

## TRACHEAL DIVERTICULUM AND CONGENITAL OESOPHAGO-TRACHEAL FISTULA WITHOUT OESOPHAGEAL ATRESIA

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The following case is reported because of its unusual interest in three respects: the presence of a congenital oesophago-tracheal fistula without atresia in a girl of 16 years; the co-existence of a tracheal diverticulum; an anatomical anomaly of the thoracic duct which caused a post-operative chylothorax.

### CASE REPORT

N. R., born on March 19, 1934, was referred to us on November 6, 1950, with an oesophago-tracheal fistula. The first abnormal symptoms were noted shortly after birth, when, at the beginning of each feed, she coughed and became cyanosed, expectorating a whitish sputum. Change of position had no effect in preventing these symptoms, and only feeds given drop by drop were tolerated without great distress.

At the age of 1 year she had bronchopneumonia, followed two years later by a bilateral pulmonary infection, and her childhood was repeatedly interrupted by similar infections.

In November, 1948, following an appendicectomy, a focus appeared in the left lower pulmonary lobe, and this was accompanied by pleural effusion.

At the beginning of 1949 bronchography with lipiodol revealed a moderate degree of cylindrical bronchiectasis of the cardiac bronchus of the lower lobe of the right lung, and on the left side bronchiectasis of the basal segments of the lower lobe and lingula.

A barium swallow showed no oesophageal stenosis and the trachea was faintly outlined by barium. Bronchoscopy (Dr. Couder) confirmed the presence of an oesophago-tracheal fistula. No other significant abnormality was detected in the patient or her relatives.

On admission in November, 1950, when aged 16 (height 5 ft., weight 5 st. 12 lb.), the patient's complaints were of breathlessness on exertion and difficulty in swallowing, together with violent attacks of coughing associated with the expulsion of ingested food.

Such attacks always occurred after taking liquids but seldom after solid food. They occurred imme-

diately after drinking, being manifested by the return of fluid into the mouth with a burst of coughing, followed by a fresh act of swallowing which would resolve the difficulties.

On examination she was seen to be underdeveloped for her age and had cyanosed cheeks. The fingers were not clubbed. Auscultation revealed no abnormality. No abnormality was detectable in the abdomen or in the cardiovascular and nervous systems. Blood pressure was 105/65 mm. Hg. Intellectual development was below normal. Menstruation had been normal since the age of 16.

Radiographs of the chest showed normal lung fields with enlarged hilar shadows and a left-sided cervical rub.

The tuberculin test was negative.

The blood picture was 4,960 R.B.C.s, 5,200 W.B.C.s (50% polymorphs, 16% eosinophils, 13% monocytes, 20% large lymphocytes, and 1% small lymphocytes). Bleeding and clotting times were within normal limits, and her blood was group B Rh negative.

At operation on November 28 the anaesthetic was given by Dr. Chateaufreud.

An L-shaped incision was made outlining the scapula, and the third right intercostal space was opened. No pleural adhesions were encountered and the oesophagus and trachea under the azygos arch were freed.

There was a large tracheal diverticulum with diameter of 2.5 cm. opening into the right posterior wall of the trachea at the level of the bifurcation; its base was applied to the oesophagus.

During the dissection of the fistula a little drop of milky fluid appeared when a small pedicle was divided. It seemed likely that a small lymph vessel had been injured, but after swabbing nothing else was seen and the dissection was continued. The fistula was divided and the oesophageal opening closed in two layers with interrupted nylon sutures. The first included the entire thickness of the oesophagus and was reinforced by a superficial invaginating layer of sutures. The repair was difficult because the muscular layer was practically non-existent at the level of the fistula. The tracheal orifice was located

on the posterior aspect about 3 cm. above the bifurcation, and it was closed by interrupted nylon stitches.

The tracheal diverticulum, containing air and mucus, was removed. It communicated with the trachea through an orifice about 3 mm. in diameter, which was also repaired with interrupted nylon sutures. The thoracotomy was closed without drainage.

Difficulties arose after operation. Expectoration was frequent, amounting to three spittoons of mucus in 24 hours. Radiographs showed a shadow in the right hemithorax. Aspiration of the right pleural cavity withdrew 300 ml. of serous, viscous fluid which agglutinated in the receptacle thereby preventing clinical or cytobacteriological examination. A test swallow with coloured fluid suggested that a fistula remained, and re-operation was decided upon.

On December 2 the original incision was reopened and 2 litres of chyloform fluid evacuated. The lung was freed and the region of the fistula explored. Chyle issued from the top of the thorax, and a careful endothoracic dissection of the base of the neck was carried out, examining the subclavian, vertebral, and internal mammary arteries, the oesophagus, and the trachea. It seemed that a fistula of an abnormal right thoracic duct existed, situated under the right mediastinal pleura behind the superior vena cava. After dissection and ligation of two or three small lymphatic and one voluminous vessel, chylostasis seemed complete. The trachea was explored and was watertight on testing. The oesophagus was suspicious at a point level with the suture line of the first operation. There was probably a small fistula there, and this was closed in two layers.

The thoracotomy was closed with dependent pleural drainage.

No complications ensued, and the pleural drain was removed in two days. Drinking no longer caused outbursts of coughing, and normal feeding was gradually permitted after a fluid diet in the early post-operative phase. The patient had an epistaxis on the fourth day, but the blood count was 4,170,000 R.B.C.s and 5,000 W.B.C.s with 29% eosinophils and a decrease of platelets. Radiographs revealed a clear right hemithorax. Skin sutures were removed on the sixth day.

Bronchoscopy on December 29 by Dr. Oustrière demonstrated on the posterior tracheal wall 4 or 5 cm. below the glottis several nylon stitches marking a suture line about 1.5 cm. long.

The patient was discharged on January 11, 1951. She was seen again on January 16. She no longer experienced bursts of coughing when drinking, but she was still unsure about swallowing, choking sometimes when taking fluid rapidly.

The radiograph of the chest was practically normal, but a barium swallow showed the right lower lobe bronchus partially outlined by barium, suggesting a persistent oesophago-tracheal communication.

Oesophagoscopy performed by Dr. Bourgeois showed an area of granulation tissue on the right

anterior wall of the oesophagus. Bronchoscopy, also by Dr. Bourgeois, detected two nylon stitches not well attached and partially obstructing the trachea; these were removed. Two other stitches were also just visible, with granulation tissue between, but neither saliva nor methylene blue ingested by the patient was visibly infiltrating into the trachea.

A lower fistula no longer existed. Deglutition of fluids appeared somewhat awkward and methylene blue spilled into the larynx. There was, furthermore, a paralysis of the right recurrent laryngeal nerve.

The patient returned to the hospital on July 10, 1951. She did not present any difficulties of deglutition or of respiration, but she complained of persistent headache with lipothymia, of which Dr. Marty thought the origin was probably neurotic.

Pathological examination by Dr. Renault showed that the diverticulum consisted of a little pouch formed by a very inflamed membrane of bronchial origin.

## DISCUSSION

The first interesting point of our case is the fact that it was an oesophago-tracheal fistula with oesophageal atresia. It was congenital because the difficulties of deglutition were noticed from birth although tolerated until the age of 16 years.

The first observation would have been the one reported by Richter in 1792, but this case, considered to be a simple fistula by Cautley, is quoted by Plass as a peculiar abnormality in which trachea and oesophagus formed a single entity.

In 1873 Pinard and Tarnier reported the case of a newborn child presenting several congenital abnormalities among which was a high oesophago-tracheal fistula, situated just beneath the glottis, the oesophagus being otherwise normal.

At the same time in 1873 Lamb published his clinical and anatomico-pathological observations of a child who lived for seven weeks with a simple fistula, without any other abnormality of trachea and oesophagus. In 1917 Cautley collected six cases of single oesophago-tracheal fistula among the previously reported congenital abnormalities of the oesophagus. In 1919 Plass could only discover one case of fistula among 136 instances of oesophageal abnormalities.

In 1941 Gengenbach and Dobos published the necropsy report of a child of 3 months who, besides having an oesophago-tracheal fistula, had only a left kidney and a patent ductus arteriosus. Two other cases are reported by the same writers but without surgical or post-mortem confirmation.

In 1943 Hübner reported a case of double oesophago-tracheal communication in a man of 36 who had had a chronic pulmonary infection

and bronchiectasis since childhood, which the writer supposed to be of congenital origin.

In 1946 Patten quoted the anatomical facts of a case of Haight's when the diagnosis was made only at post-mortem examination. In 1948 Haight mentioned two cases quoted by Leven and Lannin, one case of Shaw's and one other of Barkley's. Brown and Brown published in 1950 two cases observed at Buffalo Children's Hospital, one in 1931 and the other in 1950. Cardullo and Berens in 1950 reported the case of a newborn child with a single fistula and spoke of the difficulties of diagnosis and anatomical facts. Fluss and Poppen (1951) quoted the anatomical observations of a baby of 8 months with simple oesophago-tracheal fistula who died of bronchial obstruction shortly after the operation.

We found some records of cases where surgical intervention was successful. The first one is that of Imperatori in 1939. This writer reports the history of a 6-year-old child with an oesophago-tracheal fistula who was treated successfully by a median cervical incision as for tracheotomy and closing of the fistula. In 1948 Haight reported the case of a boy of 6 years who had an oesophago-tracheal fistula cured by a right extrapleural approach. In 1951 Helmsworth and Pryles mentioned the case of a 6-month-old child with an oesophago-tracheal fistula which was 8 mm. in diameter; this was divided and closed through a thoracotomy.

In 1951 Knox, besides mentioning three cases of Gross's without any comment, reported his personal observations of a 14-day-old child in whom he found an oesophago-tracheal fistula without oesophageal atresia but with a cervical oesophageal diverticulum. Gastrostomy was performed on the twentieth day. At the age of 3 months a left cervical incision allowed closure of the fistula which was attached to the posterior wall of the trachea about 2 cm. above the carina. The oesophageal diverticulum was removed later by the same left cervical approach.

We also found in the literature three cases of congenital broncho-oesophageal fistula. In May, 1950, Morton, Osborne, and Klassen reported the case of a man of 26 who had a congenital right oesophago-bronchial fistula with bronchiectasis of the right lower lobe; this was treated by closing the fistula and a right lower lobectomy. In November, 1952, Berman, Test, and McArt noted a case of a woman of 40 who had a left oesophago-bronchial fistula with bronchiectasis of the left lower lobe and lingula; this was cured by closure of the fistula in the first instance and pneumon-

ectomy performed 18 days later at the same time as the ligation of a double patent ductus arteriosus. Lastly, in December, 1952, Boquien, Geffriaud and Cornet recorded the history of a girl of 16 who had bronchiectasis of the right lower lobe and a fistula between the oesophagus and a bronchiole of the right lower bronchus at the junction of the segment of Nelson with the basal pyramid. The closing of the fistula by a right thoracotomy effected a cure.

In all these cases the clinical picture is almost the same: difficulty in deglutition and a burst of coughing following the ingestion of foods, particularly of fluids. Pulmonary complications are evident more or less early in life. They appear from time to time in the history of the patient in whom the fistula is only diagnosed in adult life. To explain this tolerance of the fistula until adult life it is necessary to postulate the smallness of the fistula and its direction obliquely backwards and downwards with the oesophageal orifice at a lower level than that of the trachea; another explanation is the presence of a flap of mucous membrane able to prevent the passage of foods and fluids from the oesophagus to the trachea until adult life (Chevalier-Jackson) or the temporary presence of a thin membrane acting as a valve preventing pulmonary inspiration (Berman and others, 1952).

The diagnosis of oesophago-tracheal fistula may be confirmed and the site of the fistula decided after complementary examination. Lamb (1873), Haight (1948), and Helmsworth and Pryles (1951) pointed out the importance of the presence of an abnormal quantity of gas in the stomach and bowels on radiological examination. The use of barium, being excluded, the oesophageal swallow with lipiodol may show the fistula, and Holt, Haight, and Hodges (1946) emphasized the importance of an x-ray examination in the prone position. Cardullo and Berens (1950) said that it was necessary to use an aqueous fluid such as 35% solution of diotrast with a low viscosity with therefore every opportunity to enter into a small and oblique fistulous tract. Also, this solution has the advantage of being non-irritant to the bronchial mucous membrane, and it is absorbed within 24 hours, leaving, unlike the oily solutions, no trace in the respiratory tree. Nevertheless, interpretation of pictures obtained by injection of the respiratory tree is made more difficult by the possibility of tracheal inspiration following a quick regurgitation of the ingested opaque fluid, and this is most so because in these patients the pharyngeal phase of swallowing is frequently disordered. Oesophagoscopy must always be performed, but fol-

of oesophageal mucous membrane can conceal a minute orifice. Tracheo-bronchoscopy can demonstrate the fistula, especially if methylene blue is injected or instilled into the oesophagus, but this examination was negative in the cases of Cardullo and Berens.

As far as the associated malformations are concerned, it is possible to encounter oesophageal diverticulum (Knox), vertebral and costal abnormalities (Cardullo and Berens, personal case), cardiac malformations (interventricular communication in the observations of Cardullo and Berens), and abnormalities of the aortic arch or of its branches. As for the associated bronchiectasis, it is probably the result of the repeated bronchopulmonary infections suffered by these patients.

The diagnosis of a single oesophago-tracheal fistula indicates clearly the need for surgical intervention, and the pulmonary infection usually present is a formal indication, rather than a contra-indication, to surgical treatment (Haight, 1948). These single fistulae can be compared to the type III of Ladd's classification of congenital abnormalities of the oesophagus. In the cases of a high fistula, the cervical approach of Imperatori and Knox is successful. Haight employed the right extrapleural approach with resection of the posterior part of the fourth and fifth ribs. Helmsworth and Pryles (1951) performed a right thoracotomy in the fifth intercostal space in a 6-month-old baby. This postero-lateral thoracotomy seems to us the best approach, the choice of intercostal space depending upon the site of the fistula. After dissection and section of the fistula the orifices should be closed with interrupted stitches of nylon "000" or silk "00000." Haight, anticipating difficulties in closing the tracheal opening, cut near the oesophageal wall to preserve a muff of fistulous track adherent to the trachea. He closed the tracheal orifice in one layer and the oesophageal opening in two layers, and he interposed between the oesophagus and the trachea a pedunculated flap of mediastinal fat and cellular tissues. In our patient the difficulties were met in the suture of the oesophageal hole because of the lack of oesophageal musculature at that level, and the reintervention showed the presence of a little fistula which was obturated in two layers.

It is necessary to emphasize a point in relation to post-operative care: the difficulty in swallowing fluids can last for several weeks, as it did in our patient. This fact made us fear the possibility of reappearance, or of an underlying undiagnosed oesophago-tracheal fistula, emphasized because a barium swallow had shown a partial injection of

the right lower bronchus. Oesophagoscopy and tracheo-bronchoscopy and the quick, successful convalescence showed that there was no cause for anxiety. Knox (1951) reports similarly the case of his 4-month-old patient who for nearly two months found ingestion of fluids caused discomfort because he had to grow accustomed to swallowing without inspiring the fluid into the respiratory tract.

Finally a note must be made concerning the treatment of the accompanying bronchiectasis in adult patients. Osborne (Morton and others, 1950) performed a right lower lobectomy, Berman (Berman and others, 1952) a left pneumonectomy. In our case, we thought that the bronchiectasis of both lower lobes might decrease after the fistula, the cause of the chronic bronchial infection, had been closed. Follow-up proved we were right.

The second point in our patient, and no less exceptional, is the operative discovery of a tracheal diverticulum. We found in the literature one case of Arce published in October, 1943. In an adult with a hydatid cyst of the right upper lobe bronchoscopy revealed a normal carina, and at the level of the right main bronchus another complete sagittal ridge situated in front of the right upper bronchus. A catheter was introduced into the orifice which was thought to be that of aberrant bronchus and some lipiodol was injected. The lateral radiograph of the chest showed a shadow in the shape of a bubble with a horizontal level corresponding to a diverticulum of the main right bronchus.

The first anatomico-pathological observations were made by Rokitsansky, who wrote in 1838 on three cases of tracheal diverticula. The pathogenesis and anatomical pathology of these tracheal diverticula were described in two articles, one by Przewoski published in May, 1898, the other by Czyhlarz published in September, 1897. According to these papers it is necessary to distinguish between two kinds of tracheal diverticula. (1) The congenital diverticula are situated at the level of the bifurcation and of the main right bronchus, when, according to Aeby (1880), the trachea is actually divided into three. The structure of the walls of these diverticula is the same as that of the bronchi. According to Chiari they are diverticula developed at the level of supernumerary bronchi. (2) The acquired diverticula are, according to Rokitsansky, diverticula due to traction caused by hypertrophy of the mucus glands of the trachea with enlargement of the excretory duct. On the contrary, according to Przewoski (1898) and Czyhlarz (1897), they are diverticula

caused by pulsion from degeneration of the elastic and muscular fibres of the posterior wall of the trachea.

Przewoski classifies two kinds of acquired diverticula: (1) Circular diverticula with a wide opening, which have the appearance of a hollow spherical cavity occupying the greater part of the posterior wall of the trachea. Such diverticula have only been found in cases of old emphysema. They may be localized to the upper part of the posterior tracheal wall or they may occupy all this wall which is more or less atrophied and thinner. (2) Sacciform diverticula with a small neck and opening usually found at the level of the posterior right wall of the trachea at the junction of membranous and cartilaginous parts; they may be encountered also at the level of the main bronchi. They are of various sizes and perhaps the size of an egg. They may be multiple, the upper ones then being deepest. From the anatomico-pathological point of view, they are formed of the same morphological layers as the posterior tracheal wall. Przewoski, in post-mortem examinations of children and adults, never met a case without bronchopulmonary infection. A chronic infection of the respiratory tree is therefore the aetiological factor determining an endotracheal rise of pressure during bursts of coughing and changes in tracheo-bronchial walls.

In our patient, in spite of the fact that childhood was punctuated by many bronchopulmonary infections, it was probably a congenital diverticulum because there was only one situated at the level of the bifurcation and particularly because of its association with oesophago-tracheal, bony, and vascular abnormalities.

The final point of interest in our case is the existence of an abnormality of the thoracic duct, which was the cause of a right-sided post-operative chylothorax, the cure of which was effected by ligature at reintervention. It is not our purpose to emphasize here post-operative chylothoraces; a study of these abnormalities was made in France in the theses of Mouchet (1933) and Barbier (1951).

On the other hand, we have investigated the papers describing in detail the embryology and possible abnormalities of the thoracic duct. It is in the foetus of 24 mm. (seven or eight weeks' gestation) that the beginning of the upper left and right lymphatic ducts may be found, formed by two buds from the subclavian veins, the thoracic duct being constituted in the foetus of 30 mm.

Abnormalities of development may be the cause of complete reduplication, partial reduplication

(particularly frequent at the level of the middle part of the canal), or multiple reduplications giving the appearance of a plexus. Davis (1915) studied these malformations precisely, isolating nine anatomical types of which we noted that the first seven had an oblique transverse anastomosis at the top and to the right or the left as the case might be. The anastomosis was situated between the posterior face of the oesophagus and the spine. The various abnormalities are more frequent than the normal type.

The knowledge of these abnormalities of the sub-aortic part of the thoracic duct has a double interest: the eventual damage to the canal, as was the case in our patient, and the possible influence that such a vascular abnormality may have in the determination of the oesophago-tracheal malformation. The importance of vascular abnormalities in the pathogenesis of congenital oesophageal atresia and oesophago-tracheal fistula has been underlined. In 1902 Krauss, in four cases of oesophageal atresia, found three instances of an abnormal origin of the right subclavian artery. Keith and Spicer in 1907 reported three anatomical observations of oesophageal atresia associated with a right aortic arch, the left subclavian artery passing behind the trachea just below the upper end of the oesophagus in two cases, and the right subclavian artery passing between the two parts of the oesophagus in one case. Neuhauser (1946) wrote that he found an abnormality of the right subclavian artery in two of the eight cases of oesophago-tracheal fistula which he had the opportunity to examine. In 1951 Fluss and Poppen reported the results of post-mortem studies in three cases of oesophago-tracheal fistula, associated with atresia in two cases. The first patient had an aberrant right subclavian artery coming from the left subclavian, passing between the two parts of the oesophagus, behind the trachea. The second patient had an aberrant right subclavian artery coming from the aortic arch by a common opening with the left subclavian and passing between the two oesophageal parts together with an aortic coarctation and a patent ductus arteriosus. The third patient had on the posterior wall of the aorta near the origin of the first pair of intercostal arteries, the origin of an abnormal artery which passed in front of the spine, behind the oesophagus at the level of the fistula, and led to the second right intercostal space, this abnormality having passed unnoticed during operation.

In 1952 Berman and others reported a case of left-sided congenital oesophago-bronchial fistula in which two patent ducti were found, the larger

at the usual site, the second coming from the aorta immediately below the former near the site of the broncho-oesophageal fistula and ending in the lower branch of the left pulmonary artery.

In our patient we think that there was probably an abnormal thoracic duct of the type III or VII described by Davis (1915) with the transverse portion passing obliquely and to the right between the spine and the posterior wall of the oesophagus, constituting a lymphatic abnormality similar in disposition to the arterial abnormality of the third patient of Fluss and Poppen.

It remains to try to describe the eventual action of these vascular abnormalities. The lymphatic system appears after the circulatory system in the foetus of 24 to 30 mm. at about the stage at which the oesophagus and the trachea are completely separated and the repermeabilization of the oesophageal tract takes place. One of the vascular abnormalities described here could interfere, either by direct pressure or by causing deviation of the oesophago-tracheal septum, leading subsequently to the formation of a fistula by atrophy and pressure (Fluss and Poppen, 1951).

#### SUMMARY

A case is reported of a congenital oesophago-tracheal fistula without atresia with a tracheal diverticulum and an anatomical anomaly of the thoracic duct in a girl of 16 years.

The previously published cases of congenital oesophago-tracheal fistula without atresia are reviewed and their diagnosis and treatment discussed.

The aetiology of the tracheal diverticulum is discussed.

After a study of the anomalies of the upper part of the thoracic duct and a review of the vascular abnormalities associated with oesophageal atresia, the role of the lymphatic abnormality in the aetiology of the post-operative chylothorax and the pathogenesis of the oesophago-tracheal fistula is discussed.

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