

A CASE OF INTRATRACHEAL COLLOID STRUMA; OPERATION;  
RECOVERY.\*

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AMONG the problems that the throat specialist is called upon to solve there are few, perhaps, that present more difficulties than some of the conditions causing tracheal stenosis.

A large majority of these cases of tracheal stenosis are due to pressure on the trachea by tumors external to it, to old syphilitic adhesions or gummata, and to certain intratracheal new-growths, benign and malignant.

One of the rarest forms of intratracheal growth, usually causing great stenosis, and making operative procedures imperative on account of the alarming dyspnoea by which it is accompanied, is, without question, the tumor springing from the thyroid tissue in the lower laryngeal cavity and the upper part of the trachea, sometimes called an intralaryngeal or tracheal "accessory" thyroid tumor.

There are now on record, including the case which I have the pleasure of reporting to you this evening, ten positive and two doubtful cases of intralaryngeal and tracheal tumors, originating in thyroid tissue. Eight of these were observed in different parts of Germany.

The author's case is the second reported in the United States, and the tenth on record. The other case was reported by Freer, in the *Journal of the American Medical Association*, March 30, 1901. There is no doubt that other tumors of this nature, in which the diagnosis was not verified by operation and microscopical examination, have been observed, but a very thorough search of the literature failed to reveal the report of any other case (with the exception of Freer's) like the author's.

I may mention in passing, in order to show the rarity of this form of benign intratracheal growth, that Bruns,<sup>7</sup> up to 1898, had collected altogether ninety-one cases of benign tumors occurring in the trachea, and of these only seven were cases of intratracheal struma. Since then two others, those of Baurowics and of Freer, have been reported, making nine only on record.

There have been, of course, a considerable number of accessory thyroid glands and thyroid tumors reported, occurring in more or less remote

\* Presented to the Section on Laryngology, New York Academy of Medicine, January 22, 1902.

situations, viz.: the cases of intrathoracic struma, those that are sub-sternal in position and connected directly with the thyroid gland, the accessory thyroid tumors at the base of the tongue, the parathyroid glands found in the upper third of the anterior surface of each lobe of the thyroid, which Gley has found in almost all animals, and some in still other parts of the air passages. (Osler,<sup>10</sup> Gruher,<sup>11</sup> Gley,<sup>12</sup> Warrea,<sup>13</sup> Schadle,<sup>14</sup> Virchow, Wolf,<sup>15</sup> Butlin,<sup>16</sup> and others.) These cases are only mentioned here because they are of some interest in connection with the subject under discussion.

The history of the writer's case, which in some respects differs from any of the other recorded cases, is as follows:

Mrs. M. R., aged thirty-two years, was referred to me by Dr. J. B. Harvie, of Troy, N. Y. Her father died of some kidney trouble at the age of thirty-three years; her mother is living and well. Her two brothers and two sisters are living. One sister, it is interesting to note, has a goitre of considerable size. One sister and brother died in childhood. She had the usual diseases of childhood, but otherwise has always been well. Her menstruation commenced at the age of fourteen. She has always been regular, but has usually had considerable pain the first two days of each period. Her last menstruation was in June, 1901. She is now five months pregnant, and has two children, both of whom are living and in good health. About seven years ago the patient noticed a contraction of the muscles of the left side of the neck, and that the glands on the same side were swollen. Three weeks later she first noticed a difficulty in breathing, which was increased by the least exertion. The dyspnea, which had irregularly but steadily increased, particularly during her present pregnancy, finally became so alarming that she was sent to the Troy Hospital, November 12, 1901, by her physician, Dr. Harvie. I saw her, with Dr. Harvie, on the following day for the first time. She was breathing with considerable difficulty, and there was some cyanosis of the lips.

On examining the neck a goitre of moderate size was found, the left lobe of the thyroid being somewhat, and the isthmus decidedly, enlarged. There was a marked inspiratory stridor, and on palpation over the upper part of the trachea a decided inspiratory thrill could be felt, a symptom that appears to be pathognomonic of high tracheal stenosis. No other enlarged glands in the neck were found. The examination of the lungs was negative, with the exception of a somewhat prolonged expiratory murmur at the left apex. Other organs normal. An examination of the nose, nasopharynx, pharynx and larynx, showed normal conditions.

On laryngoscopic examination, however, a tumor of considerable size, springing from the posterior and left lateral walls of the trachea, almost completely filling its lumen, was seen during deep inspiration. (Fig. 1.) The growth appeared to extend from about the first tracheal ring downward along the posterior wall. The tumor was regular in outline, and covered with a perfectly smooth, normal-looking mucous membrane. Several vessels were seen running over its surface.

During the next few days iodide of potash was given, 30 grains every three hours. The dyspnea, however, becoming more serious every

day, an operation was considered imperative, and was performed November 16th.

Under ether, with the patient's head low, an incision extending from the lower margin of the thyroid cartilage to a short distance above the episternal notch was made. On account of the enlarged isthmus which covered the parts it was not considered wise to attempt to perform a preliminary low tracheotomy. The isthmus was found to be much enlarged, making the exposure of the trachea difficult. At this time the patient's breathing became very bad, and a high tracheotomy was performed, the isthmus being pushed down as much as possible. In cutting through the tracheal wall a small incision was accidentally made in the tumor, causing a most troublesome hemorrhage, which was finally controlled with a sterile solution of adrenalin chloride, 1:1000. The thyroid isthmus was then firmly ligated on both sides with catgut, and divided. The trachea being now thoroughly exposed a low tracheotomy was performed, the upper tube taken out, and the tracheal rings above the tube were split, as was also the cricoid. The tumor, which

FIG. 1.



was over 5 cm. long, extended from the first tracheal ring downward along the posterior wall, and almost filled the tracheal lumen. It was resilient to the touch, and was found to be firmly attached, by a very broad base, to the posterior and left lateral walls of the trachea, from which it could not be separated. It was, in fact, part of the tracheal wall. As much of the growth as possible was removed with snare and curette, the adrenalin solution being used continuously, on account of the severe and troublesome hemorrhage.

Because of the hemorrhage, and because the patient's condition was not entirely satisfactory, attempts to separate the portion of the tumor that remained from its attachment to the posterior wall had to be finally abandoned. The trachea was brought together with catgut sutures, the external wound being closed by a continuous subcutaneous silkworm-gut suture. A gauze tampon was inserted above the tracheotomy tube to prevent blood from oozing down.

Patient's condition was very satisfactory for several days after the operation, but on the fourth day her temperature, which had been nearly normal, suddenly went up to 102° F., and she developed a

double pneumonia. Nitroglycerin was administered, at first  $\frac{1}{60}$  grain every hour by mouth, with  $\frac{1}{30}$  grain of strychnine every three hours hypodermically. Whiskey was also given every hour. On the following night, November 21st, patient was in a very dangerous state, several times being in a condition of extreme collapse. Hypodermic injections of 5 minims of a 1 per cent. solution of nitroglycerin every twenty minutes during the collapse were each time followed by the most marked results, the pulse improving in quality within a few minutes after each hypodermic.

The next morning, November 22d, the patient again collapsed, respiration almost ceasing. It was evident that the trachea below the tube was filled up. The tube was taken out, the patient held over the side of the bed with the head very low, and with a long, narrow curved forceps I was finally able to remove from low down in the trachea several masses of almost solid mucus which nearly filled its lumen, and which she could not have expelled through the tube. The patient's breathing improved at once, and from this time on she steadily improved. For nearly two weeks, however, from the onset of the pneumonia her heart acted badly, the pulse being rather irregular, not of good quality, and never getting below 120. During this time nitroglycerin was administered continuously at hour intervals, with strychnine  $\frac{1}{30}$  grain hypodermically every four hours. The nitroglycerin had to be stopped at times for part of a day when the patient complained too much of throbbing and pain in the head, and digitalis was given instead, but I would like to mention at this time that I do not believe there is any doubt that the large amount of nitroglycerin the patient received, and particularly the large doses given hypodermically during collapse, saved her life. She was discharged from the hospital in excellent condition on the twenty-ninth day after the operation, still wearing the tracheotomy tube. She was at that time about six months pregnant.

The portions of the growth removed were sent to Dr. Blumer for examination, who reported as follows:

BENDER HYGIENIC LABORATORY,  
ALBANY, N. Y., December 21, 1901.

I enclose report on the tumor from the interior of the trachea:

The specimen submitted consists of ten small fragments of growth, averaging half a centimetre in diameter. The tissue is pinkish in color and somewhat translucent. Microscopical examination shows that covering the growth on one side is normal tracheal epithelium. Beneath this is submucosa, but tracheal glands are not present in the section, and presumably lie behind the tumor. The tumor itself is made up of tissue which may be described in brief as exaggerated thyroid tissue. The alveoli are in arrangement exactly similar to normal thyroid, and consist of spaces lined by cubical epithelium and containing quantities of colloid. They differ from normal thyroid in the great variation in size and shape of the alveoli. (Fig. 2.) The amount of connective tissue between the alveoli is about that normally seen in the thyroid gland. The connective tissue contains a large number of bloodvessels.

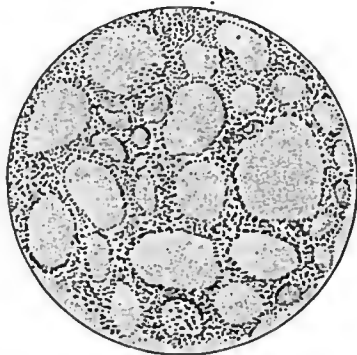
*Diagnosis.* Colloid struma, originating in thyroid tissue situated beneath the submucosa of the trachea.

GEORGE BLUMER.

It was not considered safe to permanently take out the tracheotomy tube until after her confinement, on account of the danger of a sudden swelling of the part of the tumor that had to be left. When the last laryngeal examination was made I found that this remnant had contracted considerably, so that she breathed without difficulty when the tracheotomy tube was temporarily removed, and I have no doubt that after she is confined it will almost disappear.

The origin of intralaryngeal and tracheal thyroid tumors was up to the time of Paltaut's investigations one of the most interesting enigmas in medicine, and for its solution it will be necessary to briefly review the reported cases.

FIG. 2.



Drawing of section; low power.

CASE I.—(Ziemssen's, 1875.) The patient, a shoemaker, aged thirty years, developed laryngeal dyspnoea during the latter part of October, 1875, which rapidly increased. He had a goitre of moderate size. A tracheotomy was performed for the relief of the dyspnoea. Death on the following day of erysipelas developing in the wound. At the autopsy a tumor was found in the larynx, left side, which extended from the middle of the cricoid cartilage, and was about 2 cm. long and 1 cm. thick. It had a perfectly smooth surface and an intact mucous membrane. Microscopical examination disclosed that the tumor was made up of thyroid tissue, and, also, that the goitre had penetrated on the left side, between the cricoid and thyroid cartilages, into the lower part of the larynx.

CASE II.—(Reported by Bruns.) The patient, a male, aged thirty-two years, had had since his seventeenth year difficulty in breathing, which had gradually increased. Examination of the neck showed a slight enlargement of both lobes of the thyroid. On laryngeal exam-

ination a tumor was seen in the lower part of the larynx, almost filling its lumen, and springing from the right lateral wall and part of the posterior wall. Operation, laryngo-tracheotomy. Growth extended from the fourth tracheal ring upward to close under the right vocal cord. On microscopical examination it was found to consist of thyroid tissue, with slight colloid degeneration.

CASE III.—(Heise-Bruns,<sup>3</sup> 1877.) Case of a girl, aged fifteen years, who for three years had had difficulty in breathing. Lateral lobes of the thyroid were not enlarged, but the isthmus could be felt as a node as large as a cherry. On laryngeal examination a subglottic tumor was seen on the posterior and right lateral walls of the larynx, slightly nodular, and covered with intact mucous membrane, taking up two-thirds of the lower laryngeal cavity. Operation, laryngo-tracheotomy. The tumor was found to extend from the second tracheal ring to close under the glottis, and had a very broad base. On microscopical examination it was found to consist of thyroid tissue.

CASE IV.—(Heise,<sup>4</sup> also from the Tübingen clinic, 1885.) The patient, a male, aged twenty-six years, had had slight dyspnoea after exertion, since his twenty-fourth year, which gradually got worse. The thyroid was not enlarged. Laryngeal examination, followed by tracheotomy, showed the presence of a tumor, 5 cm. long, extending from the first tracheal ring downward along the posterior tracheal wall. On microscopical examination the tumor was found to be made up of thyroid substance, showing slight colloid degeneration.

CASE V.—(Roth's,<sup>5</sup> Reported by Bruns, 1888.) Case of a woman, aged forty years. At the autopsy a small tumor, covered with mucous membrane, was found under the cricoid cartilage. On microscopical examination it was found to consist of small follicles with fatty degenerated epithelium.

CASE VI.—(Paltauf's,<sup>6</sup> 1892.) A young woman, aged twenty-nine years, was brought to Albert's clinic, in Vienna, on account of alarming dyspnoea, and an immediate tracheotomy was performed. Four weeks before difficulty in breathing had developed, and became alarming three days before the operation. Examination of the neck showed the presence of a struma of moderate size. On laryngeal examination several red tumors were seen under the vocal cords, nearly closing the laryngeal lumen. Three weeks after the operation a large abscess of the left thyroid lobe developed, death resulting a week later. At the autopsy a small tumor was found, extending from the lower border of the thyroid cartilage to the cricoid cartilage. Microscopical examination showed this to be typical thyroid tissue.

CASE VII.—(Bruns,<sup>7</sup> 1898.) Case of a girl, aged twenty-four years, who had a small median goitre. On laryngeal examination a tumor was seen on the anterior wall of the lower laryngeal cavity and upper part of the trachea. Operation, laryngo-tracheotomy. An adhesion of the isthmus to the trachea was found, and directly opposite in the trachea the tumor was situated. On microscopical examination it was found to be a struma colloides.

CASE VIII.—(Banrowicz's,<sup>8</sup> 1898.) Case of a girl, aged twenty-one years, who had had no operation for tracheotomy. Under the left vocal cord a tumor was seen attached to the left lateral and posterior walls, filling the lumen of the lower laryngeal cavity, and extending down to the fourth tracheal ring. Operation, laryngotomy. On microscopical

examination the growth was found to consist of thyroid tissue, with slight colloid degeneration.

CASE IX. (Freer's,\* 1901.)—This patient, a woman aged thirty-two years, had a thickening of the mucous membrane below the cords, forming a ledge which projected into the laryngeal lumen around its whole circumference. This thickening extended down the trachea to the third or fourth ring, forming a prominent tumor on the posterior wall. On examination of the neck the thyroid could be felt. A tracheotomy was performed on account of a sudden subglottic swelling. Portions of the intratracheal growth were afterward removed through the tracheal wound, and were found to be made up of thyroid tissue. An operation for the removal of the growth itself was not performed.

*Doubtful cases.* Radestock's<sup>17</sup> case was formerly included under this head. In his case a tumor was found low down in the trachea, at the entrance to the main bronchus, closing its lumen entirely. Since the publication of his case it has been claimed by Paltauf and Kolisko<sup>18</sup> that it was not a true case of intratracheal struma, but that the microscopical examinations proved it to be a case of adenoma. Radestock's case was undoubtedly similar to the one reported by Scheuer<sup>19</sup> in 1893. Scheuer's case also turned out to be a true adenoma on the posterior wall of the trachea, extending from the sixth to the eighth rings. The other doubtful case was reported by Ball.<sup>20</sup> In this there was a tumor filling the upper part of the larynx and complicating a goitre. The laryngeal tumor was probably malignant, although Semon<sup>21</sup> was of the opinion that it might have consisted of hypertrophied thyroid tissue. Mayer's case,<sup>22</sup> in which there was a struma nodule on the right wall of the trachea which had grown through the wall from a large struma, was also proved to be a malignant adenoma. For the reasons given, and because the clinical data were insufficient, Radestock and Ball's cases were not included in the above table. The statement made by Freer that his case is the *tenth* case on record is then not correct. His was the *ninth*. He also stated that five cases have been reported from Bruns' clinic. Only *four* have been reported from the Tübingen clinic, two by Bruns and two by Heise.

The two theories as to the origin of this most interesting form of tumor that have received the most consideration are those of Bruns and Paltauf. Bruns and Heise,<sup>2</sup> the chief exponents of the embryonal theory, held that in *intra-uterine* life a small accessory thyroid lobule from aberrant embryonic rudiments of the thyroid gland (*Aangeborene Verlagerung von Schilddrüsengewebe*), must have, in such cases, been present in the foetal larynx or trachea. This lobule, developing about the time of puberty, resulted in the true intralaryngeal or tracheal "accessory thyroid tumor." Paltauf<sup>23</sup> was able to prove in his case, by microscopical examinations, that this theory was not tenable. In his case there was a connection between the intratracheal growth and the thyroid gland externally. The thyroid was so firmly attached to the cricoid cartilage and upper three tracheal rings that it could not be separated from them. The space between the cricoid and first tracheal ring, and a portion of the external lateral lobe of the thyroid, which

were firmly adherent to the cartilage at this point, were examined microscopically, and he was able to prove positively that the thyroid tissue had penetrated (through the interstitial membranes) to the perichondrium and submucosa on the inner surface of the trachea, the cartilage itself remaining intact. So it is not necessary, as Heise thought, that in order for the thyroid tissue to penetrate into the trachea there must be a perforation, or at least a defect in the tracheal wall. Ziemssen,<sup>1</sup> too, observed that the struma had penetrated the lower cavity of the larynx between the cricoid and thyroid cartilages, and Baurowicz was able to demonstrate the same origin of the intratracheal growth in his case. This observation of Paltauf's is the first instance on record of normal thyroid gland tissue penetrating to the interior of the larynx and trachea, although Orth,<sup>2</sup> in his *Pathological Anatomy*, makes the statement, "that strumas, but particularly the malignant neoplasms, could penetrate into the air passages, *i. e.*, into the larynx."

The only way that this penetration of normal thyroid tissue in Paltauf's case can be explained is by the intense adhesion of the right and left lateral thyroid lobes to the upper tracheal rings and cricoid. They were really part of the tracheal wall. This probably took place during fetal development, although the thyroid tissue in all probability did not penetrate into the larynx until during extra-uterine life, and at a time when enlargements of the thyroid commonly take place—the period of puberty.

Paltauf's conclusions, that these intralaryngeal and tracheal tumors springing from thyroid tissue do not owe their origin to an intra-uterine deposit of thyroid tissue are of great interest. They should therefore not be called *accessory* thyroid tumors. They originate in extra-uterine life by penetration of the gland tissue between the cricoid and thyroid cartilages, between the cricoid and first tracheal ring, between the upper tracheal rings, and through the interstitial tracheal membrane itself, *from without*. When this occurs it should be considered as a direct extension of an enlarged thyroid gland—a parenchymatous struma. Proof of this assertion is that the thyroid gland has really grown fast to the cricoid cartilage, interstitial membranes, and upper tracheal rings. This abnormal adhesion can neither be explained by pressure, nor by an inflammatory process, but can only have occurred during the formation and development of the thyroid gland, and only in the way that the *fatal* gland, in these cases, must be united with the perichondrium of the cartilage and the interstitial membranes.

Gruber's<sup>3</sup> investigations, perhaps, would give additional support to Paltauf's theory. He found, after examinations of many Russians and Bohemians, that there was often an accessory or really an extra lobule extending downward from the lowest posterior margin of the lateral lobes and lying very close to the trachea. These extra lobules may



also lie in the crico-thyroid space. The only parallel cases that possibly give some support to Bruns' theory are those observed by Streckeisen.<sup>22</sup> He found on sections through the hyoid bone seven times the so-called glandulae intrahyoidea, that is, thyroid tissue was shut in the bone. He believes that this was shut in during the ossification period.

In my own case it could not be determined during the operation whether the left lateral thyroid lobe, which was enlarged, was adherent to the trachea or not. The isthmus was not, because in ligating it the ligatures could be readily passed between it and the trachea. It is a rational deduction, however, from the fact that the intratracheal growth was really part of the tracheal wall, and springing as it did from the posterior and left lateral wall, that in this case, too, the intratracheal tumor was really an offshoot from the left lateral thyroid lobe.

An analysis of the ten cases of this rare condition brings out a number of interesting points: (a) The location of the tumors in all the cases, with the exception of Bruns' last case, was characteristic. They were all situated in the lower part of the larynx and upper part of the trachea and attached to the lateral and posterior walls. Bruns' last case was the only exception to this rule, the growth being attached to the anterior wall. (b) This point decides another interesting fact before mentioned, that these tumors, for this reason, should not be called *accessory* thyroid tumors, because they are really offshoots from the thyroid, as proved in the majority of the cases by the adhesion of the thyroid to the tracheal wall, and in Pnltauf's case, microscopically, by the infiltration of the interstitial membranes with the follicles of the thyroid gland. (c) They were all observed early in life, from the fifteenth to the thirty-third year, with the exception of Roth's case (the fortieth year). (d) A goitre of moderate size was present in all except in one of Heise's and in Freer's case. (e) They occurred in both sexes—three in males, and seven (including the author's) in girls and young adult women. A special consideration of my own case reveals some additional interesting points: It is the only case of which I could find any record in which the intratracheal struma was present in a pregnant woman; it is the second case on record in which the struma was confined to the trachea, extending from the first ring downward, the other case being one of Heise's. It is one of the largest, if not the largest recorded intratracheal tumor of this kind, extending as it did down the posterior tracheal wall for a distance of over 5 cm., or nearly two inches. The fact that it was present in a pregnant woman suggests another rather interesting connection between the intratracheal struma and pregnancy. I do not think there is much doubt that the pregnant condition had a great influence on the development and increase in size of both the extratracheal and intratracheal struma, because the dyspnoea was not nearly

so great before she became pregnant. It is well known that the thyroid frequently enlarges during pregnancy to cope with the increased metabolic changes of that period. Freund,<sup>11</sup> in his classical article on the relations between the thyroid gland and the female genital organs, brings out the fact that the gland is frequently enlarged in pregnant women, and that it almost always increases markedly in size during the birth of the child. This was true in forty-five out of fifty women examined by him. Tait<sup>12</sup> found an enlargement of the thyroid body in twenty women who had borne children. In his cases the increase in size developed during the latter months of pregnancy, and subsided very much after delivery. Wölfler<sup>13</sup> has shown that in the normal thyroid gland embryonic masses of thyroid cells are found here and there, particularly toward the periphery of the gland, and these, under certain stimuli, take on a kind of normal growth. Such a change occurs during pregnancy.

**PROGNOSIS.** The prognosis, on the whole, may be said to be favorable. Tumors of this nature, when completely removed, do not appear to have a tendency to recur.

**TREATMENT.** When the growth is large enough to seriously interfere with respiration a laryngo-tracheotomy for its removal should be performed as soon as possible, on account of the peculiar capacity tumors of this kind have for increasing very suddenly in size. I could not find any record of internal thyroid medication being used for intratracheal struma, but should think that it might be of value in certain cases.

**CONCLUSIONS.** 1. Cases of intratracheal struma are, perhaps, not so rare as the few cases on record would indicate. If laryngeal and, as far as possible, tracheal examinations of all cases of goitre were made, particularly when the thyroid is only *slightly* enlarged and the dyspnoea is severe, more cases would probably be discovered.

2. The term *accessory*, when applied to such tumors, is *not* correct, because they really are "off-shoots" of the thyroid gland.

3. Pregnancy, as in the writer's case, undoubtedly has a great influence on the development of the intratracheal struma.

I wish to take this opportunity to express my thanks to Dr. Harvie for his valuable assistance at the operation, and also to my clinical assistant, Dr. Fairweather, for so carefully following out the after-treatment of the case.

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## EXENCEPHALIC (INIENCEPHALIC?) MONSTER WITH BILATERAL HARELIP AND CLEFT PALATE.<sup>1</sup>

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THE monster which I wish to present this evening is a seven months' white female foetus which was stillborn, being the first child of an extremely neurasthenic woman of seventeen and a half years, whose family history I cannot obtain, but whose pregnancy up to the time of the miscarriage was uneventful. The father is about twenty-five years of age, healthy and robust, but having a strong family history of tuberculosis, his father having died of phthisis, and his mother is now suffering with the same disease.

About one week before the miscarriage the mother fell directly forward upon her abdomen, followed by severe lumbar pain and uterine

<sup>1</sup> Read at the meeting of the Philadelphia Pathological Society, held February 13, 1902.