

TRACHEAL STENOSIS IN INFANCY

BY

JOHN BORRIE

From the Department of Surgery, University of Otago Medical School, and Southern Thoracic Surgical Unit, Dunedin Hospital, Dunedin, New Zealand

(RECEIVED FOR PUBLICATION OCTOBER 26, 1959)

Little has been written on the management of post-tracheostomy tracheal strictures in infants. The long and difficult management recently encountered, therefore, and its ultimate success, prompt this case report.

CASE REPORT

INTRALARYNGEAL FOREIGN BODY.—On the afternoon of Saturday, December 10, 1955, Stephen, a boy aged 14 months, was playing on the kitchen floor when his mother suddenly noticed that his breathing had become wheezy. His medical practitioner diagnosed a foreign body in the air passages and he was brought to Dunedin Hospital for emergency treatment. Immediate bronchoscopy under general anaesthesia showed a small piece of bone ($\frac{1}{2}$ cm. \times $\frac{1}{2}$ cm. \times $\frac{1}{8}$ cm.) wedged firmly between the vocal cords. It was readily removed with peanut grasping forceps.

OEDEMA OF GLOTTIS AND TRACHEOSTOMY.—Eighteen hours later, on December 11, he was discharged home. He was apparently well throughout the following day, but on the evening of Tuesday, December 13, he was readmitted seriously ill with acute oedema of the glottis accompanied by deep indrawing of the chest wall with every breath. Under local anaesthesia a tracheostomy was performed with immediate relief.

ENDOBONCHIAL CASTS AND TRACHEAL STRICTURE.—Two days later, on December 15, he again suddenly developed acute respiratory obstruction. Emergency bronchoscopy through the tracheostoma revealed plugs of caked bronchial secretions forming bronchial casts and blocking both the right and left main bronchi up to the level of the carina. They were removed with fine grasping forceps. Thereafter he was nursed in a "croupette" in an atmosphere of nebulized detergent (alevaire) and fine water vapour.

Despite this treatment, because of continued caking of endobronchial secretion, he caused the utmost concern, and between December 15 and 29 required 36 life-saving bronchoscopies. Each was a real emergency, and was precipitated as further plugs of inspissated secretions continued to form and block the bronchi (Table I).

On December 17, in the belief that the tracheostomy tube itself was aggravating this caking of bronchial

secretions, a small portion of the anterior surface of two of the tracheal rings was excised through the tracheostomy opening and the tube temporarily removed for 12 hours. Nevertheless, it was obvious from recurrent indrawing of the chest that the tracheostomy tube was still required. It was re-inserted and used continuously until finally removed 17 months later.

Progress was slow. The oedema of the larynx persisted until the beginning of January, 1956, when a smaller tracheostomy tube was used in the hope that he could then breathe through the larynx in the normal manner. Although the glottic oedema had subsided by January 6, none the less he could not breathe if the tracheostomy tube was removed. This was confirmed by a peroral bronchoscopy which revealed a tight tracheal stricture at the point of entry of the tracheostomy tube, approximately half an inch below the glottis. After dilatation, although a bronchoscope could be passed through this region, the moment it was withdrawn the anterior and posterior tracheal walls became apposed, totally occluding the airway. The posterior wall also appeared to be kinked forward on the anterior wall by contraction of fibrous tissue around the tracheostoma (Fig. 1a).

At this stage the possibility of excising the stricture completely and re-anastomosing the trachea was investigated experimentally in lambs, but the results made it clear that no such procedure could be applied to infants with their problem of a tiny trachea and the growth of adolescence still to come. After further assessment it was planned to (1) create a lower tracheostomy, (2) insert a skin graft to re-epithelialize the trachea at the site of entry of the first tracheostomy tube, and (3) close this first tracheostoma by suture.

CLOSURE OF UPPER TRACHEOSTOMY AND CREATION OF A LOWER ONE.—Operation was therefore performed on March 1, with the aid of my colleague, Mr. S. Horowitz. The operative details were as follows:

The baby was anaesthetized with open ether. Peroral bronchoscopy was performed and the stricture inspected. It lay as before just below the cricoid cartilage and just above the tracheostomy tube. This tube was removed, the stricture dilated and the bronchoscope passed onwards. The bronchoscope was next removed, the child intubated through the

TABLE I

SUMMARY OF PERTRACHEOSTOMY BRONCHOSCOPIES
REQUIRED FOR FORTNIGHT DECEMBER 15-29, 1955

Date	Bronchoscopy	Bronchoscopy No.
13/12/55	Admitted, acute oedema of glottis, in croupette 8.00 p.m., tracheostomy, local anaesthetic, routine tracheostomy treatment	
15 12 55	Respiratory distress + + +, blue and obstructed, as on all subsequent occasions when emergency bronchoscopy had to be performed	
	11.00 a.m. Bronchoscopy	1
	8.00 p.m. ..	2
16 12 55	6.30 a.m. ..	3
	3.00 p.m. ..	4
	8.00 ..	5
	10.30 ..	6
17 12 55	8.30 a.m. ..	7
	7.00 p.m. ..	8
	11.00 ..	9
18/12/55	5.45 a.m. ..	10
	2.00 p.m. ..	11
	6.30 ..	12
	10.45 ..	13
19 12 55	5.30 a.m. ..	14
	8.30 ..	15
	11.00 ..	16
	3.00 p.m. ..	17
	6.00 ..	18
20/12/55	10.30 a.m. ..	19
	5.15 p.m. ..	20
	Midnight ..	21
21, 12 55	5.30 a.m. ..	22
	5.55 ..	23
	5.30 p.m. ..	24
	8.00 ..	25
	11.00 ..	26
22 12/55	2.45 ..	27
	6.45 ..	28
23/12/55	5.30 a.m. ..	29
	4.45 p.m. ..	30
	Midnight ..	31
24 12/55	3.00 p.m. ..	32
	10.30 ..	33
25 12 55	5.00 ..	34
	11.00 ..	35
28/12/55	9.00 a.m. ..	36
29/12/55	Laryngoscopy—still oedema of glottis	

larynx and stricture with a No. 2 Magill tube, and ether anaesthesia continued.

A vertical incision was made from the level of the upper border of the thyroid cartilage to the suprasternal notch. The skin around the tracheostoma was excised, the normal pretracheal muscles isolated, the isthmus of the thyroid gland divided, and the cervical trachea exposed. The existing tracheostoma was further inspected and surrounding granulation tissue removed.

In order to establish a permanent airway below the narrowed area a lower tracheostomy was now performed, the endotracheal tube withdrawn a little, and a No. 2 tracheostomy tube inserted. A sterile anaesthetic tube was thereafter connected to the tracheostomy tube and anaesthesia continued.

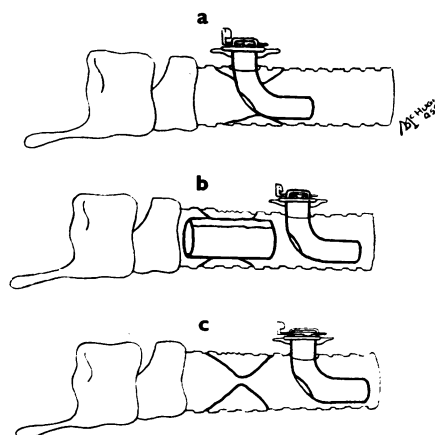


FIG. 1.—(a) The first tracheostomy tube and stricture at that level. (b) The second, lower tracheostomy tube in place together with a rubber prosthesis holding a skin graft in place. (c) This shows the problem to be treated once the upper tracheostomy had been closed.

The original endotracheal tube, which was still in position, was divided in two through the upper, original tracheostoma, and the upper, proximal portion of the tube removed through the mouth. The lower, distal portion was next brought out through the first tracheostoma; it formed a snug fit in the trachea, and was reinserted with the bevelled end pointing up towards the larynx. A split skin graft, 1 cm. square, taken from the chest wall, was then inserted through the first tracheostoma, and, using arterial suture hooks, spread out over the rubber tube which acted as a prosthesis. The graft was sewn on to the inside of the trachea with stay sutures placed at both upper angles. These stay sutures were then passed out through the pretracheal muscles and on to the skin. In order to anchor the rubber prosthesis a fine nylon suture was next passed through the skin of the neck, the trachea, the rubber tube, and out again (Fig. 1b). At this stage of the operation endotracheal suction was required, and accomplished by temporarily disconnecting the anaesthetic tube from off the low tracheostomy tube, and sucking with a fine Jaques catheter.

In closing the wound the pretracheal muscle layers were approximated with interrupted sutures, taking the pretracheal fascia to obliterate dead space. The skin was approximated with interrupted nylon sutures. Thereafter the three sutures holding the graft were tied. The new tracheostomy tube was sutured in position, and the wound covered with tulle gras.

PROGRESS.—The baby rapidly recovered from the operation.

Inspection Bronchoscopy showing Persistence of Stricture.—Twenty days after operation, under general anaesthesia, the skin sutures were removed, and bronchoscopy performed. The larynx was normal. The small piece of endotracheal tube lying as a prosthesis in the upper trachea and measuring

22 mm. long, was removed through the larynx with grasping forceps. The trachea at the first tracheostoma level appeared granular. When the tracheostomy tube was removed below, the patient could not tolerate occlusion of the tracheostoma, because at the strictured area the tracheal walls again collapsed and did not allow of normal breathing (Fig. 1c).

It was felt that a prosthesis would be required in the trachea for at least another six months. A piece of No. 2 Magill endotracheal tube measuring 3.5 cm. in length was therefore re-inserted through the larynx to lie above the tracheostomy tube, but without any stay suture.

On March 21, at 11.00 p.m., while the tracheostomy tube was being changed, the patient became breathless, and the tracheostomy tube could not be re-inserted.

Inspection bronchoscopy through the tracheostoma showed that the prosthesis had slipped down to the carina. It was removed with fine grasping forceps, and replaced as before. Four days later this episode was repeated. A suture passed transversely through the skin, trachea, and prosthesis did not appear to be a satisfactory solution.

As a result, on March 28, under a general anaesthetic the trachea was again inspected. During the week, granulation tissue had disappeared, but the narrowing remained. It responded to dilatation with the bronchoscope. With the bronchoscope in position the tracheostomy tube was removed and an attempt was made to insert a special flexible metal prosthesis, which had been made from an old flexible "fish-tail" tracheostomy tube. On it had been soldered a flange which could sit snugly above the normal tracheostomy tube and prevent the flexible tube (which was aimed to be placed through the stricture area from below) from slipping down like the rubber prosthesis. The existing tracheostoma was widened by an incision and by dilatation using a graduated set of tracheostomy obturators as dilators. Thereafter it was possible to insert the tube into the trachea; but, although it could be rotated upward, the flange could not be made to enter the trachea (a) because the prosthesis was too large, and (b) because the bulky junction between the two component parts could not negotiate the acute angle between the tracheostomy and upper trachea in one so small.

From this experience it was felt that successful management of this intractable stricture required (1) a small malleable tube based on this pattern, (2) that the junction area be hinged, and (3) that plastic material be used instead of metal.

Despite this aim, it was at least two months before the desirable piece could be found and fashioned. Many attempts to make varying types of tube had ended in failure; but ultimately the one illustrated in Fig. 2 (a, b) proved satisfactory. It was a piece of "portex" polythene tubing 5 cm. long and 6 mm. in diameter. One half had its lumen complete and the other half its wall cut away. This trimming allowed of a hinging movement at the junction of

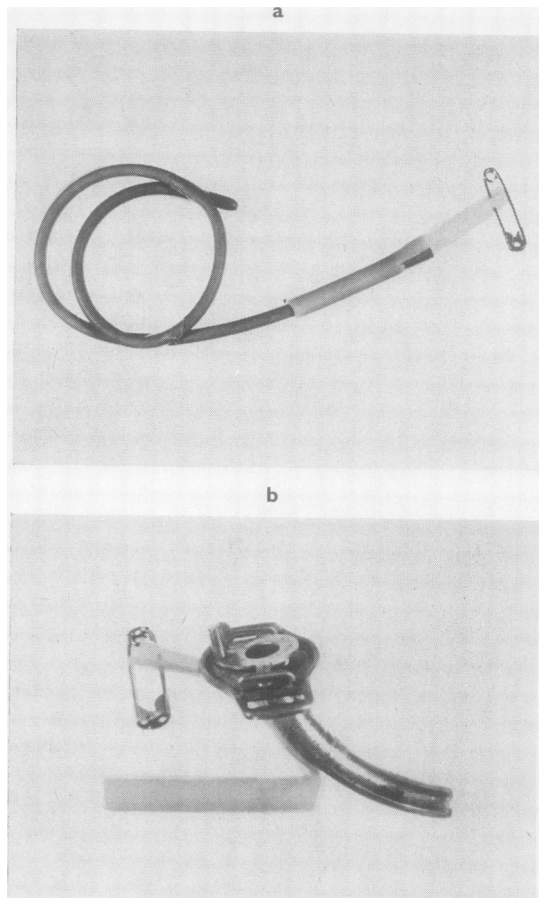


FIG. 2.—(a) The piece of portex tube 5 cm. long and 6 mm. in diameter, cut to the required shape, and mounted on to a Jaques No. 9 Fr. catheter, ready for inserting into the upper trachea. (b) Scheme to show how the polythene splint lay in the upper trachea, held in place by the polythene bevel which lay above the tracheostomy tube.

the tracheostoma with the trachea, and so allowed angulation of the polythene tubing to any angle at this point. Further, the half circle of the polythene would easily fit snugly above the tracheostomy tube, which would thus hold the prosthesis in position.

Insertion of Polythene Prosthesis.—On July 17 it was inserted. The method used was as follows:

(1) General anaesthesia was induced with open ether given through the tracheostomy tube.

(2) The polythene tube was cut to the required shape, and a No. 9 Jaques catheter was threaded through it in readiness for inserting as shown in Fig. 2a. It was hoped that the flange of the catheter would hold this tube as it was being inserted.

(3) When anaesthesia was deep enough, an infant bronchoscope was passed through the larynx and through the constricted area, down to the tracheostomy tube.

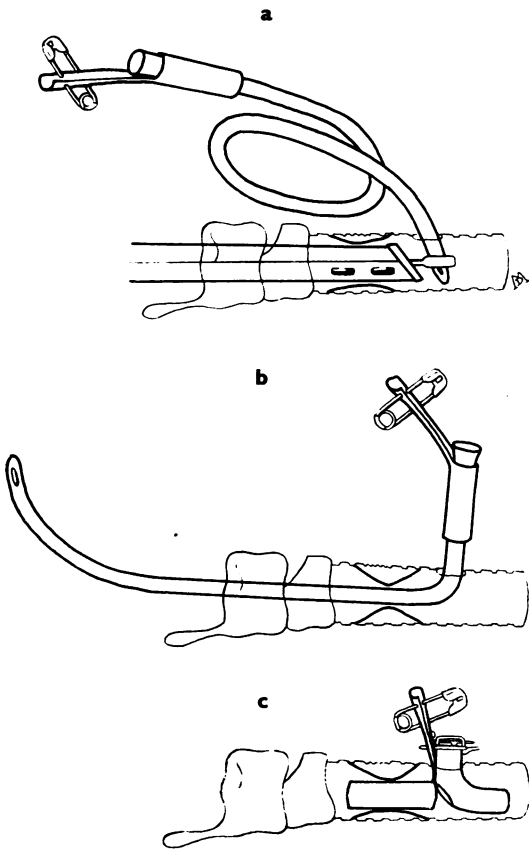


FIG. 3.—(a) The infant bronchoscope is passed through the larynx and stricture to the tracheostomy. The tip of a Jaques No. 9 catheter holding the polythene prosthesis is grasped with peanut grasping forceps. (b) The bronchoscope is then removed. The catheter now lies along the upper trachea and passes out through the mouth. (c) By gentle traction the polythene prosthesis is guided into place, and the tracheostomy tube re-inserted.

(4) The tracheostomy tube was next removed, and the tip of the catheter inserted down the stoma. During this manoeuvre the bronchoscope provided an adequate airway (Fig. 3a).

(5) When the catheter became visible through the bronchoscope, its tip was grasped with an infant peanut grasping forceps and the catheter guided along the bronchoscope.

(6) The bronchoscope was then removed. The catheter now lay through the tracheostoma, along the upper trachea, through the larynx and out through the mouth. At its lower, wide end was the polythene tube (Fig. 3b). By gentle traction on the catheter with the right hand, and guiding the polythene prosthesis into the tracheostoma with the left hand, it was possible to guide the polythene around the corner into the upper trachea and so on up through the stricture. Once this corner had been negotiated, normal breathing through the larynx was possible.

This manoeuvre, lengthy to explain, was quickly performed—a matter of seconds—and was possible because of the tight fit between the catheter and polythene prosthesis.

(7) Thereafter a safety-pin was inserted through the flap of the polythene tube still remaining outside the tracheostomy opening, so preventing it passing in. The catheter was next removed by pulling it firmly through the mouth, thus stretching the rubber and so lessening the snug fit between polythene and catheter. Finally, the No. 2 tracheostomy tube was re-inserted so as to lie immediately below the bevelled half of the prosthesis and thus keep it in place (Fig. 3c).

The patient now had two airways: (1) through the larynx via the prosthesis and (2) through the tracheostomy tube. Trial and error was required to get the exact length of prosthesis. The first proved too short, and the second proved too long and protruded through the vocal cords. It could be pushed down through the larynx with forceps via a bronchoscope. The desired length was finally obtained.

PROGRESS.—As a result of these manipulations, throughout the second half of 1956 the patient had a satisfactory polythene internal tracheal splint, 6 mm. in diameter, and held in position by the low tracheostomy tube supporting the bevel of the splint. Inspection at monthly intervals confirmed that the splint was lying immediately below the vocal cords.

To recapitulate, on December 10, 1955, a foreign body was removed from the larynx. Within three days the baby developed laryngeal oedema and required tracheostomy. During the succeeding fortnight 36 bronchoscopies were required through the tracheostoma for acute respiratory obstruction from inspissated mucus forming bronchial plugs. A tracheal stricture resulted. Two months later an endotracheal skin graft was applied to the strictured area, the upper tracheostomy closed, and a lower tracheostomy created. When the area was inspected four weeks after this operation, the tracheal wall was still strictured at the site of original tracheostomy. Six months after the onset of the stricture a polythene tube plastic prosthesis was finally satisfactorily placed in the upper trachea to act as a non-irritating endotracheal splint. Its position was checked at four-weekly intervals for a further six months, being finally adjusted under anaesthesia on December 27, 1956.

Final Removal.—That same evening, when he drank milk it was noted to flow down the trachea and emerge from the tracheostomy tube. The prosthesis was removed by withdrawing it through the tracheostoma; the tracheostomy tube alone was left in position.

By now the baby had been in hospital for a year. He had grown considerably, for his weight had risen from 1 stone 4 lb. on March 17, 1956, to 2 stones on March 24, 1957. Once the prosthesis had been successfully inserted he was given speech training by a speech therapist (Fig. 4).



FIG. 4.—Stephen at the time when the stricture was held open by the polythene prosthesis.

During January and February of 1957 it was noted that he could articulate despite the absence of the prosthesis. The tracheostomy tube was therefore plugged at intervals. On March 30, the tracheostomy tube was taken out for two hours without any respiratory discomfort. It was next removed for two hours each day. On May 10, 1957, one year and five months after the original intralaryngeal foreign body incident, peroral bronchoscopy showed a satisfactory lumen through the upper trachea and complete absence of any stricture. The tracheostomy tube was therefore finally removed and the stoma allowed to close by granulation.

DISCUSSION

The danger of intralaryngeal foreign bodies causing acute oedema of the glottis before or after removal, especially in infants, is very real and will frequently require emergency tracheostomy.

In a previous experience with a baby aged 10 months, the foreign body—a piece of egg shell in the larynx—remained unrecognized for three weeks until the onset of laryngeal oedema and respiratory obstruction (Borrie and Begg, 1955). Both cases were followed by secretions caking in the bronchi. Although earlier experience with nebulized sodium lauryl sulphate and the instilling of two or three drops of varidase had arrested the caking process, such treatment had no effect here. Time and the gentle removal of caked secretions alone secured the desired result.

The trimming of the anterior extremities of the tracheal rings on December 17, 1955, in the depths of the tracheostoma probably played a greater part in the later development of the tracheal stricture than was apparent at that time. On the other hand, the inevitable trauma of 36 bronchoscopies through the tracheostoma undoubtedly

contributed to this complication which declared its presence at the end of a gruelling three weeks of constant care. Though tight, the stricture could be dilated.

The management of the stricture in itself presented major problems. Search of the literature gave little help, nor could enquiry from colleagues through Australasia or from selected clinics in the United States of America give any parallel experience or help with such strictures in infants. The method of Erich (1945) using Stent moulds was not directly adaptable to one so young, nor with such a small trachea. A hinged, flexible splint was obviously required.

EXPERIMENTAL PROCEDURES.—The possibility of excising the stricture completely was also considered and the possible results explored by a series of survival experiments performed on young lambs.

Various methods of tracheal reconstruction have been tried for tracheal stenosis in adults, but none has been described for use in infants (Daniel, 1948; Gebauer, 1950; Grindlay and Waugh, 1951; Swift, Grindlay, and Clagett, 1952; Craig, Holmes, and Shabart, 1953; Rob and Bromley, 1953; Pacheco, Rivera, and Porter, 1954; Flavell, 1959).

Experiments were carried out on 12 lambs to assess the effects of excision on tracheal rings and the most desirable means of tracheal reconstruction (Borrie, 1957). The animals were anaesthetized and intubated as described elsewhere (Borrie, 1955), the trachea was exposed in the neck, and the experiments performed proximal to the inflated cuff of the endotracheal tube. There were no operative deaths, but seven died within two months from the effects of the implants or from stenosis. The remaining five were killed at varying times up to one year.

PARTIAL EXCISION OF TRACHEA IN FOUR ANIMALS.—Defects measuring 1.2 cm. square were made in the anterior tracheal wall, and covered with (a) cartilage autograft, (b) polyvinyl chloride sponge, or (c) tantalum wire mesh in double thickness. Because of the growth of wool, skin, which had been successfully used by Gebauer (1950) in humans, is quite unsuitable in sheep.

The cartilage autograft was rejected; one animal with polyvinyl chloride sponge developed fatal secondary haemorrhage and the other a fatal pneumonia. The tantalum wire mesh became epitheliated, caused minimal tracheal narrowing and was functioning normally after two months.

TRACHEAL RING EXCISION AND RE-ANASTOMOSIS IN FOUR ANIMALS.—One, two or three rings of

the trachea were excised. The tracheal segments, which separated for 3 cm., were readily approximated with interrupted 0000 silk sutures on curved cutting needles.

Follow-up examination showed that one and two tracheal rings could be excised and the trachea resutured with virtually no stricture formation; but when three rings were excised a tight stenosis (diameter 4 mm.) developed in six weeks.

TRACHEAL RING EXCISION AND TANTALUM MESH REPLACEMENT IN FOUR ANIMALS.—In one animal the trachea was divided, and the gap bridged by a piece of double tantalum wire mesh 3 cm. wide so that the tracheal ends were 1.5 cm. apart. In six weeks the defect was epitheliated but there was stenosis of half the tracheal lumen. In two animals two tracheal rings were excised and the resulting 3.5 cm. defect bridged as above. One had not become epitheliated in two and a half months but allowed normal breathing. The other formed a fatal tracheal stenosis in two and a half weeks. Where three rings were excised and the defect bridged with tantalum mesh respiratory obstruction caused death in four weeks.

CONCLUSION FROM EXPERIMENTS.—In lambs, while complete excision of one or even two segments of the trachea will allow of successful resuture without stricture formation, excision of three segments causes a tight stricture. Partial tracheal defects can be adequately repaired by tantalum wire mesh, but it is not a reliable method of bridging total defects made by excising two or three tracheal rings. None of these methods could be recommended for treating tracheal stenosis in infants.

When considered as a means of treating our patient, the possibility of a further stricture developing as a result of any attempt to excise the present one, either immediately or as a result of differential growth through adolescence, put excision and resuture out of the question. As it ultimately proved, by using a polythene tube prosthesis, the natural process of growth was the way to ultimate success.

In the final assessment the following offers the most likely reason why this stricture, which was related to the anterior tracheal wall, ultimately disappeared. With differential rates of growth it appeared that, once the narrowed area of the trachea was effectively held patent with a satisfactory prosthesis, differential rates of growth of the lateral tracheal wall proceeded normally.

The posterior tracheal wall thus grew away from the anterior wall, so that after six months, when the prosthesis was finally removed, an adequate lumen not only was present, but it remained and has increased in size with increasing years and growth.

SUMMARY AND CONCLUSION

The management of an intractable stricture of the upper trachea in a child aged 14 months is described. This followed high tracheostomy, done for oedema of the glottis after an intralaryngeal foreign body had been removed, and an episode of bronchial obstruction requiring 36 life-saving bronchoscopies.

Experiments on lambs established that, while complete excision of one or even two segments of the trachea will allow of successful resuture without stricture formation, excision of three segments causes a tight stricture.

The method finally adopted included:

(a) Epithelializing the first tracheostomy at stricture level with a skin graft and the establishing of a lower tracheostomy.

(b) Dilating of the stricture with a simple prosthesis made from polythene tubing, this being guided into position over a No. 9 Jaques rubber catheter via the second tracheostomy opening. The technique used is illustrated. Many other methods were initially tried before this simple device was discovered.

Some three and a half years after his final discharge the patient remains well, has started school, and has no evidence of recurrence. His speech is normal.

It is concluded that, in infants with post-tracheostomy strictures, provided the strictured area can be kept dilated with a simple polythene tube prosthesis held in position as illustrated, inherent differential rates of growth of the trachea will ultimately ensure a permanent and normal airway.

REFERENCES

- Borrie, J. (1955). *J. thorac. Surg.*, **29**, 426.
 — (1957). *Proc. Univ. Otago med. Sch.*, **35**, 15.
 — and Begg, N. C. (1955). *N.Z. med. J.*, **64**, 178.
 Craig, R. L., Holmes, G. W., and Shabart, E. J. (1953). *J. thorac. Surg.*, **25**, 384.
 Daniel, R. A. (1948). *Ibid.*, **17**, 335.
 Erich, J. B. (1945). *Arch. Otolaryng. (Chicago)*, **41**, 343.
 Flavell, G. (1959). *Proc. roy. Soc. Med.*, **52**, 143.
 Gebauer, P. W. (1950). *J. thorac. Surg.*, **20**, 628.
 Grindlay, J. H., and Waugh, J. M. (1951). *A.M.A. Arch. Surg.*, **63**, 288.
 Pacheco, C. R., Rivera, O., and Porter, J. K. (1954). *J. thorac. Surg.*, **27**, 554.
 Rob, C., and Bromley, L. L. (1953). *Thorax*, **8**, 269.
 Swift, E. A., Grindlay, J. H., and Clagett, O. T. (1952). *J. thorac. Surg.*, **24**, 482.