

INTRATHORACIC TRACHEAL TUMORS: DEVELOPMENT OF SURGICAL TECHNIQS FOR THEIR REMOVAL*

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SERIOUS PATHOLOGIC PROCESSES of the trachea are uncommon. Yet this organ and organs closely associated with it are exposed to a variety of diseases. It is remarkable that the increased incidence of bronchogenic tumors has not been paralleled by an increase in the incidence of tracheal tumors. Certainly the trachea is exposed to dangers as great as those to which the bronchi are exposed. Because tracheal tumors are rare and because direct surgical attack on them has not seemed feasible, they have received little attention even from endoscopists and thoracic surgeons. Treatment has been limited to endoscopic removal of the intraluminal portion of the tumor, implantation of radon seeds and application of roentgen rays. This has accomplished some palliation but has never been curative of malignant lesions.

It is now apparent that direct surgical attack can be made on some tracheal tumors. Many such tumors are cylindromas or squamous cell carcinomas, tumors that grow relatively slowly and which therefore lend themselves to surgical extirpation. Anesthesiologists have provided means of maintaining anesthesia and respiration in the presence of a surgically opened trachea. Surgical procedures performed on experi-

mental animals have demonstrated that resection of segments of the trachea is technically feasible. Increasing experience with pulmonary and intrathoracic operations has reduced the danger of surgical intervention on the trachea. We have used methods and technics developed in the experimental laboratory to operate on a small number of patients with tracheal tumors. Reports of these cases are included in this presentation.

Although tracheal tumors do not occur commonly there is considerable literature regarding them. Most of it consists of reports of one or two cases. In 1898, von Bruns²¹ was able to collect from the literature reports of 31 cases of carcinoma of the trachea. D'Aunoy and Zoeller⁶ in 1931, found reports of 351 cases. In 1938 Culp⁴ made a collective review of 433 primary tumors of the trachea. According to Ellman and Whittaker,⁷ 507 cases had been reported in the literature up to December, 1945. Only a few additional cases have been reported since.

Tumors can develop anywhere in the trachea but usually they are in the posterior or lateral wall of its lower third. The anterior wall is rarely concerned. The disease affects men predominantly, as is true also of bronchogenic neoplasms. Most patients are between 40 and 70 years of age.

Pathologic studies of tracheal tumors have resulted in considerable confusion.

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According to Ellman and Whittaker,⁷ 20 different pathologic types of tracheal tumor have been described. Although critical study probably would reduce the variety of tumors that have been described, obviously the trachea can be the site of a wide assortment of neoplasms. Tinney, Moersch and McDonald²⁰ made careful pathologic study in 27 cases; 11 of their tumors were squamous cell carcinomas, eight were cylindromas, six were adenocarcinomas and two were hemangio-endotheliomas.

Squamous cell carcinoma may produce either a fungating or an ulcerating growth. It tends to invade directly through the tracheal wall, and the adjacent lymph nodes may be involved. Cylindroma of the trachea is identical histologically to the tumor of the same name that occurs in the salivary glands, lacrimal glands, nasal sinuses, mouth, nose or throat. It probably arises from mucous glands in the wall of the trachea and is covered with an intact mucosa. Cylindroma extends through the wall of the trachea, and the extraluminal portion of the tumor may be larger than the intraluminal portion. These tumors grow slowly but are not encapsulated and tend to infiltrate. This characteristic has a bearing on treatment, as will be explained.

Adenoma usually originates in the bronchus but may involve the lower part of the trachea secondarily. In evaluating older reports of cases of adenoma of the trachea it must be remembered that many of these lesions might now be classified as cylindroma. Adenocarcinoma originates from the mucosa and may produce either a polypoid mass or an ulcerated lesion. This tumor is malignant, but fortunately it is of uncommon occurrence.

SYMPTOMS

The symptoms produced by tracheal tumors are dependent primarily on mechanical factors (Fig. 1). A tumor with a wide

base and which extends into the lumen of the trachea will interfere with both inspiration and expiration. A pedicled tumor, on the other hand, may cause obstruction to either inspiration or expiration. Dyspnea has been the earliest and most prominent symptom in cases of tracheal tumor and was present in 90 per cent of cases studied at the Mayo Clinic. The dyspnea may be con-

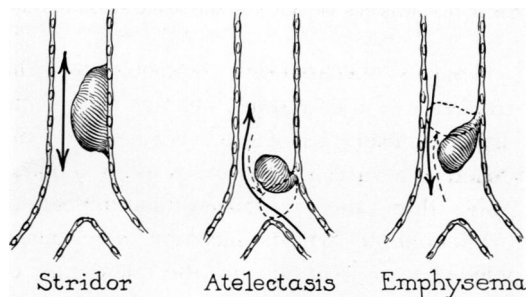


FIG. 1.—Mechanical factors that are responsible for some of the symptoms of tumor of the trachea. [From H. J. Moersch: Symposium on Certain Tumors of the Bronchi (Adenomas and Cylindromas) and on Tumors of the Trachea: Clinical Aspects. Proc. Staff Meet., Mayo Clin., 21: 410, 1946. (By permission)]

stant or paroxysmal and is often affected by the position assumed by the patient. Most patients with tracheal tumors are thought to have asthma for some time before the possibility of an obstructing lesion is considered. As might be expected, wheezing is a common symptom and occurred in 63 per cent of our cases. Tracheal tumors commonly produce cough. At first the cough may be dry and unproductive but as the tumor ulcerates or obstructs the tracheo-bronchial tree it becomes productive. Hemoptysis has occurred in 44 per cent of the cases encountered at the clinic. Hoarseness is a fairly common symptom of tracheal tumor and is often due to involvement of the vagus or recurrent laryngeal nerves by peritracheal extension of the tumor or by infiltration of lymph nodes.

DIAGNOSIS

Physical examination may give a negative result. The presence of a wheeze or stridor is significant. The usual postero-anterior thoracic roentgenogram may reveal nothing abnormal unless the tumor is causing obstruction of a main bronchus. Occasionally the extratracheal portion of a tracheal

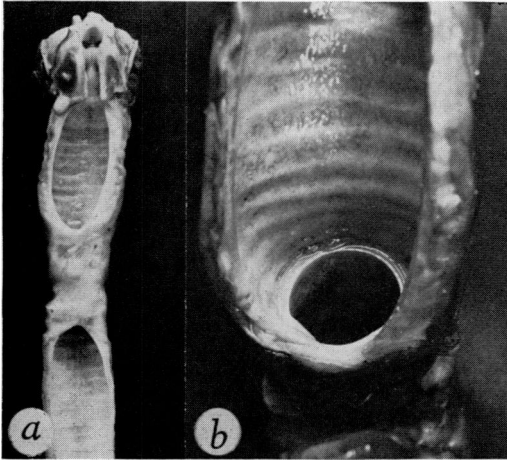


FIG. 2.—(a) Trachea of a dog in which a polythene prosthesis had been placed following tracheal resection. Note absence of reaction in the trachea above and below prosthesis; (b) closeup view of prosthesis in trachea of a dog. Note snug fit and absence of reaction.

tumor can be demonstrated. Tomography is valuable. Roentgenograms taken after iodized oil has been instilled into the trachea may show the presence of a tracheal tumor. Occasionally the tumor is seen on direct laryngoscopy. Accurate diagnosis of the pathologic type of tracheal tumor, together with determination of its location and extent, can be accomplished best by bronchoscopy. There is considerable variation in the bronchoscopic appearance of the tumors. They may be polypoid, or flat and infiltrating. They may occur as smooth and rounded protrusions with normal mucosal covering, or as excavated ulcerating lesions. In some cases the endoscopist can remove all or most of the intraluminal por-

tion of the tumor. Specimens can be obtained for biopsy.

TREATMENT

In the past the treatment for tracheal tumors has been the province of the endoscopists. This treatment consisted of piecemeal removal of the intraluminal portion of the tumor with bronchoscopic biopsy forceps, diathermic cauterization of the base of the tumor, implantation of radon seeds and radiation. Obviously, a more nearly ideal treatment, and one more in keeping with the accepted principles of surgery, would be complete excision of the tumor with a surrounding margin of normal tissue and, if possible, of the lymphatic vessels and nodes draining the site of tumor. Until recent years such treatment has not been considered technically feasible.

In 1946, we began experiments in the course of which we found that short segments of the tracheas of animals could be removed and primary anastomosis of the ends of the trachea could be performed successfully. We have been able to bridge long gaps between the cut ends of the trachea with polythene plastic tubes and many of the animals have lived for four and five years without difficulty (Fig. 2a and b). The defect left by excision of only a part of the circumference of the tracheal wall has been repaired successfully in a variety of ways. Resection of an entire lung together with the tracheal bifurcation, followed by primary anastomosis of the bronchus of the remaining lung to the trachea, has been performed successfully in a number of instances. Much of this work has been summarized in our previous reports.¹¹⁻¹³

Experimental studies of surgical procedures performed on the tracheas of animals have been reported in recent years by Hanlon,¹⁴ Daniel,⁵ Maisel and Dingwall,¹⁹ Carter and Strieder,² Jackson and his associates,¹⁵ Gebauer,⁹ Juvenelle¹⁷ and Fergu-

son and his associates.⁸ These studies have confirmed the fact that surgical procedures involving the trachea are feasible and they have resulted in the development of necessary technics.

Clinical experience with surgical resection of tumors involving the intrathoracic

weeks stenosis of the trachea made replacement of the tube necessary. Thirty-three months after operation the patient died of generalized pleural and pulmonary metastasis. In 1948 Gibbon¹⁰ reported a case in which resection of a bronchial adenoma required sacrifice of a portion of the lower

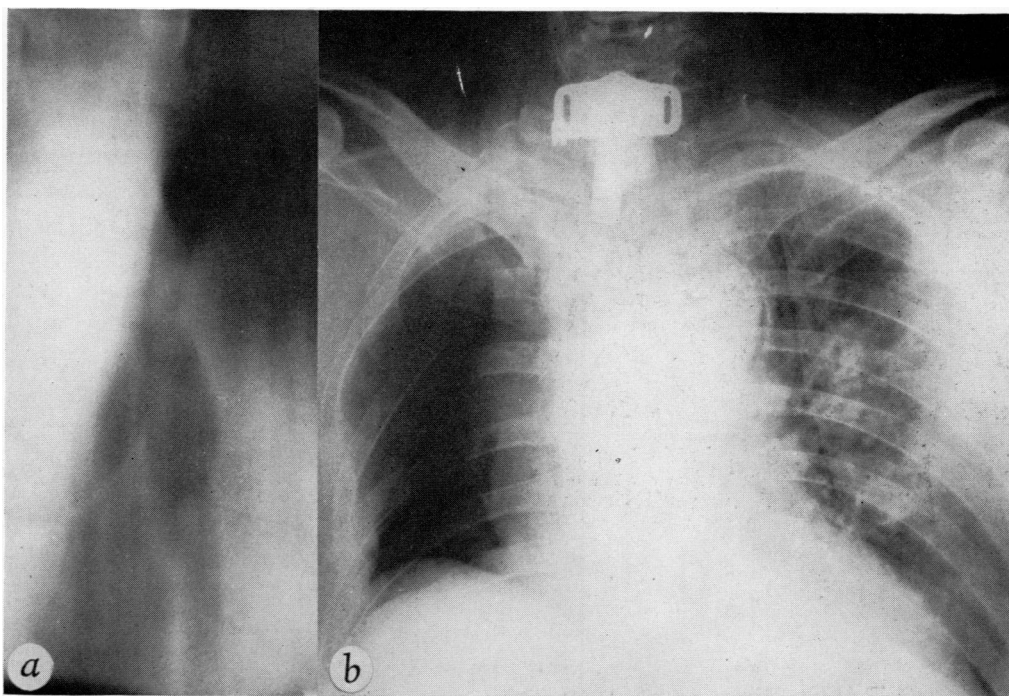


FIG. 3.—(a) Tomogram demonstrating marked constriction of trachea by extensive cylindroma located just above the bifurcation of the trachea; (b) roentgenogram immediately following pneumonectomy and resection of the lower trachea with connection of left main bronchus to the trachea by means of a polythene tube.

portion of the trachea is limited. In 1946 Belsey¹ reported a case of adenoma of the trachea in a woman, 42 years of age. The lesion of the lateral wall of the trachea was resected and the defect was repaired with a fascial patch reinforced with steel wire. The patient survived the operation but the report does not indicate the course of events after two weeks. In 1950 Jarvis¹⁶ mentioned a case in which a tracheal cylindroma located just below the sternal notch was excised. Tracheal continuity was maintained with a tube of stainless steel. After a year the tube was removed. Within three

part of the trachea. The defect was successfully repaired with a pedicled flap of parietal pleura wrapped around a piece of costal cartilage. Recently Kergin¹⁸ reported an instance of squamous cell carcinoma on the right wall of the lower part of the trachea in a man 51 years of age. Local excision was performed and the defect was closed with a free graft of pericardium. The lesion recurred locally and radical resection was performed. The right lung and right wall of the trachea were resected. However, a long segment of the right main bronchus was preserved and brought up as

a pedicle flap to cover the defect. The patient's death on the third postoperative day prevented further evaluation of the procedure.

REPORT OF CASES

Case 1.—A man, 49 years of age, registered at the clinic January 16, 1941. He gave a history of cough and recurrent bouts of pneumonia of 3

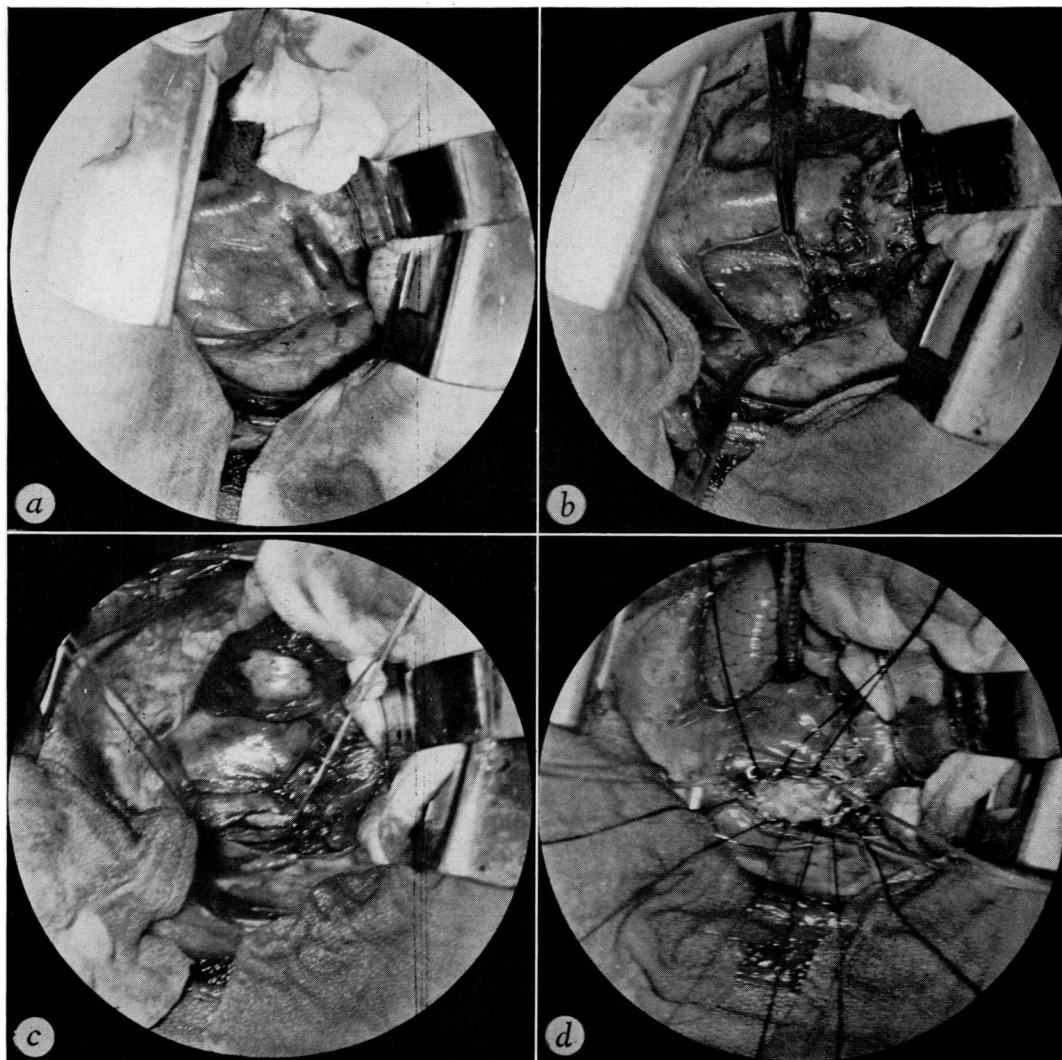


FIG. 4.—(a) Tumor in the right wall of the trachea just above the azygos vein; (b) mediastinal pleura dissected away from tumor; (c) tracheal defect following excision of tumor; intratracheal tube visible within the trachea; (d) fascial patch sutured over tracheal defect.

Our experience with resection of tracheal tumors consists of surgical procedures performed in four cases. Three of the lesions were cylindromas and one was a squamous cell carcinoma. Three of the patients were men and one was a woman. Their ages were 47, 49, 59 and 60 years.

years' duration. About 6 months prior to his admission, wheezing had developed and a diagnosis of asthma had been made.

Physical examination and laboratory studies, including roentgenograms of the thorax, revealed no significant abnormalities. Bronchoscopy disclosed a large tumor almost filling the lower part of the trachea. The intratracheal portion of the

tumor was removed bronchoscopically and examination of the tissue resulted in a diagnosis of cylindroma. During the next 6 years bronchoscopic removal of recurrent tumor was performed on 12 occasions. In addition, the tumor was treated by roentgen rays. The patient's condition remained good and there was no evidence of metastasis.

In spite of all endoscopic measures the lumen of the trachea gradually diminished and by January, 1947, it became apparent that nothing fur-

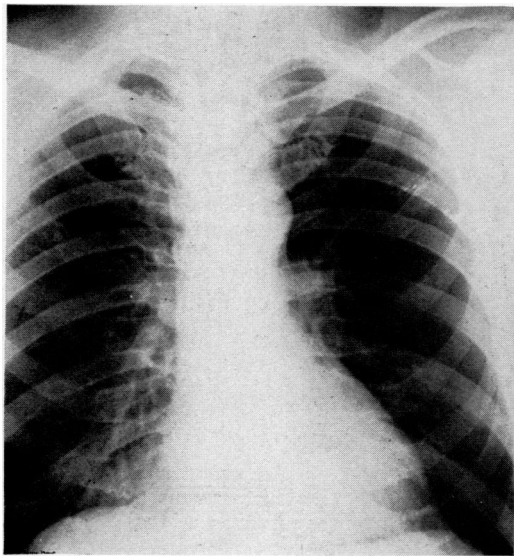


FIG. 5.—Roentgenogram revealing a slight bulge in the right superior mediastinum indicating location of tracheal tumor.

ther could be accomplished by conservative means and operation was performed (Fig. 3a). The tracheal lumen was so narrowed that it was necessary to insert a bronchoscope beyond the obstruction before the anesthetic agents could be administered. Nitrous oxide, oxygen and ether were then given through the bronchoscope. A posterolateral incision was made on the right side of the thorax. The surgeon mobilized the trachea without opening the left pleural space. Plans were made to remove the tumor and replace the resected segment of trachea with a polythene tube. The extent of the tumor made this procedure impossible. It was necessary to resect the lower third of the trachea together with the right lung. The left main bronchus was connected to the trachea with a long polythene tube which was secured at each end with silk ligatures. There was no leakage of air. The mediastinal pleura was repaired and the thoracic wall closed. Tracheotomy was performed to facilitate aspiration of secretions.

The patient tolerated the operation well and the postoperative course for the first four days was uneventful, although accumulation of secretions in the tracheobronchial tree required frequent aspiration (Fig. 3b). On the fourth day evidence of bronchopneumonia appeared and this complication progressed in spite of administration of antibiotics. The patient died on the sixth postoperative day.

Necropsy revealed that the polythene tube partially blocked the bronchus of the upper lobe of the left lung and this was undoubtedly responsible to a large extent for the bronchopneumonia. (This case was reported by us in 1948.³)

Case 2.—A woman, 60 years of age, registered at the clinic October 28, 1949. She gave a history of cough, hemoptysis, wheezing and dyspnea of only 4 weeks' duration. Thoracic roentgenograms made elsewhere had given negative results. Bronchoscopy revealed a large polypoid tumor arising from the right wall of the trachea. The intratracheal portion of the tumor was removed bronchoscopically. It proved to be a cylindroma. Tomograms demonstrated a small mass outside the tracheal wall at the site of the tumor.

On November 17, thoracotomy was performed on the right. The tumor of the trachea was excised (Fig. 4a, b and c). The defect in the tracheal wall was closed with a fascial patch reinforced with steel wire (Fig. 4d). This patch was further reinforced with a small sheet of gelatin-foam sponge. The mediastinal pleura was closed and tracheotomy was performed.

The postoperative course was uneventful. The patient was kept in the hospital for 3 weeks. She was examined 11 months after operation. There were no complaints and when bronchoscopy was performed it was difficult to recognize any abnormality of the trachea.

Case 3.—A man, 59 years of age, registered at the clinic in December, 1949. He gave a history of cough and hemoptysis of about 6 months' duration. Bronchoscopy elsewhere had revealed a lesion of the trachea and biopsy had been reported to show squamous cell carcinoma.

Physical examination and laboratory studies, including thoracic roentgenograms, gave negative results (Fig. 5). Bronchoscopy confirmed the presence of an ulcerating lesion of the right wall of the middle third of the trachea and biopsy confirmed the previous diagnosis of squamous cell carcinoma.

Operation was performed January 3, 1950. The tumor was in the right wall, and near the upper limit of the intrathoracic portion of the trachea (Fig. 6a). Several adjacent lymph nodes

were obviously involved by carcinoma. The right recurrent laryngeal nerve was involved in the tumor and the invaded segment was removed. The patient was dismissed 3 weeks after operation.

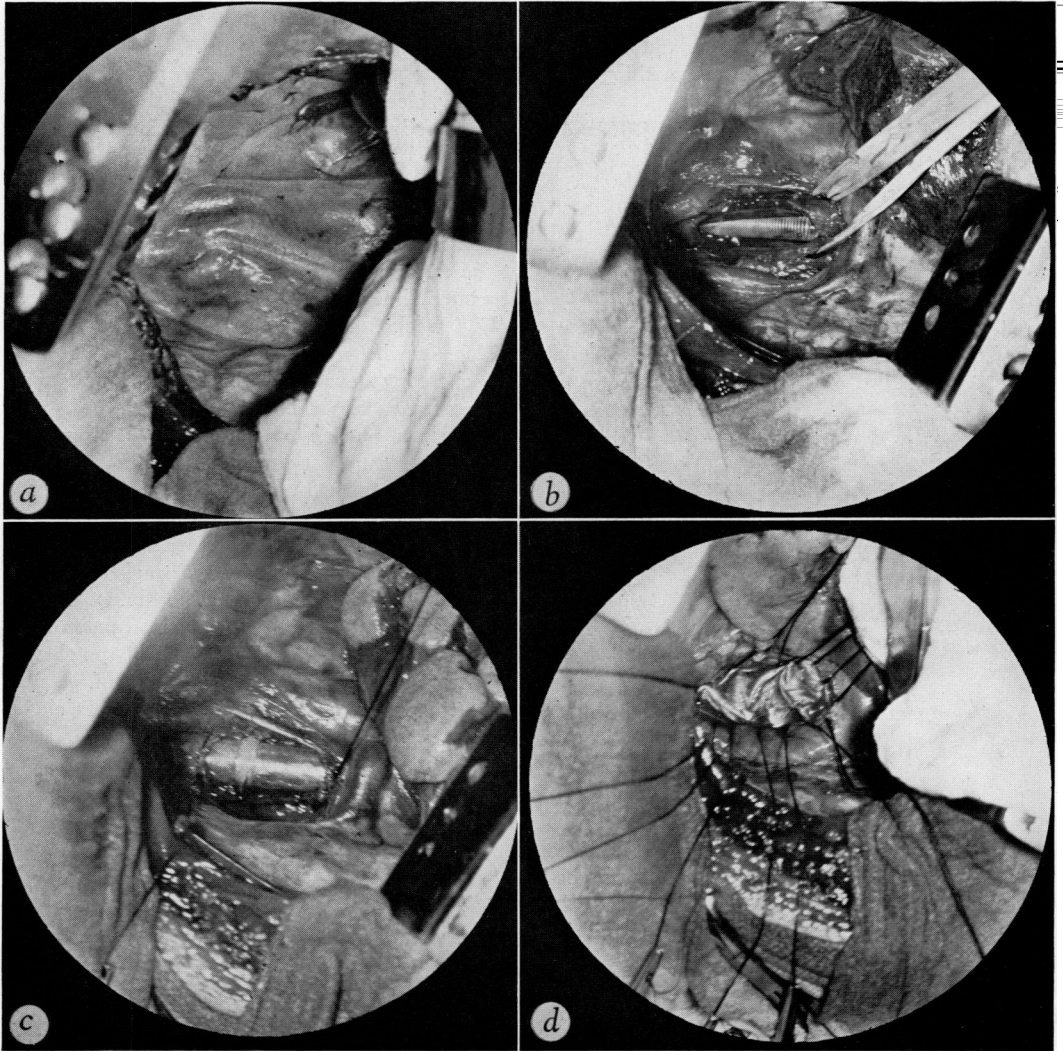


FIG. 6.—(a) Tracheal tumor bulging beneath the pleura just behind the trachea in the upper part of the thorax. (b) Tracheal defect following resection of the tumor. Intratracheal tube is visible within the trachea. (c) Polythene tube in place in tracheal lumen; (d) fascial patch sutured over the tracheal defect.

6b). This required resection of more than half the trachea. The patient had a severe hemoptysis but there was no evidence of metastasis.

During the next year this patient had an increasing amount of trouble and widespread metastasis appeared, causing death a little less than 2 years after operation. The polythene tube had provided a well-functioning airway until his death.

Case 4.—A man, 47 years of age, registered at the clinic in January, 1948. He gave a history of cough, wheezing, dyspnea and hemoptysis of about 3 years' duration. Physical examination re-

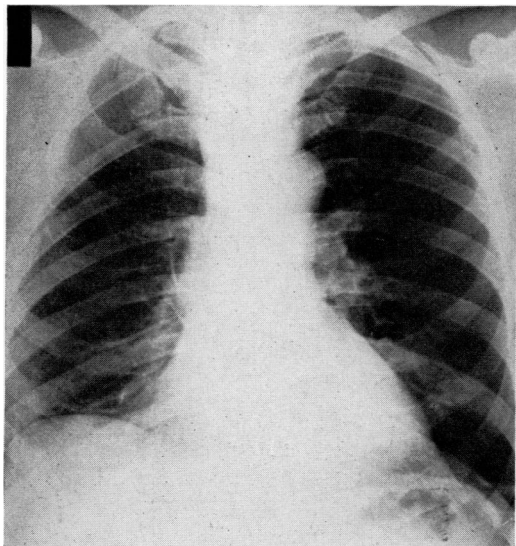


FIG. 7.—Thoracic roentgenogram one year after tracheal resection.

vealed the presence of wheezing respiration. There were no other abnormal findings. Roentgenographically, the thorax seemed to be normal except for a small region in the apex of the left lung that was interpreted as healed tuberculosis. Bronchoscopy revealed a rounded tumor arising from the right wall of the lower part of the trachea. This tumor was removed endoscopically and was reported to be a cylindroma. Surgical resection was advised but the patient refused. During the next 3 years recurrent tumor was removed by bronchoscopic means on 14 occasions.

By May, 1951, a large extraluminal tumor had developed and at this time the patient accepted operation (Fig. 8a and b). The tumor involved about half the circumference of the trachea. It was about 3 cm. in length and involved the point of origin of the right main bronchus. The tumor was excised. The defect was repaired with a free fascial graft reinforced and contoured with steel wire and sutured to the edges of the defect with interrupted silk sutures. The trachea was wrapped

with a sheet of gelatin-foam sponge. The mediastinal pleura was repaired and the thorax was closed. Tracheotomy was performed. The resected specimen measured 7.5 by 5 by 4.5 cm.

The postoperative course was satisfactory and the patient was dismissed 3 weeks after operation. Bronchoscopy was performed 8 times at various intervals after operation. Considerable amounts of granulation tissue appeared at the site of repair and produced cough and expectoration. The steel wire that had been used to reinforce the fascia could be seen in the granulations.

Six months after operation this wire was removed through the bronchoscope. Cough disappeared following this procedure and 9 months after operation bronchoscopy revealed a well-healed and only slightly narrowed tracheal lumen. Whether stricture of the trachea will develop in the future cannot be determined at this time. The patient is now well and free of symptoms 11 months after operation.

COMMENT

It seems unlikely that one surgeon will ever have the opportunity to carry out surgical excision of a large number of tracheal tumors. As has been indicated, these lesions are rare and in many instances the location of the tumor or the extent to which it has spread by the time diagnosis is made will preclude any attempt to remove it. More experience and further improvement in technic may widen the field of application of surgical procedures to tracheal lesions.

Tracheal tumors should be considered among other possibilities when the physician is confronted by patients who present themselves with a history of cough, expectoration, dyspnea and wheezing. Certainly a patient more than 40 years of age who complains of wheezing of recent origin should not be considered to have bronchial asthma until the possible presence of an obstructing lesion of the trachea has been ruled out. At one time or another in three of the four cases reported in this paper, the diagnosis of asthma was entertained.

The failure of postero-anterior thoracic roentgenograms to reveal presence of the condition has been pointed out. Results of

routine thoracic roentgenography were reported to be essentially negative in all four of our cases of tracheal tumor. Furthermore, in our experience, about 10 per cent of bronchial adenomas that involved the major bronchi did not produce any roentgenographic evidence of their presence.

gical resection. Evidence of the slow growth of cylindromas is the fact that the lesions had been present in one of our cases for nine years and in another for six years before surgical resection was carried out. In neither case was there evidence of metastasis at the time of operation. How-

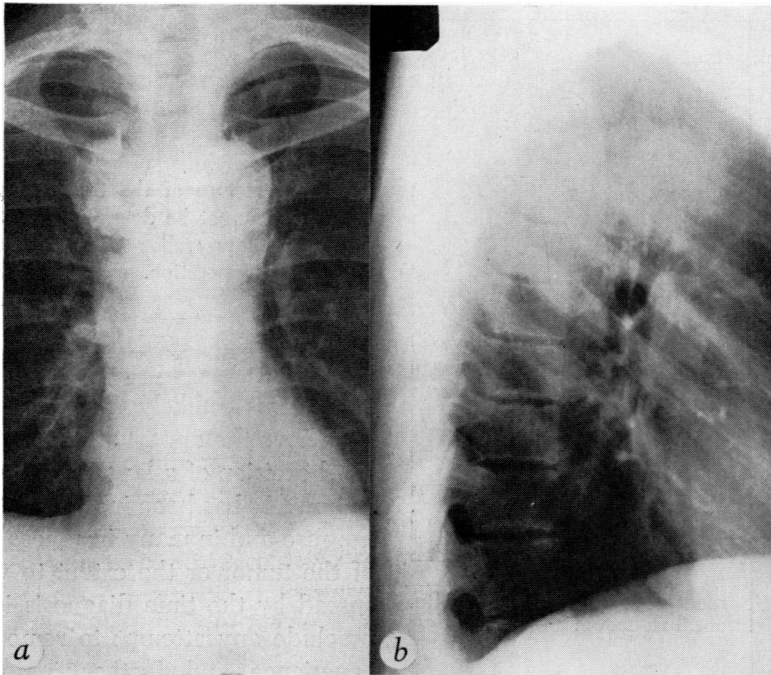


FIG. 8.—(a) Large cylindroma arising from the right tracheal wall and extending into the right pleural space; (b) lateral view of tumor involving the right wall of the trachea.

The investigation of any case of unexplained cough, hemoptysis, wheezing or dyspnea should invariably include careful bronchoscopic examination of the tracheo-bronchial tree.

That the more common pathologic types of tracheal tumor are squamous cell carcinoma and cylindroma is fortunate because these tumors grow more slowly than do the less common varieties such as adenocarcinoma and hemangio-endothelioma. From a surgical standpoint this fact offers some encouragement since these more common lesions offer better opportunities for sur-

ever, if experience with resection of cylindromas of the trachea parallels that of resection of cylindromas of the mouth, nose, throat or salivary glands, we have little reason to expect that these tumors will be permanently cured. They grow slowly but tend to infiltrate and eventually to metastasize even after fairly radical excision. The operation, therefore, must include removal of normal adjacent tissue. Fortunately for the surgeon, tracheal tumors tend to develop in the lower third of the trachea where they are most readily accessible. We have been particularly fortunate in the four

patients upon whom we have operated in that the tumor involved the right wall of the trachea. Obviously the arch of the aorta would interfere considerably with the exposure of tumors involving the left wall of the lower part of the trachea.

There is obviously a need for better technics for repair of the tracheal defects than those we have used. The use of wire-reinforced fascia seems to be a fairly satisfactory type of repair for small tracheal defects, but experience with our fourth case, in which the wire had to be removed bronchoscopically, indicates that this method of repair is not adequate for larger defects. Further experience will be necessary to determine whether grafts of skin, pericardium or some other tissue may be preferable or whether a prosthesis of some metal or plastic material can be used to best advantage.

We believe that cervical tracheotomy should be performed routinely in all cases in which operations involving resection of the trachea are performed. It permits frequent removal of tracheobronchial secretion and adds immeasurably to the comfort and safety of the patient during the early postoperative course.

This presentation can be considered only as a preliminary report. The questions and problems that we have been able to answer are fewer than are those that remain unanswered. We cannot predict that surgical resection of tracheal tumors will ever offer a greatly rewarding field of endeavor. However, our experience thus far encourages us to continue our experimental and clinical efforts.

SUMMARY

Tracheal tumors do not occur commonly. They are usually found in men between 40 and 70 years of age. These tumors usually involve the lateral walls of the lower third of the trachea. The symptoms usually are

cough, hemoptysis, dyspnea and wheezing. Routine thoracic roentgenograms often show no detectable evidence of a tumor. Tomograms frequently are necessary to demonstrate these tumors roentgenographically.

Tracheal tumors are best diagnosed by bronchoscopy. The most common types are squamous cell carcinoma, cylindroma, adenocarcinoma and hemangio-endothelioma. Cylindromas of the trachea tend to grow slowly and to metastasize late, and thus are the most favorable type of tracheal tumor for surgical resection.

Extensive experimental studies have resulted in the development of technics that permit surgical resection of some tracheal tumors.

Four cases of tracheal tumors are reported in which surgical resection of the tumor was performed. Three of the tumors were cylindromas and one was a squamous cell carcinoma. One patient died of bronchopneumonia six days after operation and one died two years after operation of local recurrence and extensive pulmonary metastasis. One patient is alive and well 11 months after operation and one is alive and well one year after operation.

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DISCUSSION.—DR. RICHARD H. SWEET, Boston: I wish to compliment Dr. Clagett upon the excellence of his presentation, and the most interesting experience which he has rehearsed.

The rarity of these tumors prompts me to mention an experience of my own with a cylindroma, which brings up a question I would like to ask of Dr. Clagett.

The patient, an elderly man, had one of these tumors in the cervical segment of the trachea, and I found that it was necessary, in order to remove it, to do a segmental or sleeve resection of the trachea. Much to my amazement, I found that it was possible to mobilize the lower end sufficiently to bring it up for an end-to-end anastomosis. That patient is still living; it has been about five years since then.

I just wondered if Dr. Clagett thought in some of these cases that might be done in the thoracic segment. I have not had an opportunity to test the possibility, however.

DR. OTTO C. BRANTIGAN, Baltimore, Md.: Dr. Clagett's work was very well presented. He deserves a lot of credit for his pioneer effort in this field. I would like to report on work done by Dr. Penton and me. To repair the trachea we con-

ceived the idea of a viable pedicle graft obtained from the chest wall. One or more ribs are removed in a subperiosteal manner, depending on the desired width of the graft. A strip of chest wall is then cut free except at its source posteriorly; it includes periosteum, pleura, intercostal muscles, nerves, veins and arteries.

Since the blood and nerve supply is intact, almost any type of tracheal repair has been possible on the dog, including the replacement of the complete circumference of the trachea. There has been no tendency toward stricture formation.

We have not had a patient with a tumor of the trachea. However, a patient with carcinoma of the esophagus complicated by an esophago-tracheal fistula was subjected to operation. An esophagectomy was done. In order to remove the tumor from the trachea and bronchi, a little more than half the posterior circumference of the lower two to two and one-half inches of the trachea and both main bronchi had to be resected. This defect was easily repaired with the viable chest wall graft. No rigid support was used in the graft but a soft rubber tube was placed in each bronchus and brought up through the trachea and out through a tracheotomy wound in the neck. The patient made an uneventful recovery but died four months later of generalized metastasis.