tracheotomy in infants is reported to be from 0-5% [50]. Complications, including mortality, are related to younger age and longer duration of the tracheotomy [34,49,50]. The risks are probably further increased if a stent is connected to the cannula. The period of tracheotomy and stenting should therefore be as short as possible. Operation at an early age, and a short postoperative period until decannulation should be aimed for. The duration of tracheotomy in Rotterdam does not exceed those in other centers (Table IVc).

Serious dysphagia and aspiration, continuing for months after decannulation, occurred only in infants. This problem is probably related to long-term nasogastric tube feeding resulting in inefficient reflexes, and requires an intensive therapeutic training program.

The treatment results in the infants were better than those in the children of the reported series, both in terms of decannulation rate and of complaints after decannulation. The stenoses, however, were more serious in children than in infants (Fig. 3). Other centers report a relation between grade of stenosis and treatment result [27,76,81], but do not report a relation between age and treatment result, or between age and grade of stenosis [27,59,70,76,81].

It is interesting to compare the decannulation rate of laryngofissures without graft in Rotterdam (83% in the combined groups of infants and children) with the results of the same type of operation in Cincinnati (Table IVb). Cotton (Cincinnati) reports a 66% decannulation rate after 73 laryngofissures without a graft [27]. The stenoses in this group of patients, however, were more serious than those in Rotterdam: 11 grade 4 stenoses of 73 in Cincinnati, versus 3 of 30 in Rotterdam [27]. Another sizeable group of laryngofissures without graft is reported from London [8]. Unfortunately, separate decannulation rates of the operations with and without graft are not presented; the first operation decannulation rate is 63%.

The decannulation rate of our patients treated with laryngofissures without graft (83%) in comparison with those of other types of operations are summarized in table IVa and b. The decannulation rate of operations with a graft in Cincinnati is

Table IV: Laryngotracheal stenosis treatment in 6 centers. Rotterdam, Cincinnati, London, Zürich/München, Philadelphia, and Lausanne [8,26,27,59,70,76,81,82].

a. Types of treatment

Center	Rot	Cin	Lon	Z-M	Phi	Lau
Patients (n)	30	203	108	63	27	15
Types of operation						
graft	2	174	74	63	33	0
no graft	30	73	75	4	2	0
resection	0	0	0	0	0	15
other	0	4	0	0	0	0
Total	32	251	149	67	35	15

b. Decannulation rates

Center	Rot	Cin	Lon	Z-M	Phi	Lau
Decannulation rate of first operations (%)						
graft	-	78	?	-	-	-
no graft	83	66	?	-	-	-
combined	-	75	63	78	67	-
resection	-	-	-	-	-	93
Decannulation rate in patients (%) (after 1 or more operations)	90	92	83	84	78	93

c. Pre- and postoperative period

Center	Rot	Cin	Lon	Z-M	Phi	Lau
Time between tracheotomy and first operation (m)	17	?	?	?	?	?
Minimum age for operation (m)	9	12-18	24	18?	12?	3
Operations/patient	1.07	1.24	1.38	1.06	1.3	1
Stenting period (m)	4	1-6	1.5-6	1.5-2	1	0.5
Time between last operation and decannulation (m)	6	?	15	6	8	7
Mortality rate (%)	7	0-1	4	5	0	0

78%, the severity of the stenoses in that group of patients is comparable with that of the Rotterdam patients: 17 grade 4 stenoses of 174, versus 3 of 30 in Rotterdam [27]. The partial cricoid resection and laryngotracheal anastomosis in Lausanne has a very high first operation decannulation rate (93%) [76]. Though the number of patients is not large, and the indications are limited to subglottic and tracheal stenoses, the results are promising, especially in patients with a reported grade 4 stenosis. However, a grade 4 stenosis in the current Cotton classification implies obliteration and no vocal cords identifiable [27]. As such a high stenosis cannot be cured by cricoid resection, it seems likely that the old Cotton classification was used in Lausanne, and that the reported grade 4 stenoses are in fact grade 3 (subglottic obliterations) [28].

The length of the tracheotomy period is determined by the preoperative period, of which no figures are reported in literature, and the postoperative period. The latter period in Rotterdam does not exceed those of the other centers (Table IVc).

The number of major operations per patient in Rotterdam is low compared to other centers, the number of minor procedures (endoscopies, replacement of stents) are not reported by other centers.

Other important factors for assessing the long-term results are:

- growth of the larynx, especially the subglottic diameter;
- voice;
- respiratory function;
- growth of the patient.

These factors have not yet been subject of systematic research [30], but could add to an adequate evaluation of treatment strategies.

CONCLUSIONS

The method used in Rotterdam, laryngofissure without graft, compares favorably with methods used in other centers in terms of decannulation rate, duration of tracheotomy, minimum age for operation, and number of major operations. The longer duration of stenting, the repeated endoscopic procedures, and the observed higher mortality rate are disadvantages.

The presence of a stent in the larynx may contribute to dysfunction and damage to the glottis, formation of granulation tissue, and increased tracheotomy risks. This is why methods to reduce the stenting period have to be explored. A cartilage graft,

though gradually replaced by fibrous tissue [83], prevents retraction of scar tissue. Moreover, the perichondrium could provide a surface that is easily overgrown by regenerating mucosa, and therefore gives rise to less granulation formation than an anterior gap not closed by a graft.

Evaporation of granulation tissue by laser surgery could also positively influence wound healing.

Comparison of the results of various operation techniques, however, does not lead to unambiguous conclusions in this respect.

ECTOPIC CARTILAGE IN SUBGLOTTIC STENOSIS: HAMARTOMA OR REACTION TO TRAUMA?

ABSTRACT

In an experimental study in growing rabbits an endolaryngeal injury to the subglottis resulted in the development of a stenosis due to the formation of scar tissue containing ectopic cartilage. For comparison, biopsies taken from the subglottic stenosis in 8 children were studied histologically. In 6 cases ectopic cartilage was observed; all patients had a history of endotracheal intubation. In 3 children the diagnosis hamartoma was made. In the remaining 3 cases the formation of ectopic cartilage might have been a direct reaction to the endolaryngeal intubation. The observations suggest that the formation of ectopic cartilage in acquired subglottic stenosis is not always due to a developmental aberration such as a hamartoma.

INTRODUCTION

Endotracheal intubation can lead to the formation of a subglottic stenosis. Especially in infants and children the treatment of acquired stenosis is a difficult clinical problem. The causes for failure of therapy are still poorly understood. To investigate basic characteristics in the development of a subglottic stenosis an experimental study in growing rabbits was performed. The morphometric results illustrated specific patterns of disturbed development of the cricoid ring after trauma [5]. The histology showed the formation of ectopic cartilage in the subepithelial scar tissue and thickening of the cricoid cartilage at some sites [2,3].

From 1985 tot 1990 a laryngofissure was performed in 12 children for repair of chronic subglottic stenosis. In all patients repeated endotracheal intubations had been necessary for ventilatory support. In 8 cases a biopsy of the stenosis was taken. In this paper the histology of the biopsies will be compared with data obtained from the animal experiments.

MATERIALS AND METHODS

Twenty-one female New Zealand White rabbits were used. At the age of 4 weeks the subglottic airway was damaged circumferentially using a cutting burr as previously described [5]. In 10 animals the injury was restricted to the epithelium and subepithelial layer. In the remaining specimens the perichondrium and the

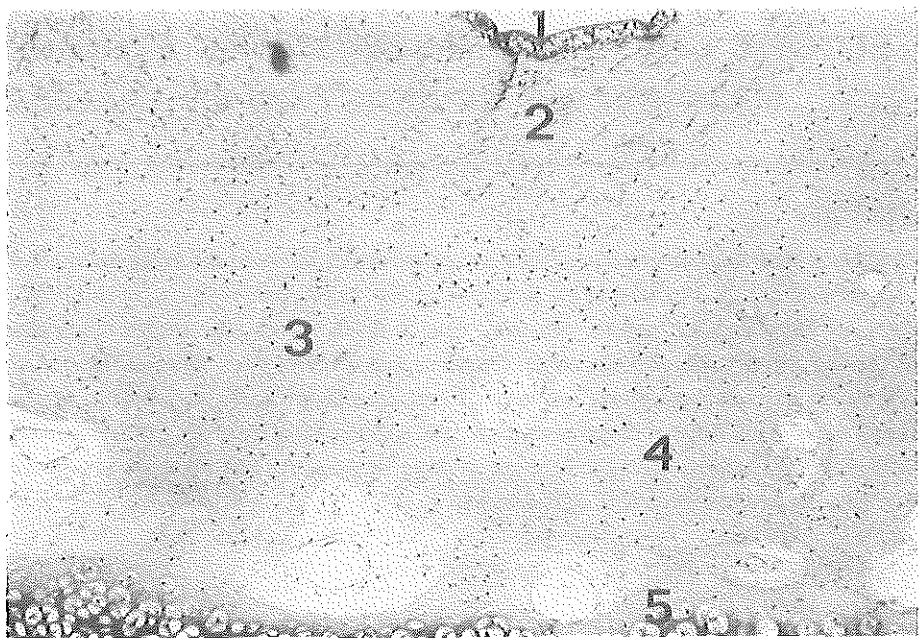

Figure 1: Experimental animal: part of the subglottis 20 weeks after endolaryngeal trauma: regenerated epithelium (1), fibrous tissue (2) in thickned subepithelial layer with areas of ectopic cartilage of the hyaline (3) and of the fibro-cartilaginous type (4) in contact with cricoid (5). (haematoxylin-azophloxin, magn. 25x).

innermost layer of the cricoid cartilage were damaged as well. At the adult stage of 24 weeks the animals were sacrificed. The subglottic part of the larynx was excised and processed histologically: 5μm transverse sections were prepared and stained with hematoxylin-azophloxin.

From 8 patients tissue obstructing the laryngeal airway was excised at different sites in the subglottic region (see Results). After histologic processing 5μm sections were obtained and stained with hematoxylin-azophloxin.

RESULTS

EXPERIMENTAL ANIMALS (N = 20)

A detailed histologic description has been reported previously [2,3]. The data will be summarized. The subglottic airway is lined with normal respiratory epithelium. Glands as well as the tunica elastica are missing. The regenerated subepithelial layer consists of fibrous scar tissue, fatty tissue and blood vessels. Regardless of

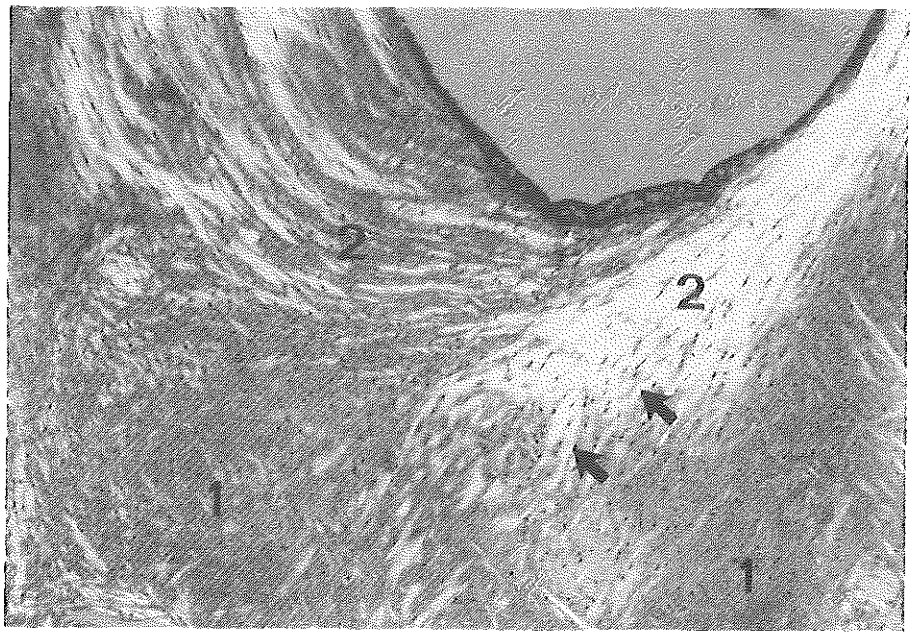

Figure 2: Experimental animal: collagen fibres (arrows) continue from ectopic cartilage of the fibrocartilaginous type (1) into fibrous tissue (2). (haematoxylin-azophloxin, birefringent light, magn. 40x).

the depth of the endolaryngeal trauma ectopic cartilage is present in all but one specimen (Fig. 1). The newly formed cartilage is distributed randomly in the scar tissue and resembles either a hyaline or a fibro-cartilaginous type (Fig. 2). The cricoid cartilage and perichondrium display a normal adult aspect.

PATIENTS (N = 8)

In 6 of 8 biopsies ectopic cartilage was present. The histology of this subgroup shows:

(1) D.R.: dense fibrous tissue with leucocyte infiltration and a fibro-cartilaginous type of cartilage (Fig. 3); polarization shows the collagen fibres to continue their course from the cartilage into the surrounding fibrous tissue without an intervening perichondrial layer (Fig. 4).

(2) A.T.: fibrous tissue with areas of a fibro-cartilaginous type of cartilage and secondarily formed bone.

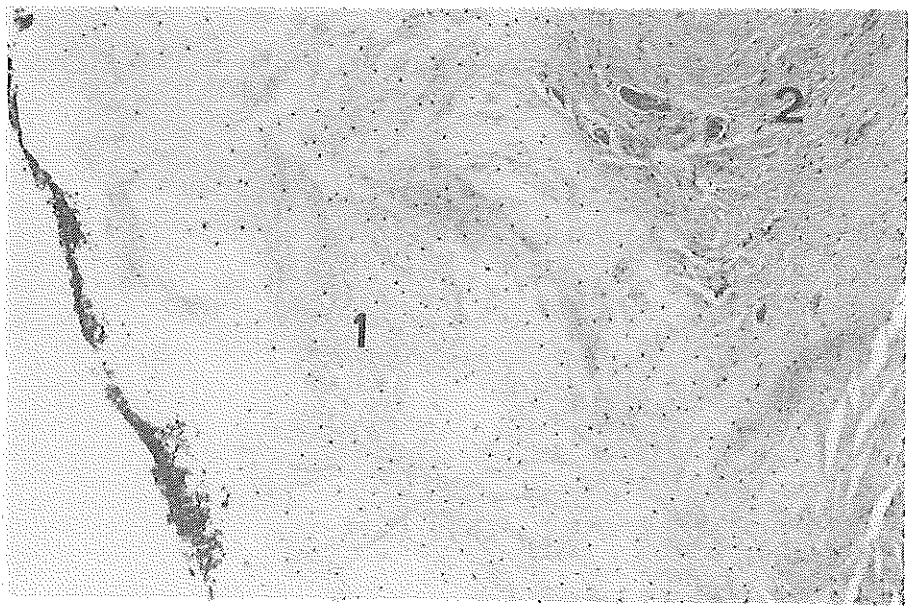

Figure 3: Human specimen (D.R.): dense fibrous tissue (2) and ectopic cartilage of the fibro-cartilaginous type (1). (haematoxylin-azophloxin, magn. 40x).

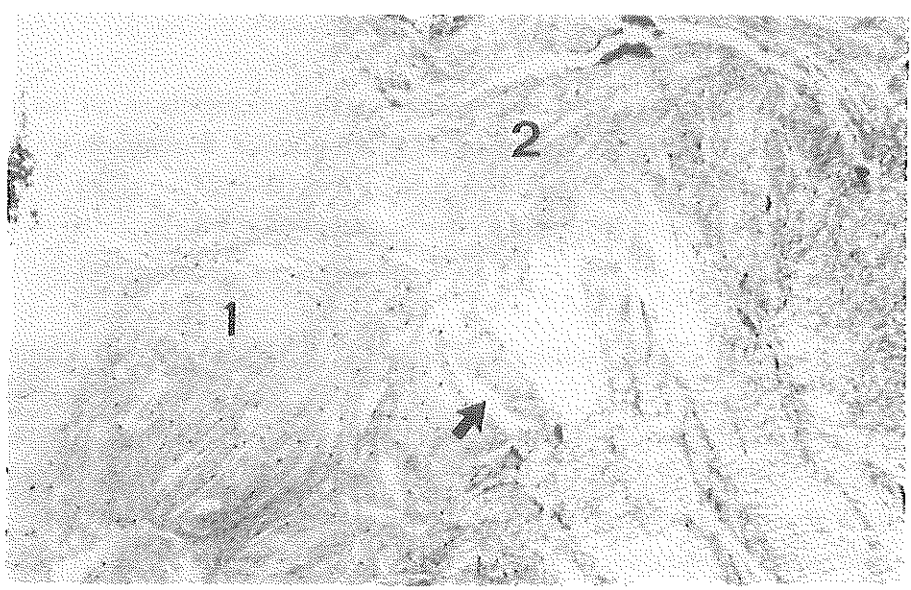

Figure 4: Human specimen (D.R.): fibro-cartilaginous type of ectopic cartilage (1); collagen fibres (arrow) continue into fibrous tissue (2) without perichondrial layer. (haematoxylin-azophloxin, birefringent light, magn. 40x).

(3) A.B.: fibrous tissue with a hyaline and fibrocartilaginous type of cartilage; also areas of fatty tissue and secondarily formed bone containing marrow (Fig. 5).
(4) G.K.: hyaline cartilage surrounded by fibrous tissue, orderly arranged muscle tissue and glands (Fig. 6).
(5) G.O.: fibrous tissue, a hyaline type of cartilage and muscle tissue; also some nervous tissue is present.
(6) J.P.: fibrous tissue with leucocyte infiltration and both a hyaline and fibrocartilaginous type of cartilage, muscle tissue, nerve trunks and glands.

DISCUSSION

The experimental study in growing rabbits showed the chondroneogenesis in the larynx to be the product of endolaryngeal trauma [2,3]. To date the formation of ectopic cartilage after injury has not been reported in cases of acquired subglottic stenosis in infants and children. However, 'thickening' of the cricoid has been observed macroscopically in pediatric cases of subglottic airway narrowing after endotracheal intubation [31,35,56].

In humans differentiation of granulation tissue into cartilage with secondary formation of bone is a well-known phenomenon in the process of wound repair in muscle tissue (myositis ossificans) [88]; the formation of ectopic cartilage and bone is a reaction to injury and should not be considered as a hamartoma. A hamartoma is defined as a developmental aberration in which the tissues of a particular part of the body are all present, but arranged randomly with excess of one or more of the components [101].

In 3 of the patients (D.R., A.T., A.B.) the cartilage was the only ectopic tissue amidst the fibrous tissue and could, therefore, not be classified as a hamartoma of the larynx. The fibrous matrix of the cartilage merges into the surrounding fibrous tissue indication the chondroneogenesis to occur within the scar formation (Fig. 4). In these patients the formation of ectopic cartilage might have been a direct reaction to the intubation injury;

The biopsies of 3 other patients (G.K., G.O., J.P.) contain cartilage apart from muscle tissue, nerves and glands. In two of these cases (G.K. and G.O.) the biopsies were taken from the lateral or posterior subglottic area, so the lateral cricoid arytenoid muscle might have been included in the biopsy of the first patient. In one patient (J.P.) the biopsy was taken from an obstructing mass based on the

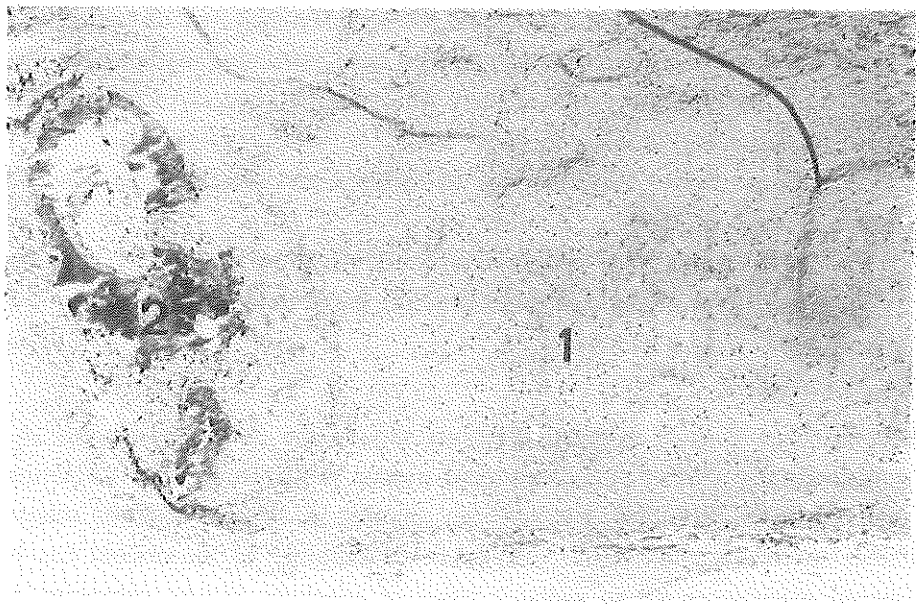

Figure 5: Human specimen (A.S.): ectopic cartilage (1) of the hyaline type with secondarily formed bone (2). (haematoxylin-azophloxin, magn. 100x).

Figure 6: Human specimen (G.K.): ectopic cartilage of the hyaline type (1), glands (2) and muscle tissue (3). (haematoxylin-azophloxin, magn. 40x).

laryngeal side of the epiglottis. In the normal anatomy no muscle is situated in this region, hence the diagnosis hamartoma seems to be justified.

The results of this comparative histologic study suggest that: (1) the formation of ectopic cartilage is not always caused by an aberrant development such as in a hamartoma; if ectopic cartilage is found in the larynx, suspicion of a post-traumatic lesion should be raised; and (2) the presence of ectopic cartilage in cases of acquired subglottic stenosis in infants and children might be a cause for therapy failure.

CHAPTER 9

SUMMARY AND CONCLUSIONS

The subject of this thesis, the study of some aspects of the diagnosis, treatment, and pathogenesis of airway obstruction in children, is introduced in the first chapter.

Bronchoscopy, either flexible or rigid, is indispensable in the diagnosis of airway obstruction. Function of the laryngeal entrance and mobility of the vocal cords are best observed in a spontaneously breathing patient with undisturbed anatomy. Chapter 2 deals with a method of flexible bronchoscopy aimed at achieving that goal in a patient under general anesthesia. Anesthetic gases and oxygen are administered through a breathing circuit and a mask. The patient is not intubated, and muscle relaxants have not been administered. The flexible bronchoscope is introduced into the breathing circuit through an air-tight valve, and subsequently passed through the nose until the larynx becomes visible. This method gives the endoscopist ample time to examine laryngeal function.

Structure and contents of the laryngobronchoscopy (LBS) data base are described in Chapter 3: sex and age of the patient, the used technique (rigid or rigid/flexible), the indication for LBS, diagnosis, procedures during LBS, and possible complications. The code table, which provides a code number to indications, diagnoses, procedures, and complications, is demonstrated. Most LBSs were performed in infants under one year of age. Frequent indications were inspiratory stridor, atelectasis, bronchopneumonia, and foreign body aspiration. The diagnoses made in patients with inspiratory stridor at LBS included laryngomalacia, intubation injury, subglottic laryngitis, subglottic hemangioma, tracheomalacia, and vocal cord paralysis. Diagnoses in patients with atelectasis made at LBS included pneumonia, bronchial stenosis, bronchial granulations, mucous plug, and tracheomalacia. Many procedures (a mean 1.2 per LBS) were performed: therapeutic ones such as bronchial lavage, intubation, foreign body extraction, dilation of a stenosis, or diagnostic ones such as bacterial sampling, biopsy, bronchoalveolar lavage (BAL). Some aspects of this series of bronchoscopies were compared with those of other reported series [48,52,84,85]. The data base provides an easy access to aspects of laryngobronchoscopy, which is of great clinical importance, and facilitates further research.

Chapter 4 describes all 25 complications, that occurred in 1332 rigid laryngobronchoscopies performed within eight years, a complication rate of 1.9%. The most frequent complications were hemorrhage, cardiac arrhythmia, and subglottic edema. Complications were statistically associated with the following factors: tetralogy of Fallot, biopsy or drainage of an abces, and foreign body extraction. Hemorrhage is a typical complication of biopsy and foreign body extraction. Patients with tetralogy of Fallot appeared to have an increased risk of cardiac arrhythmia during bronchoscopy. Manipulations during bronchoscopy or repeated introduction of a rigid bronchoscope may result in postoperative subglottic edema. Complication rates in other reported series of rigid laryngobronchoscopies range from 1.9 to 3.8% [48,52,85,69,74], and in flexible bronchoscopy from 2.2 to 8.0% [44,47,79,108]. Although the indications and the type of complications differ in rigid and flexible bronchoscopy, flexible bronchoscopy does not seem to be safer than rigid bronchoscopy.

In Chapter 5 a group of 115 children suspected of foreign body aspiration is described [19,66,71,98,103]. In 85 patients a foreign body was identified, in the others aspiration was excluded. LBS was decisive in the diagnosis. Choking, cough, stridor, abnormal auscultation, or abnormal chest X-rays were frequent and therefore rather sensitive symptoms in the group of children with foreign body aspiration. The same symptoms, however, were also frequently present in the group without a foreign body, so that the specificity appeared rather low. The abcense of specific symptoms of foreign body aspiration may easily result in missing the diagnosis. Laryngobronchoscopy is the best diagnostic tool, and the rigid type rather than the flexible LBS, the best therapeutic tool. In patients in our study group in whom the diagnosis was made immediately, the mean delay was only 6 days, but in other patients, in whom first another diagnosis was suggested the delay increased to a mean 55 days.

Early manifestations of post-intubation injury are (sub)mucosal edema, ulcerations, and granulations. Late manifestations are scar tissue formation and narrowing of the airway [36,37,43,95]. A method to treat the early manifestations of laryngeal injury in preterm infants is presented in Chapter 6. A group of 23 preterm infants with post-intubation laryngeal injury treated at the neonatal intensive care unit in the Sophia Children's Hospital was retrospectively studied. The patients were categorized according to the findings at laryngobronchoscopy: a. edema or

superficial lesions, b. ulcerations and edema, and c. granulations [15]. Treatment of the injury consisted of reintubation, which lasted a mean 17 days. The therapy was successful in 22 patients, and a failure in one. Outcome and duration varied with the category of the injury. In the group of infants with laryngeal granulations the intubation had to be continued longer, and the results were not as good as in the group of infants with edema or ulcerations. Therapeutic reintubation appears to be a valuable therapy that should precede the decision for surgery [55,29,25,33].

The late manifestation of prolonged intubation, cicatricial laryngeal stenosis, is surgically treated as a rule. A great variety of operations has been reported [25,8,27,76,59,70]. The treatment results in Rotterdam are described in Chapter 7 and compared with those of other centers. Thirty young patients with acquired laryngeal stenosis were treated by means of laryngofissure without a graft and long-term stenting with a silicone rubber stent or with intubation, in Rotterdam. Two subgroups could be distinguished: 24 infants and 6 children. Members of the group differed in age, cause and grade of the stenosis, and in treatment result. Twenty-one of the 24 infants, and 4 of the 6 children, were decannulated. Stenting was continued for about 4 months in the subgroup infants, and about 5 months in the subgroup children. In both groups decannulation was possible after a mean 6 months. Two patients died in other institutions, and in three patients failures occurred; two of them were succesfully reoperated (laryngofissure with a cartilage graft). The decannulation rate in the Rotterdam series is similar or better than that in other centers, and the number of reoperations is lower. It is impossible to compare the results of laryngofissures with and without graft, because separate figures have not been reported. The longer duration of stenting in Rotterdam is a disadvantage, which however did not result in a later decannulation moment. Mortality, which was high in Rotterdam, is theoretically related to the presence of the tracheotomy and the stent [49,50]. We recently started to perform laryngofissures with a cartilage graft, hoping to shorten the stenting period.

A comparative histological study of scar tissue produced by endolaryngeal trauma to the subglottis of growing rabbits, and of biopsies obtained from laryngeal stenoses in children, is described in Chapter 8. The animal scar tissue appeared to contain ectopic cartilage amidst fibrous tissue [2,3], which was also observed in 6 of the 8 human specimens. All biopsied children had a history of endolaryngeal intubation. In three the diagnosis hamartoma was made. In the other three the

formation of ectopic cartilage might have been a direct reaction to the endolaryngeal intubation. These observations suggest that the formation of ectopic cartilage in an acquired subglottic stenosis is not always due to a developmental aberration such as hamartoma [88,101].

CONCLUSIONS

Endoscopic examination of the pediatric airway is in experienced hands, safe, quick, and reliable. Many indications for bronchoscopy, such as airway obstruction, intubation or ventilation difficulties, or problems with a tracheostomy, arise in Neonatal and Pediatric Intensive Care Units (ICU). These intensive care units for their part are indispensable in the treatment of such disorders. Therefore children with serious airway pathology are usually managed in close cooperation with the otolaryngologist and the ICU-pediatrician in University Hospitals with pediatric ICUs. Pediatric Otolaryngology as a profession, particularly with respect to the airway, is in demand, and can only develop in such hospitals.

The clinical knowledge and experience can be gathered in data bases, study of which may help to deepen the understanding. The LBS data base appeared to be useful in demonstrating the association between complications and cardiac defects or administered drugs, thus isolating risk factors. The stenosis data base provided data about patients with post-intubation laryngeal injury, and laryngeal stenosis. Study of these data helped to find out which type of injury may be succesfully treated with reintubation, and indicated that age may be a factor in the outcome of surgical stenosis treatment.

Experimental work with laboratory animals and clinical studies are mutually dependent. Study of biological processes such as wound healing and laryngeal growth in experimental animals, provides the clinician with information that would otherwise be unattainable. On the other hand, observations in patients may initiate experimental studies. Close cooperation between clinic and laboratory is mandatory to understand the pathological processes that play a role in the formation of airway stenosis.

Children with serious airway obstruction should be treated in University Hospitals, where clinical skills and equipment are available, together with utilities for experimental research.

SAMENVATTING

De afdeling Keel-, Neus- en Oorheelkunde in een universitair kinderziekenhuis als het Sophia Kinderziekenhuis (SKZ) houdt zich voor een belangrijk deel van de tijd bezig met de diagnostiek en behandeling van luchtwegobstructie. Pasgeborenen en ook oudere kinderen met een inspiratoire stridor worden veelal verwezen naar een academisch centrum, waar de noodzakelijke diagnostiek en behandeling voorhanden is. Op de grote intensive care afdelingen voor pasgeborenen, voor pediatrische en voor chirurgische patiënten in het SKZ worden veel patiënten met ernstige luchtwegaandoeningen behandeld. De KNO-arts in het SKZ wordt geconfronteerd met aandoeningen als laryngomalacie, aangeboren larynxstenose, verworven larynxstenose na langdurige intubatie, problemen samenhangend met een tracheostoma, tumoren in de luchtwegen, obstructie op het niveau van de pharynx als onderdeel van een syndroom, en tracheomalacie. Bij de diagnostiek, en ook bij de behandeling van deze aandoeningen spelen de starre open, en de flexibele gesloten bronchoscopie een grote rol. Starre bronchoscopie is door de betere optische eigenschappen te verkiezen boven flexibele bronchoscopie bij de bestudering van anatomische afwijkingen. Een nadeel van de starre bronchoscopie is echter het optreden van vervorming van de luchtweg, en de noodzaak tot algehele anesthesie. Hierdoor wordt de diagnostiek van functionele aandoeningen zoals laryngomalacie, stembandverlamming en tracheomalacie bemoeilijkt. Indien dergelijke aandoeningen worden vermoed of dienen te worden uitgesloten moet ook flexibele bronchoscopie worden uitgevoerd. In hoofdstuk 2 wordt een methode van flexibele bronchoscopie beschreven bij een spontaan ademende patiënt onder algehele anesthesie. Deze methode maakt diagnostiek van functionele luchtwegaandoeningen mogelijk.

De afdeling Keel-, Neus- en Oorheelkunde in het SKZ verricht per jaar 200-300 bronchoscopieën. De indicaties, bevindingen en complicaties worden bewerkt en verzameld in een gegevensbestand. Een aantal gegevens van 1332 bronchoscopieën verricht bij 808 patiënten van 1982-1990 worden gepresenteerd in hoofdstuk 3. Meer dan de helft van de patiënten was ten tijde van de bronchoscopie jonger dan 1 jaar. De meest voorkomende indicaties waren inspiratoire stridor, atelectase, pneumonie, en aspiratie van een vreemd voorwerp. De meest gevonden oorzaken

van inspiratoire stridor waren laryngomalacie, intubatieletsel van de larynx, laryngitis subglottica, subglottisch hemangioom, tracheomalacie en stembandverlamming. Bij de bronchoscopieën verricht vanwege atelectase werd als oorzaak hiervan gevonden onder meer: bronchusstenose, granulaties in de bronchiaalboom, een afsluitende slijmplug en tracheomalacie. Een dergelijk gegevensbestand maakt een snel overzicht mogelijk, en bevordert de mogelijkheden tot retrospectief patiëntenonderzoek.

In deze groep van 1332 bronchoscopieën werd 25 maal een complicatie gezien, een percentage van 1,9%. In hoofdstuk 4 wordt een overzicht gegeven van de omstandigheden waaronder de complicaties voorkwamen. Intra-operatieve complicaties waren bloeding in de luchtwegen en hartritmestoornissen, postoperatief werd onder andere subglottisch oedeem gevonden. Statistisch onderzoek van de omstandigheden waaronder de complicaties voorkwamen toonde een relatie aan met tetralogie van Fallot, met biopsie of drainage van een abces, en met verwijderen van een geaspireerd vreemd lichaam. In de literatuur worden complicatiepercentages vermeld bij starre bronchoscopie van 1,9 tot 3,8%, en bij flexibele bronchoscopie van 2,2 tot 8%. De indicatiegebieden voor starre en flexibele bronchoscopie verschillen echter enigszins, evenals de aard van de complicaties.

Een groep van 115 kinderen, die een bronchoscopie ondergingen wegens aspiratie van een vreemd voorwerp, werd nader bestudeerd. De resultaten van dit onderzoek worden beschreven in hoofdstuk 5. Door bronchoscopie werd de diagnose aspiratie van een vreemd lichaam bij 85 kinderen bevestigd, en bij de overigen uitgesloten. Veel voorkomende symptomen bij aspiratie waren: zich verslikt hebben, hoest, stridor, afwijkende auscultatie, en afwijkingen bij röntgenonderzoek. Omdat deze symptomen echter ook vaak voorkwamen in de groep zonder aspiratie is de specificiteit ervan gering. Dit heeft tengevolge dat de diagnose gemakkelijk wordt gemist. Wanneer dat in ons onderzoek aanvankelijk het geval was, verstreken tussen de aspiratie en het verwijderen van het vreemd lichaam gemiddeld 55 dagen, tegen 6 dagen bij een direct goed gestelde diagnose.

Langdurige intubatie en beademing is een veelvuldig toegepaste behandelingswijze geworden op pediatrische intensive care afdelingen. Hierdoor kan larynxletsel optreden, bestaande uit oedeem, slijmvliesletsel, ulceraties of granulaties. Deze afwijkingen manifesteren zich na extubatie met tekenen van een luchtwegobstructie.

Nadat de diagnose door laryngoscopie is bevestigd kan de behandeling bestaan uit reïntubatie onder optimale omstandigheden, tracheotomie of anterior cricoid split. Een groep van 23 prematuur geboren kinderen van de neonatale intensive care afdeling met deze aandoening werd behandeld door middel van reintubatie. De resultaten van deze behandeling worden beschreven in hoofdstuk 6. De behandeling slaagde bij 22 van de 23 kinderen na een gemiddelde intubatieduur van 17 dagen. De neonaten bij wie granulaties waren ontstaan, werden gemiddeld langer geïntubeerd, en hadden minder goede lange termijn resultaten dan kinderen met oedeem, oppervlakkige lesies en ulceraties. Reïntubatie is een goede mogelijkheid tot behandeling van intubatieletsel bij prematuren, die vooraf dient te gaan aan chirurgische behandeling.

Een later optredende complicatie van langdurige intubatie is littekenvorming in de larynx, leidend tot een meestal subglottisch gelokaliseerde stenose. Deze aandoening wordt tegenwoordig algemeen operatief behandeld. In het SKZ bestaat de behandeling uit een laryngofissuur met of zonder een posterieure splijting van het cricoïdkraakbeen en plaatsen van een endolaryngeale stent van gesiliconeerd siliconrubber. Tot voor kort werd geen kraakbeenimplantaat gebruikt. In hoofdstuk 7 worden de resultaten van deze behandeling gerapporteerd. Van de 24 zeer jonge kinderen (de meeste ongeveer 1 jaar oud) konden 21 worden gedecanuleerd, 2 overleden, en bij 1 slaagde de behandeling aanvankelijk niet, maar na heroperatie wel. Van de 6 overige oudere kinderen (>6 jaar) konden 4 worden gedecanuleerd, 1 moest opnieuw worden geopereerd en is nu gedecanuleerd, bij de laatste is de behandeling niet gelukt. De stentduur was gemiddeld 4 maanden bij de jonge kinderen, en 5 maanden bij de ouderen. Decanulatie vond gemiddeld ongeveer 6 maanden na de operatie plaats. De resultaten van de Rotterdamse behandeling werden vergeleken met die van andere centra. De in Rotterdam langere stentduur leidt niet tot een later tijdstip van decanuleren, het decanulatiepercentage is hoog, en wordt bereikt met minder larynxoperaties. De mortaliteit was echter hoog, en mogelijk gerelateerd aan de aanwezigheid van een tracheotomie. Om de stentduur en de tracheotomieduur te verkorten wordt in het SKZ sinds enkele jaren meer gebruik gemaakt van kraakbeeninterpositie.

In hoofdstuk 8 wordt een vergelijkende histologische studie beschreven van biopten genomen van patiënten met een larynxstenose na langdurige intubatie, en van konijnen aan welke experimenteel endolaryngeaal letsel was toegebracht. In biopten

van zowel de konijnen als van de patiënten werden geïsoleerde velden kraakbeen omgeven door bindweefsel aangetroffen. Bij 3 van de 6 patiënten werd het ectopische kraakbeen geduid als hamartoom, omdat behalve kraakbeen ook ectopisch spier- en zenuwweefsel werd gezien. Bij de overige 3 kan het kraakbeen, analoog aan wat bij de konijnen werd gevonden, worden beschouwd als het produkt van chondroneogenesis na een endolaryngeaal trauma.

CONCLUSIES

Endoscopie van de luchtwegen is in ervaren handen een veilig en betrouwbaar onderzoek. De indicaties ervoor zijn dikwijls afkomstig van intensive care afdelingen, die op hun beurt vaak nodig zijn voor de behandeling van ernstige hogere luchtwegaandoeningen. Kinderen met dergelijke luchtwegpathologie worden in de meeste gevallen dan ook behandeld door een keel-, neus- en oorarts in nauwe samenwerking met intensive care artsen in universiteitsziekenhuizen. De pediatrische keel-, neus- en oorheelkunde, en vooral de laryngologie, zal zich daarom juist in universiteitsziekenhuizen met grote pediatrische intensive care afdelingen ontwikkelen.

De verworven klinische ervaring kan worden opgeslagen in gegevensbestanden. Twee bestanden werden gebruikt voor het patiëntenonderzoek: het bronchoscopiebestand en het stenosebestand. Beide bleken van groot belang voor het vergroten van de kennis en het begrip van luchtwegpathologie en de behandeling ervan.

Klinisch patiëntenonderzoek en experimenteel proefdierenonderzoek zijn wederzijds afhankelijk. Wondgenezing en groei kunnen goed worden bestudeerd in proefdieren. Het verworven begrip komt de behandeling van kinderen met een luchtwegstenose ten goede. Omgekeerd kunnen waarnemingen bij patiënten leiden tot gericht experimenteel onderzoek.

Kinderen met een ernstige luchtwegstenose worden daarom bij voorkeur behandeld in een universiteitsziekenhuis, waar ervaring met de behandeling van dergelijke pathologie aanwezig is, naast de mogelijkheid voor experimenteel onderzoek.

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CURRICULUM VITAE

Hans Hoeve, geboren in Leeuwarden (1946), studeerde van 1964 tot 1972 geneeskunde aan de Rijksuniversiteit in Groningen.
Gedurende de jaren 1973 en 1974 werkte hij op de afdelingen chirurgie en gynaecologie-verloskunde in ziekenhuizen in Heerenveen en Delfzijl, en volgde hij de Tropencursus in Amsterdam ter voorbereiding op het werk als arts in een ontwikkelingsland.
Van 1974 tot 1978 verbleef hij in Afrika; hij werkte in het Mulanje Mission Hospital in Malawi, waar hij zich bezig hield met verloskunde, kindergeneeskunde, de opleiding van vroedvrouwen, en nieuwbouw van het ziekenhuis.
Terug in Nederland begon hij, na een kort durend assistentschap (agnio) gynaecologie-verloskunde in Amsterdam, in 1979 de opleiding in de keel-, neus- en oorheelkunde bij Prof. Dr. E.H. Huizing in Rotterdam. De opleiding werd voltooid onder leiding van Prof. Dr. C.D.A. Verwoerd. In 1983 vond inschrijving in het specialistenregister plaats.
Sindsdien is hij als staflid verbonden aan de afdeling Keel-, Neus- en Oorheelkunde van het Academisch Ziekenhuis Rotterdam, en geeft hij leiding aan de subafdeling binnen het Sophia Kinderziekenhuis. Zijn belangrijkste interessegebieden zijn luchtwegproblemen met name larynxstenosen, en oorchirurgie bij kinderen. Daarnaast heeft hij zich voor de afdeling KNO bezig gehouden met de voorbereiding van de nieuwbouw en de verhuizing van het kinderziekenhuis naar de nieuwe lokatie bij het Dijkzigt ziekenhuis en de Medische faculteit.