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Short Trachea, a Hazard in Tracheal Intubation of Neonates and Infants: Syndromal Associations

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Short trachea results from reduction in number of tracheal cartilage rings to 15 or fewer from normal mean of 17 rings in infants. In a review of radiologic and pathologic data, the thoracic vertebral level of tracheal bifurcation as seen in anteroposterior chest radiographs of infants with congenital malformations, cardiovascular anomalies, and skeletal dysplasias, was compared with numbers of tracheal cartilage rings demonstrated in postmortem specimens. Increased frequency of short trachea was seen in patients with DiGeorge anomaly (77%), skeletal dysplasias (55%), brevicollis (57%), diaplacental rubella (40%), and patients with congenital heart disease who did not have DiGeorge anomaly (36%, with range 25–83% for different types, the highest, 83%, being interrupted aortic arch). Preintubation high kilovoltage chest radiographs to establish the level of tracheal bifurcation in patients with increased risk of short trachea can be helpful in avoiding bronchial intubation and its complications. Postintubation chest films to assure the level of the endotracheal tube tip should be considered for such patients. Growth in length of the trachea with age is accomplished both by increase in size of tracheal cartilage rings and interring membranes, and by increase in ring number. (Key words: Anesthesia: pediatric. Intubation, tracheal: complications. Lung: short trachea.)

BRONCHIAL INTUBATION is a hazard of tracheal intubation in infants.[1–6] Rosenberg and Rosenberg[6] listed seven syndromes in which patients are said to have brevicollis, which is clinically defined as shortness of the neck: Turner, Noonan, Klippel-Feil, Smith-Lemli-Opitz, Freeman-Sheldon (whistling face), Meckel, and hemifacial microsomia syndromes. It was not specified that such patients may also have disproportionately short tracheas.

That abnormally short trachea in brevicollis can be due to reduction in tracheal cartilage rings was first reported by Landing and Dixon,[7] and Sein *et al.*[8] reported short trachea in patients with the DiGeorge anomaly. Cohen[9] reported short trachea with high tracheal bifurcation, demonstrated radiographically in a patient with laryngotracheoesophageal cleft.

The objectives of this study were as follows: 1) to identify conditions in which short trachea may occur by reviewing radiologic and pathologic data on infants with syndromes where this association seemed likely; 2) to demonstrate radiographically the thoracic vertebral level of the tracheal bifurcation in such patients, and, when possible, to compare high tracheal bifurcation with tracheal cartilage ring counts of postmortem specimens; and 3) to identify conditions with which the occurrence of short trachea is associated with an increased risk of inadvertent bronchial intubation.

Materials and Methods

The thoracic vertebral level of tracheal bifurcation was recorded for all evaluable antemortem, anteroposterior (AP) chest films in the Karshner teaching film library of the Department of Radiology, Childrens Hospital of Los Angeles. All patients whose chest radiographs appeared to show a short trachea (tracheal bifurcation apparently above T4) fell into one of the categories, multiple congenital anomalies, genetic or chromosomal disorders, congenital cardiovascular anomalies or skeletal dysplasia syndromes. Evaluable chest radiographs in the radiologic file of the Gabriel C. Duque, Jr., Cardiac Registry of the Department of Pathology and Laboratory Medicine, Childrens Hospital of Los Angeles, were reviewed for all patients with cardiac anomalies considered possibly associated with short trachea, including DiGeorge anomaly, hypoplastic left heart complex with aortic valve stenosis or atresia, interrupted or atretic aortic arch, transposition of great arteries, Ivemark asplenia syndrome, polysplenia, tetralogy of Fallot, and for patients with other cardiac anomalies who had been found at autopsy to have short trachea. The determinations of thoracic vertebral level of the tracheal bifurcation were done by counting the ribs down to the 12th, identifying the corresponding vertebra, and counting the vertebrae back up to the tracheal bifurcation.

Cartilage ring counts were done for all 77 available intact tracheal specimens of patients autopsied at Childrens Hospital of Los Angeles, 38 formalin-fixed specimens, dissected, stained in .25 toluidine blue at pH_2, and cleared for tracheal cartilage ring count by the method used by Sein *et al.*[8] and 39 specimens that had been formalin-fixed but not stained. Tracheal ring numbers, including the first ring below the cricoid cartilage and the

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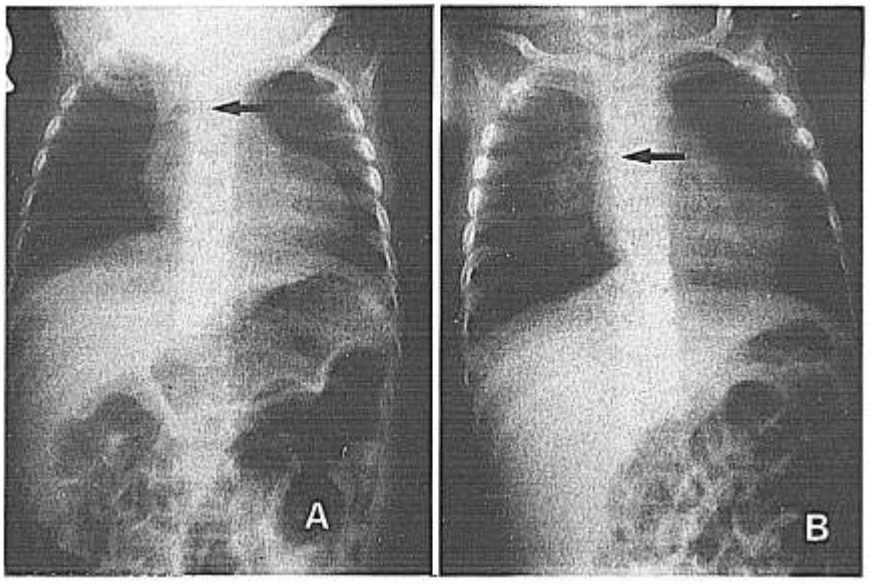

FIG. 1. Anteroposterior chest radiographs of a 3-month-old female with diaplacental rubella syndrome: *A.* The trachea is short, with level of tracheal bifurcation at T3. *B.* An endotracheal tube enters the right main bronchus. At autopsy this infant had 12 tracheal cartilage rings (mean normal 17).

last tracheal cartilage at the tracheal bifurcation, were determined by counting the rings on both right and left sides of the trachea and dividing the sum by 2.

The 49 patients who had 15.5 or more tracheal rings (the control series) ranged in age from 1 day to 24 yrs and included 12 preterm infants. The study group of 28 patients with 15 or fewer tracheal rings had the age range 1 day to 2.5 years; this group included eight preterm infants. Student's *t* test was performed for numbers of tracheal rings for the 49 control patients *versus* the study group of 28 patients with 15 or fewer rings.

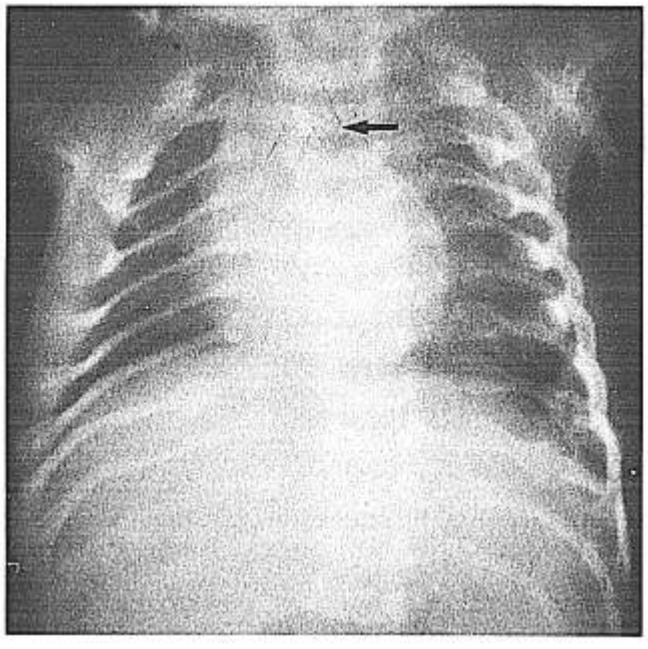

FIG. 2. Anteroposterior chest radiograph of a newborn male with osteogenesis imperfecta, showing tracheal bifurcation at the level of T2.

Results

Radiologic Aspects of Short Trachea

The AP chest radiographs shown in figures 1–3 demonstrate the high thoracic vertebral level of tracheal bifurcation of infants with congenitally short trachea (normal at T4 until age 2 yr, and at T5 thereafter), and figures 1B and 4 show intubation of the right main bronchus as a result of short trachea. The radiograph (fig. 4) of a 1-month-old infant with partial DiGeorge anomaly with right main bronchial intubation also demonstrates atelectasis of the contralateral lung, a frequent complication of bronchial intubation.[10,11] The air bronchogram (high KV technique, inspiratory phase) in figure 3, of a 3-yr-old with Ellis-van Creveld syndrome, shows short trachea with bifurcation at T3 (normal for age T4–5) and abnormal bronchial angles (right main bronchus [RMB] = 23° *vs.* 31 ± 6°; left main bronchus [LMB] = 25° *vs.* 51 ± 8°)[12].

Pathology

The control group for tracheal cartilage ring number reported by Sein *et al.*[8] was extended to 49 patients with 15.5 or more tracheal rings. The mean and SD of the number of tracheal rings in these controls is 17.01 ± 1.28. Examples of the stained and cleared tracheobronchial preparations used in the tracheal ring counts are seen in figures 5 (control) and 6, which demonstrates a short trachea with 14 tracheal rings in a patient with spondylothoracic dysplasia (Jarcho-Levin syndrome).

Table 1 presents data on disease categories of 28 patients with 15 or fewer tracheal rings. Comparison of the ring counts for the two groups *versus* the total series of tracheal specimens studied by Student's *t* test shows that the difference is significant ($P < .001$). *T* test for a com-

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bined series of patients (n = 11) with craniofacial skeletal abnormalities or with skeletal dysplasias also shows significant difference from the total values ($P < 0.01$) (table 1; fig. 7).

The diseases showing the highest incidence of short trachea in the data of this study were DiGeorge anomaly (DGA) (77%); skeletal dysplasias (55%) (including two patients with achondrogenesis type 2, with short and flattened (AP) tracheas and three patients with thanatophoric dysplasia, who had normal tracheal cartilage ring count); brevicollis, clinical (57%); interrupted aortic arch (IAA) (89%); hypoplastic left heart complex (HLH) (63%); patients with congenital heart disease who did not have DiGeorge anomaly (36%); and, congenital heart disease (CHD) excluding DiGeorge anomaly, interrupted aortic arch, and hypoplastic left heart complex (25%). Two younger patients with diaplacental rubella had short trachea, and three older patients with this disorder had normal tracheas, an overall incidence of short trachea of 40%.

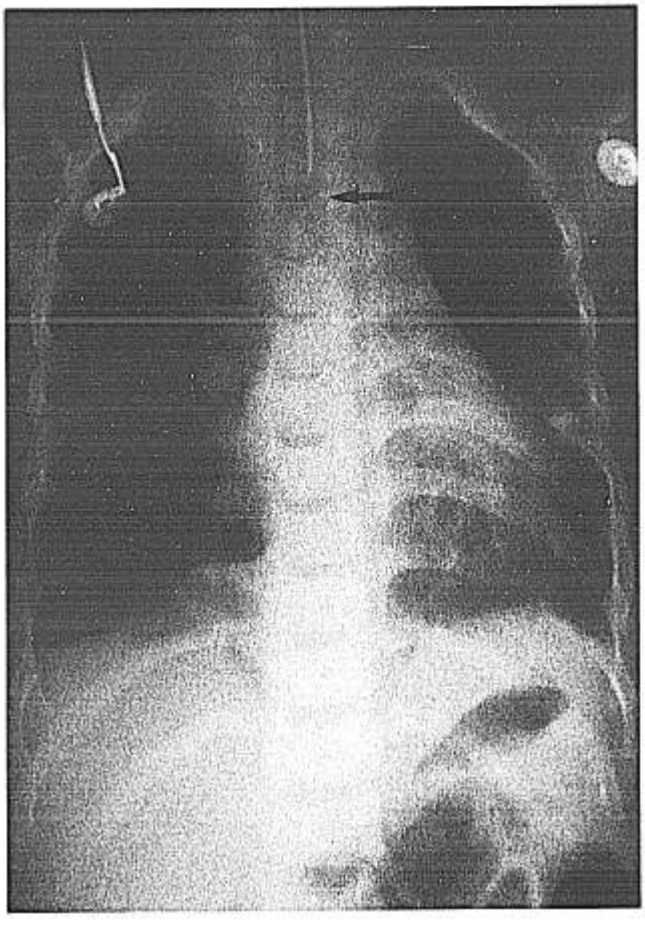

FIG. 3. Air bronchogram seen in a high KV (75) anteroposterior chest radiograph of a 3-yr-old male with Ellis-van Creveld syndrome. The tracheal bifurcation is at T3 during inspiration (normal for age T4–5) and the bronchial angles are abnormal (RMB, 23; normal 31 ± 6; LMB, 25; normal 51 ± 8).

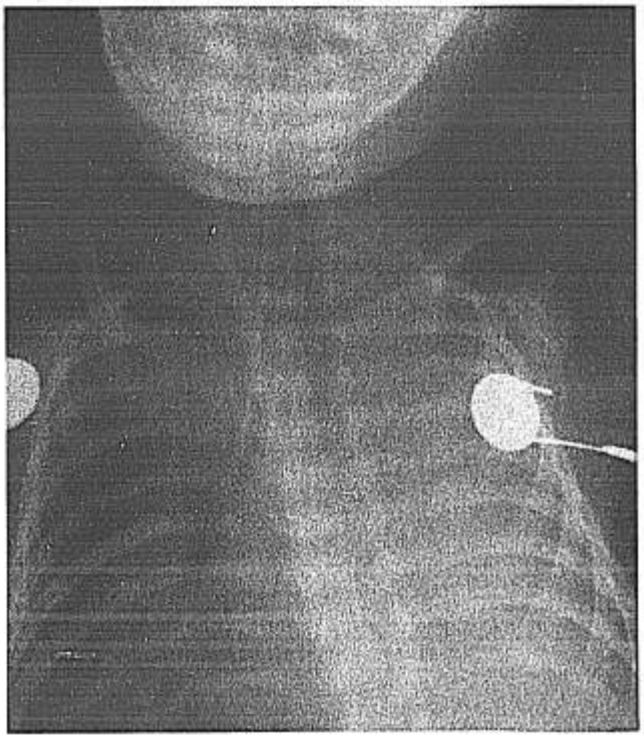

FIG. 4. Anteroposterior chest radiograph of a 1-month-old female with partial DiGeorge anomaly showing short trachea with bronchial intubation of the RMB and atelectasis of the left lung. At autopsy, this patient had 13 tracheal cartilage rings.

No difference in incidence of short trachea by sex was found.

Discussion

Intubation of a disproportionately short trachea can result in bronchial intubation, as shown in figures 1 and 4. Reported consequences of bronchial intubation include bronchial stenosis,[2] pulmonary interstitial emphysema, pneumomediastinum, pneumothorax,[13] and atelectasis of the contralateral lung,[10] but the association of short trachea and bronchial intubation has not hitherto been documented in the anesthesiology, otolaryngology, or neonatology literature.

Patients with the DiGeorge anomaly (third and fourth branchial pouch, fourth branchial arch deficiency) in this study showed a high incidence (77%) of short trachea. It has been proposed that short trachea in this condition is caused by abnormal blood flow to the region of the developing trachea.[8] That short trachea was seen in 11 of 14 patients with the DiGeorge anomaly (DGA) who had congenital heart disease, and in 83% of patients with interrupted aortic arch (IAA) who did not also have DGA may support this proposal, but the comparably high (55%) incidence seen in skeletal dysplasias presumably reflects a different mechanism. Because patients with DGA also

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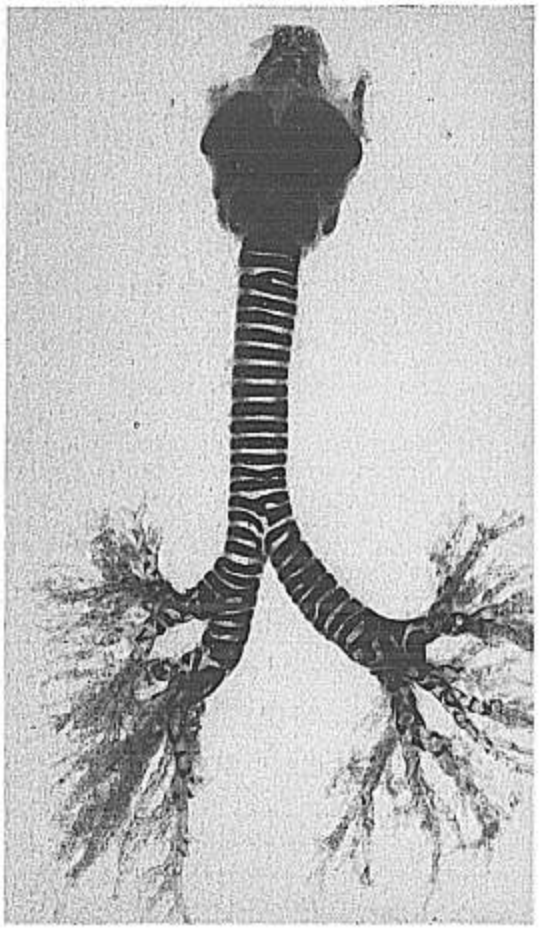

FIG. 5. Dissected, stained, and cleared tracheobronchial tree of a 1-month-old female control patient showing the normal number of tracheal cartilage rings (17) and normal bronchial anatomy.

have hypoplastic larynx,[14] an endotracheal tube of smaller diameter, as well as length, should perhaps be used for tracheal intubation of such patients.

Landing and Wells[15] have shown irregularly fused tracheal cartilages in a patient with the Ellis-van Creveld syndrome (chondroectodermal dysplasia); this lesion (tracheal splint) prevents tracheal lengthening during inspiration. Figure 3, a chest radiograph of a 3-yr-old patient with Ellis-van Creveld syndrome, specifically taken during inspiration, shows that the tracheal bifurcation descended only to the level of the third thoracic vertebra (normal for age T4–5) and also shows abnormally low angles of both main bronchi, this latter finding possibly a regular feature of Ellis-van Creveld syndrome. The ultimate tracheal splint was reported by Shimada and Misugi,[16] who showed that two patients with craniosynostosis, cloverleaf skull deformity and other skeletal anomalies had solid cartilaginous tracheal walls.

Jones and Pelton[17] listed 144 malformation complexes and genetic and chromosomal diseases with their anesthetic implications. Of these, 32 (22%) were said to have "difficult airways," "difficulty in intubating" or both, with the difficulty apparently attributed to craniofacial or vertebral anomalies, including brevicollis. Although difficulty in intubating the trachea of patients with brevicollis results in part from the more anterior position of the larynx and reduced mobility of the head, the present study shows that these patients also have an increased (57%) risk of short trachea. The tracheobronchial tree seen in figure 6, which shows a reduced number of tracheal rings and tracheal deviation by a right aortic arch, is from a 7.5-month-old patient with Jarcho-Levin syndrome (spondylothoracic dysplasia), a condition often confused with Klippel-Feil syndrome,[18] in which brevicollis is also present.

Normal tracheal growth with age results not only in increased cartilage ring size and interring distance but also in increased cartilage ring number. The perforated

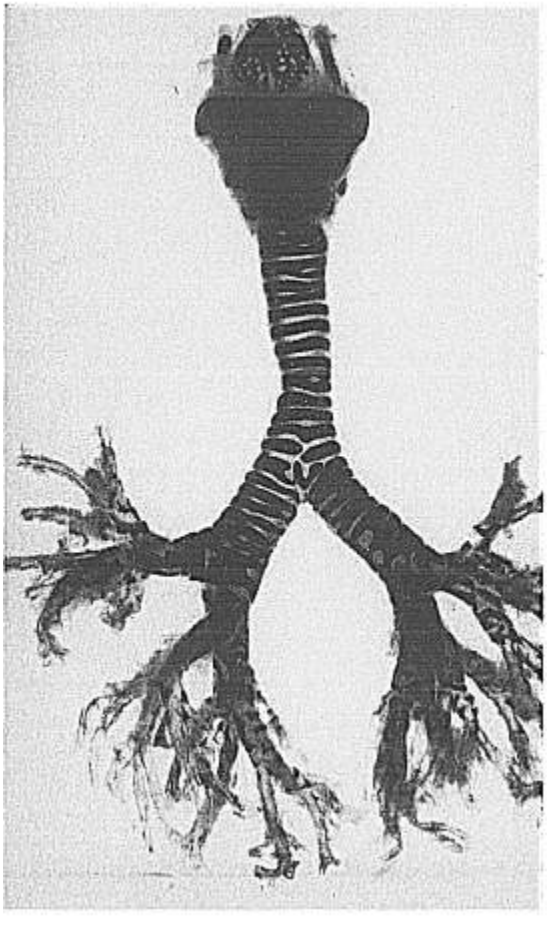

FIG. 6. Dissected, stained, and cleared tracheobronchial tree of a 7.5-month-old girl with Jarcho-Levin syndrome (spondylothoracic dysplasia). In addition to short trachea (14 rings), this patient had a right aortic arch, relatively long RMB (6 rings; normal 3–5), short LMB (7 rings; normal 7–9), and very short bronchus intermedius. The leftward deviation of the lower trachea is produced by the right aortic arch.

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TABLE 1. Summary of Radiologic and Pathologic Data on Incidence of Short Trachea in Patients with Various Syndromes*

	Tracheal Cartilage Rings 15 or Less†		Apparent Level of Tracheal Bifurcation Above T4‡		Total Patients with Short Trachea in Category§	
	#	%	#	%	#	%
Categories with Congenital Cardiovascular Disease						
DiGeorge anomaly	6/7	86	6/9	67	11/14	77
Interrupted aortic arch	7/8	89	1/1	100	8/9	89
With DiGeorge anomaly	3/3	100	—	—	3/3	100
Without DiGeorge anomaly	4/5	80	1/1	100	5/6	83
Congenital cardiovascular disease other than with DiGeorge anomaly¶	8/25	32	4/8	50	12/33	36
Hypoplastic left heart complex with aortic valve stenosis or atresia	2/5	40	3/3	100	5/8	63
Congenital cardiovascular disease with none of interrupted aortic arch, aortic valve stenosis, atresia, or DiGeorge anomaly¶	6/24	25	—	—	6/24	25
Brevicollis, clinical	8/14	57	—	—	8/14	57
Klippel-Feil syndrome	1/1	100	1/1	100	1/1	100
Skeletal dysplasia, including	4/8	50	6/9	67	10/17	59
Ellis-van Creveld syndrome** (4)						
Jeune syndrome (2)						
Osteogenesis imperfecta (3)						
Achondrogenesis type 2 (2)						
Thanatophoric dysplasia (3)†						
Kniest syndrome (1)						
Spur-limbed dwarfism (1)[23]						
Mesomelic dwarfism (1)[15]						
Diaplacental rubella						
Crown-heal length > 80 cm	0/3	0	—	—	0/3	0
Crown-heal length < 80 cm	2/2	100	2/2	100	4/4	100

* Some patients had more than one disorder (lesion), and appear in two or more categories, *e.g.*, congenital cardiovascular disease with diaplacental rubella.

† Patients in whom pathologic material was available.

‡ Patients in whom x-rays were available.

§ Represents sum of patients represented by pathologic or radiologic data except for patients with DiGeorge anomaly, Klippel-Feil syndrome, and Jarcho-Levin syndrome wherein both x-ray and autopsy specimens were available for several patients.

¶ Categories of congenital cardiovascular disease that have not, in the material of this study, contained a patient demonstrated to have short trachea, include: Ivemark asplenia syndrome not associated with Goldenhar syndrome[24] (five patients); 0-anisosplenia[25] (one patient); pentology of Cantrell (one patient); "short pancreas syndrome"[26] (one patient); tetralogy of Fallot not associated with DiGeorge anomaly (four patients); Down syndrome not associated with interrupted aortic arch (four patients); 13 trisomy (one patient); chromosome 18-q deletion (one patient); septal defects other than components of the categories listed above (three patients). Other specific syndromes not found in this study to have short trachea as a feature include 18 trisomy (one patient) and congenital muscular dystrophy (one patient). Specific syndromes not included in the categories of this table for which patients with short trachea were identified in this study include DeLange syndrome (one patient), congenital neuroblastoma (one patient), hypertrophic cardiomyopathy consistent with IHSS, ? chromosome translocation (one patient).

** Jarcho-Levin syndrome is now considered different from Klippel-Feil syndrome.[18] Both cause brevicollis.

†† In addition to short trachea and abnormal bronchial angles, found in half the patients with Ellis-van Creveld syndrome in this study, patients with Ellis-van Creveld syndrome also have tracheal splint.[15] Note that because of their abnormal tracheal cartilage anatomy,[15] tracheal ring counts cannot be done on tracheal specimens of patients with that disorder.

‡‡ Note that patients with thanatophoric dysplasia have a normal tracheal ring count and tracheal length. If thanatophoric dysplasia is removed from the skeletal dysplasia category, the incidence of short trachea in the other skeletal dysplasias we studied is 11/14 (79%).

and partially divided tracheal rings in the stained preparations seen in figures 8 and 9 may reflect the mechanism by which new ring formation is accomplished.

Calculations for endotracheal tube placement in the midtracheal (safe zone) area for orotracheal intubation were reported by Loew and Thiebeault[19] using the equation endotracheal tube length (cm) = 0.16 body length (cm). Coldiron[20] reported a similar calculation for nasotracheal tube length (cm) = 0.21 body length (cm). Use of these factors should position an endotracheal tube tip in the midtrachea of a patient with normal ratio of tracheal length to body length. However, use of these ratios for a patient with one of the conditions listed above, associated with significant incidence of disproportionately short trachea, could lead to bronchial intubation. The results of application of these ratios to infants with skeletal dysplasias or other conditions producing abnormality of the ratios of the body segments—head, trunk, and extremities—to

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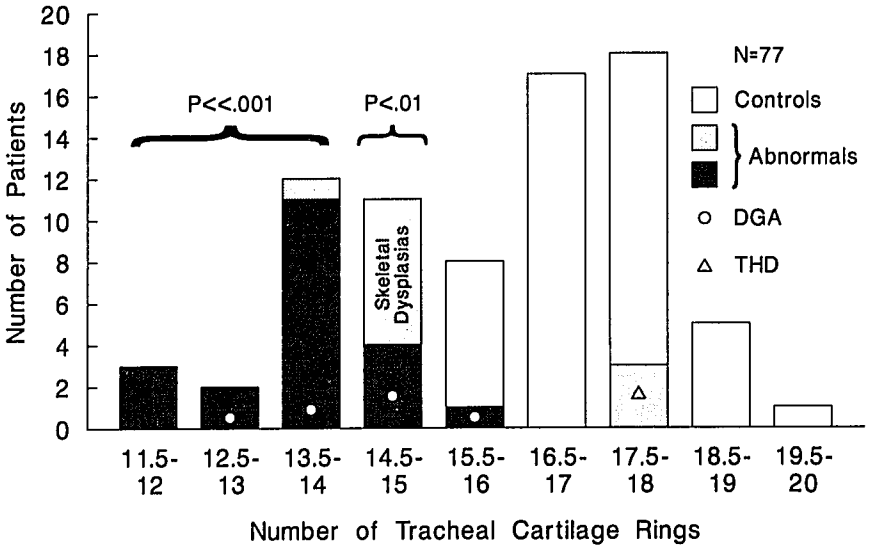

FIG. 7. Distribution of number of tracheal cartilage rings for control patients and for patients with 15 or fewer tracheal rings. Student's *t* test shows that the differences are statistically significant. DGA = DiGeorge anomaly; THD = thanatophoric dysplasia.

each other will, in principle, depend on the type of body segment disproportion produced by the disorder present. For example, relative shortness of extremities compared to trunk could by reducing the predicted endotracheal tube length, provide a margin of safety for patients with such disorders who have a short trachea. Among the many skeletal dysplasias that produce relatively greater shortening of extremities compared to trunk are achondrogenesis types 1 and 2; hypochondrogenesis; dyssegmental dysplasia; atelosteogenesis; fibrochondrogenesis; the short rib–polydactyly syndromes (type 1, Saldino-Noonan syndrome; type 2, Majewski syndrome; type 3, Verma-Naumoff syndrome); Beemer syndrome; thanatophoric dysplasia; achondroplasia; rhizomelic chondrodysplasia punctata; Elejalde syndrome; and the lethal form of osteogenesis imperfecta.[21] Conversely, the formulas given above could overestimate the proper endotracheal tube length for patients with disproportionately short trunks or thoraces, as spondyloepiphyseal dysplasia congenita, Kniest syndrome, Jarcho-Levin syndrome, and perhaps Morquio syndrome. The same consideration could apply to conditions with relative enlargement of the head, as achondroplasia, thanatophoric dysplasia, other "cloverleaf skull," syndromes and other causes of hydrocephalus.

Bednarek and Kuhns[22] have described a method of avoiding introduction of an excessively long endotracheal

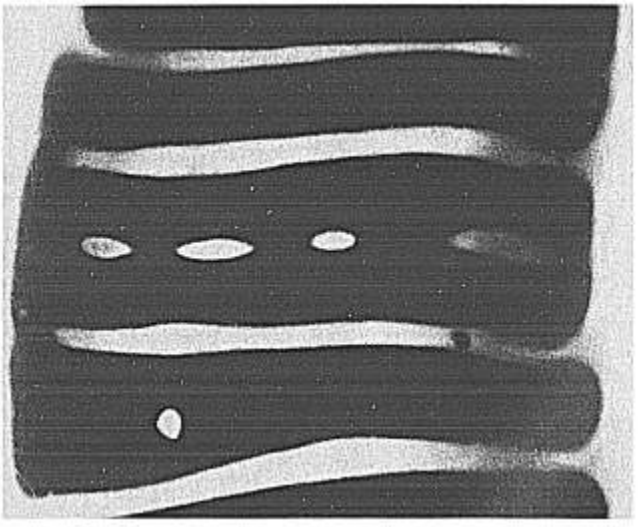

FIG. 8. Perforated and partially divided midtracheal cartilage rings of a 2.5-yr-old girl with Klippel-Feil syndrome, demonstrating the possible mechanism of new ring formation most frequently seen in the first and second tracheal rings. This patient had 14 tracheal cartilage rings. (Anterior view of trachea, toluidine blue stain at *p*H 2; original magnification ×7.)

FIG. 9. Lateral fusion of tracheal cartilage rings, possibly a result of incomplete or delayed ring separation (tracheal splint). (Same patient as figure 8, anterior view, toluidine blue stain at *p*H 2; original magnification ×7.)

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tube by having a second person palpate the trachea at the jugular (suprasternal) notch during the insertion of the tube. If the trachea is slightly compressed, the palpator can feel the tip of the tube as it is inserted. However, this method also may not prevent bronchial intubation of infants with abnormally high tracheal bifurcation.

Recognition of short trachea as an entity, and of conditions in which it may occur in neonates and infants, can be helpful for avoidance of bronchial intubation and its complications. That a patient at risk has short trachea can be demonstrated before intubation with appropriate radiologic technique (high kV). Such radiologic technique, or attention to the length of endotracheal tube inserted if the tube used has centimeter marks, can be useful also for postintubation monitoring for level of endotracheal tube placement.

One must bear in mind that the apparent vertebral level of the tracheal bifurcation in chest radiographs can be influenced by the angle of the x-ray beam, by the position of the infant (lordotic *vs.* antilordotic), and by the depth of inspiration, and also that the method described for assessing the carinal level should not be applied to a radiograph that shows tracheal buckling. Despite these reservations, it would appear prudent to consider an infant whose chest radiograph shows a higher than normal carinal level as having short trachea until proven otherwise, especially if the patient could have one of the syndromes described above with significant incidence of short trachea.

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