

HEMANGIOMA OF THE LARYNX.* **

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Although telangiectatic growths are not infrequently seen in the larynx, true hemangioma of the larynx is a fairly rare tumor. The literature has been reviewed in recent years by Phillips and Ruh,¹ by Mayer,² and by New and Clark.³ Fifty-five cases of angioma, including eight of lymphangioma, were included in the analytical table published by the last named authors in December, 1919. I have found no reports of later date, though my search of the literature has disclosed a number of cases not included in any of the above reviews.***

The case herewith reported is unique in some respects, and suggests rather important conclusions when studied together with the heretofore reported cases. My case is especially noteworthy because of the age of the patient, the type and situation of the growth, and the peculiar kind of hemorrhage.

History of the Case: Baby boy, L. L., was born prematurely at about eight months on April 20, 1920, and died on June 21, at the age of 62 days. The father and mother are living. This was the only child. He appeared to be normal for some days. At the age of nine days he had a series of attacks of cyanosis with poor respiration, tachycardia, and cardiac arrhythmia. Administration of oxygen accomplished relief. A feeding of three-quarters of an ounce of breast milk was regurgitated, deeply colored with red; this I presume to have been blood. The baby was unable to swallow the next two feedings given at three-hour intervals. From that time improvement was continuous, and he left the hospital six days later, gaining in weight and nursing from the breast regularly. There had been no fever.

About five days before death the family noticed that the baby had some difficulty in breathing. At first the family physician considered asthma, but later sent the patient to the Minneapolis Gen-

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***Since presentation of my paper, an article on "Angeliomata of the Larynx," by Irwin Moore of London has appeared in *The Journal of Laryngology and Otology* for January and February, 1921. To his review of 73 cases should be added the case reported by Fallas (11), as well as the one herein reported.

eral Hospital as a case of possible laryngeal diphtheria. On admission at 10:15 p. m., June 21, 1920, physical examination showed slight general cyanosis, dyspnea, and marked retraction of the supra-clavicular spaces on inspiration. The temperature was 100° F. by rectum. A nose and throat culture was taken, but no *B. diphtheriae* found.

Intubation was attempted; the tube passed easily into the pharynx but could not be introduced into the larynx. As the posterior pharyngeal wall seemed to bulge slightly, a few small incisions were made in the mucosa there, but no pus was obtained. Dyspnea grew rapidly worse, and the patient died at 10:40 of the same evening. I was called upon to perform a post-mortem the following morning.

Autopsy Findings: The body is that of a well developed male infant, 55 c.m. long and of about 3000 grams weight. There is no edema, cyanosis or jaundice. There are no angiomas to be seen in the skin. The serous cavities are free from excess fluid and adhesions. The stomach contains some clotted blood (presumably from the incisions in the pharyngeal wall). The liver weighs 140 grams; its cut surface is light yellowish brown, and microscopic section stained with sudan iii shows a diffuse extreme fatty change in the liver cells. There is no other noteworthy pathology in the abdomen.

The heart is normal, except for three very minute bright red nodules at the free margin of the mitral valve leaflets. The ductus arteriosus barely admits a small probe. The aorta is normal. The lungs are partially collapsed; they show neither hemorrhages nor pneumonia. The bronchi and trachea appear normal on gross examination. The thymus weighs 12 grams and shows no lesion. The tongue, tonsils, soft palate and esophagus are normal. There is some clotted blood in the tissue posterior to the oral pharynx, and in the mucous membrane there are several fresh very short incisions. No purulent exudate is found.

On opening the larynx and trachea longitudinally through the posterior wall, one sees a diffuse, flattened, grayish purple bulging of the mucous membrane within the circle of the cricoid cartilage. This bulging has its upper border at a level two to three m.m. below the vocal cords and extends downward therefrom for a distance of 5 m.m. to the lower border of the cricoid cartilage. It occupies the entire circumference of the larynx with the exception of a narrow zone at the posterior commissure. The greatest thickness is in each lateral wall. The epithelium is not ulcerated, but shows a little loss of sheen in two or three places. Cross-section

of the larynx through the cricoid cartilage (see Fig. 1) shows a flattened mass lying between the cartilage and the lining epithelium. The cut surface of the mass is mottled dark red and gray and has a slightly granular appearance. A zone directly adjoining the cartilage at each side is dark red without the mottling of gray. The thickness from epithelium to cartilage on the right side is 3 m.m., and on the left side 2 m.m. The whole space within the cricoid cartilage at a level 5 m.m. below the vocal cords measures 5 m.m. transversely and 7 m.m. anteroposteriorly. The lumen of the larynx near that level appears to be entirely closed, and a little lower it forms a triangular chink measuring 0.5 m.m. transversely by 1.5 anteroposteriorly.

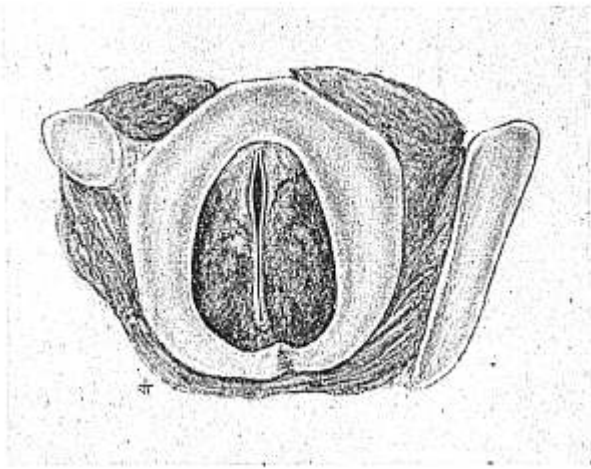


Fig. 1. Drawing of cross section of larynx through tumor. Ring of cricoid cartilage shown; also parts of thyroid cartilage and laryngeal muscles. Closure of lumen of larynx is shown to be due to compression by tumor. Darkest zone just inside cartilage is the intramural hemorrhage.

Microscopic Examination: The epithelium in some places is normal; but in other places it is thin. There the superficial layers are absent, and the cells of the remaining layers are shrunk, with pyknotic nuclei and deeply eosin-staining cytoplasm. Directly beneath the epithelium there is a narrow zone of dense fibrous tissue which appears normal except for a little dilatation of the blood vessels. More deeply there is a large abnormal area involving all the circumference excepting the posterior commissure and composed of closely-set large and small new blood channels with prominent endothelium. In some portions these channels are cavernous, in others they are quite small. The delicate stroma between the channels is a very young type of reticular connective tissue (see Figs.

2, 3 and 4). Between the blood spaces are scattered numerous mucous glands. These are in an active state; that is, the cells are filled with light-staining mucus and the nuclei are flattened peripherally (Fig. 4). The tumor laterally is separated from the cartilage by a zone of quite fresh but certainly antemortem hemorrhage. In places the hemorrhage extends into the tumor, but for the most part is in the meshes of a fibrous connective tissue of normal appearance adjoining the cartilage.

The condition found in this case corresponds in every respect to a true hemangioma. The large new formed blood vessels with very thin walls of prominent endothelium are quite characteristic, as is the delicate stroma of primitive reticulum. Such structure be-

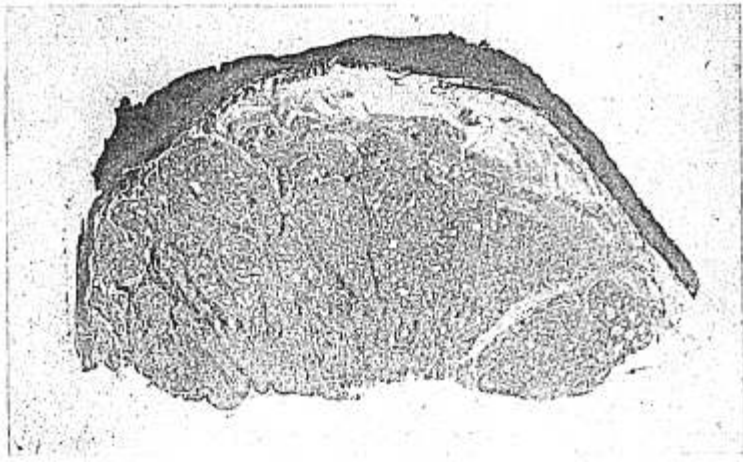


Fig. 2. Half of cross section of larynx through cricoid cartilage. Microphotograph, low power (X 15). Margin of cartilage is dark stained semicircle above; mucous membrane shows below. The larger blood spaces of the tumor can be clearly seen.

longs to a true neoplasm rather than to a telangiectasis. Active growth is indicated in this case by the absence of encapsulation.

The structure of the tumor in my case is remarkably similar to that in the case described by Phillips and Ruh.

Hemangiomas are congenital. They increase in size, as a rule, for some time after birth, and then usually tend to become encapsulated and to remain stationary⁴. In my case the tumor must have been growing since birth and the lumen of the larynx must have been markedly reduced by the time the parents called their physician for the child's dyspnea. At the time of the attempted intubation the tumor had almost entirely filled the lower part of

the larynx. It seems probable that the hemorrhage into the tissue within the unexpandible ring of the cricoid cartilage was sufficient to close completely the already markedly constricted air passage.

To Summarize the Case: An infant, aged eight weeks, became markedly dyspneic, the dyspnea increasing progressively until within five days an intubation was attempted as a last resort. Death followed quite promptly from suffocation. An autopsy, a hemangioma was found almost filling the ring of the cricoid cartilage. In addition, an intramural hemorrhage at the same place completed the closure of the air passage.

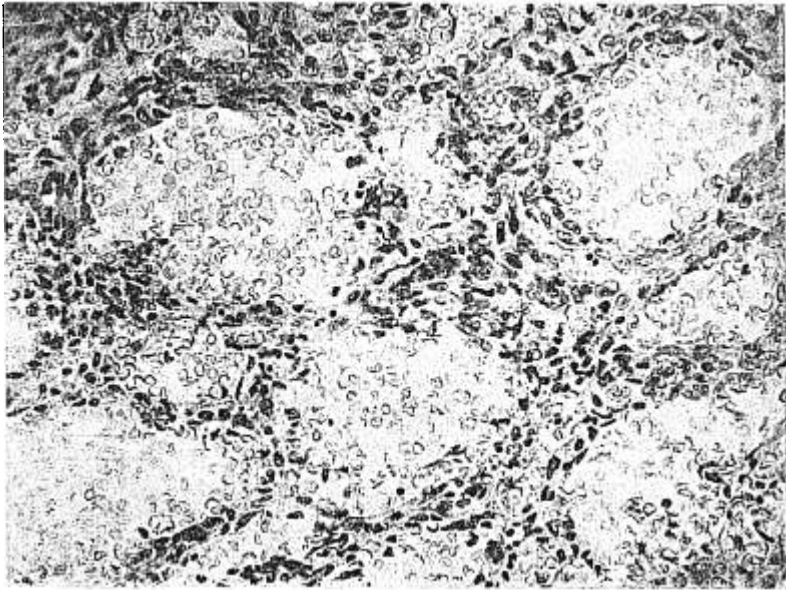


Fig. 3. Section of tumor. Microphotograph, high power. Shows some of the larger blood channels and a small amount of reticular tissue.

Discussion: In a search of the literature I have found reports of 25 hemangiomas in people more than twenty years of age, one in a girl 19 years old, one in a boy 13 years old, one in a boy 6 years old and three in infants less than one year old. Mine is the fourth in a child less than a year old, and the patient is, to the best of my knowledge, the youngest mentioned thus far.

Of the 38 cases of hemangioma, my own included, from which I have a record of sex, 66 per cent have been in males.

A study of the situations and characteristics of hemangiomas of the larynx leads to a rather striking conclusion not heretofore noted:

it appears to me that the laryngeal hemangiomas seen in adults are very different from those found in infants and children.

Among the 48 cases, including my own, in which the situation of the tumor is given, 40 have their attachment upon or above the true vocal cords, and are not attached to the laryngeal wall below the cords. At least 38 of these 40 patients were more than twenty years old when first examined, and the age of the other two is unknown. Among the same 40 patients, all the tumors described macroscopically, with one possible exception, were sharply projecting, raspberry-like or smoothly rounded growth. Some were peduncu-

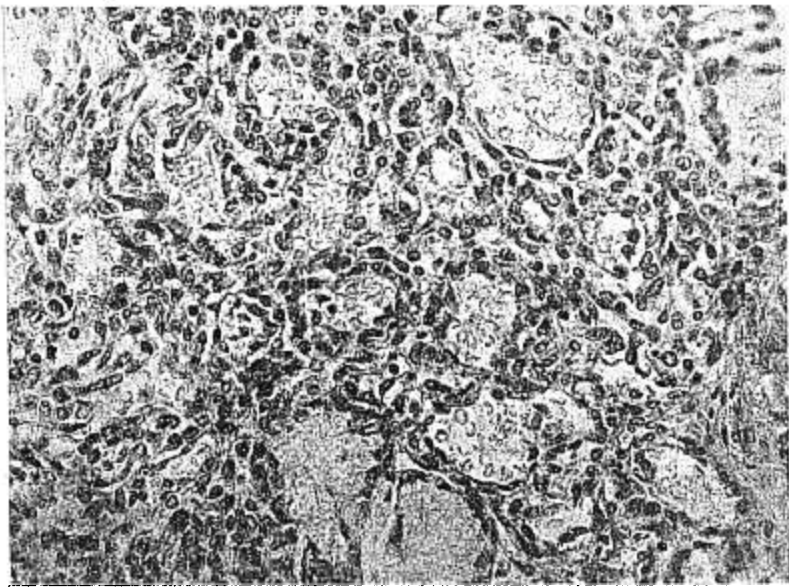


Fig. 4. Section of tumor. Microphotograph, high power. Shows smaller blood channels with more prominent endothelium, and some primitive reticular connective tissue. Below is seen part of one of the mucous glands.

lated, and some had broader bases, but all were sharply demarcated, projecting growths. Hoarseness was a predominant symptom; dyspnea was relatively unimportant. The above series I conceive to represent a definite type of tumor found in adults and not to be confused with a different type found in infants.

In 8 of the 48 cases in which the situation of the tumor was noted, the attachment has been entirely below the true vocal cords. Two of the patients were adults (one seen by Martuscelli and Porfidia⁵ and one by McKinney⁶), but in them the tumor was pedunculated

and in gross appearance otherwise resembled the type seen in adults and described above. The age of a third patient⁷ is not given. In the case of one boy 6 years old with a subglottic angioma,⁸ the description of the tumor is not clear. One boy 13 years old⁹ showed a subglottic cavernous angioma so large as practically to fill the larynx and cause severe dyspnea. The other three cases were of infants 9 months,¹ 9 months³ and 2 months old (my case), respectively. All these showed subglottic tumors, large in comparison to the size of the larynx, sessile in attachment, flattened, and somewhat diffuse. Dyspnea was the prominent symptom.

It is worthy of note that true laryngeal hemangiomas are exceedingly rare in infancy, and that they are all subglottic and sessile, and cause serious interference with respiration. On the other hand, true laryngeal angiomas are less rare in adult life, are found almost always upon or above the vocal cords, and are distinctly rounded, projecting, and sometimes pedunculated tumors; they cause relatively little dyspnea, but rather hoarseness of voice.

The following two cases have been excluded from the above classification. Levbarg's¹⁰ case of angioma in an infant ten weeks old is not included because the tumor seems to have been primary in the mouth and pharynx and to have involved the larynx only secondarily by direct extension. In a recent letter from Dr. Levbarg, he said: "I do not think that the growth originated primarily in the larynx. Upon direct laryngoscopy the tumor was slightly elevated, but spread out profusely, involving first the region above the vocal cords and then subsequently spreading below the vocal cords." A case reported by Fallas of Brussels,¹¹ would also hardly be placed in either of the types described, as the involvement included the arytenoids, false cords, left true vocal cord and subglottic region as well as the skin of the neck and possibly the lining of the pharynx.

The danger of hemorrhage has been repeatedly emphasized in the literature. Desvernine,¹² in 1888, reported a case giving occasional spontaneous bleeding from the throat, and at one time a severe hemoptysis. Wolfenden¹³ said that laryngeal angiomas occasionally lead to recurrent hemorrhage. Ferreri,¹⁴ in 1888, told of a case of angioma on the left vocal cord, which he crushed with a forceps. Alarming hemorrhage occurred at night and was controlled with difficulty by applications of ferric chlorid. Tracheotomy was performed, but another violent hemorrhage took place at that time and, although it was controlled, the patient died from pneumonia within 48 hours. Edmund Meyer,¹⁵ in 1904, strongly advised thy-

rotomy as the operation of choice in a child, because "even if intra-laryngeal operation were successful in removing the tumor, the hemorrhage would prove fatal." Ryerson¹⁰ tells of Shurly's case in which tracheotomy was performed to relieve dyspnea. "The growth entirely encircled the lower laryngeal and upper tracheal region. In opening the trachea, the growth was incised, which gave rise to uncontrollable hemorrhage, which resulted in the death of the patient." Phillips and Ruh¹ state that serious hemorrhage may occur. Mayer² emphasizes the danger of death from hemorrhage if a part or all of the tumor is removed intra-laryngeally. New and Clark³ have also emphasized the danger from hemorrhage. Thus, it is seen that there is a real danger of hemorrhage either into the lumen of the larynx or into a laryngotomy or tracheotomy wound.

My case emphasizes the danger of hemorrhage of a different sort. Here there is a hemorrhage into the tissue within the ring of the cricoid cartilage. Such a complication must be very rare, as my search of the literature has not revealed any similar case. The danger in such a condition is not from the loss of blood, but from the interference with the passage of air through the larynx, resulting in suffocation. The danger from this intramural hemorrhage is, of course, much greater in the sessile subglottic type of angioma found in infants than in the type found in adults.

Treatment: When possible, radium application is, without doubt, the treatment of choice for angioma of the larynx, as for angioma of other parts of the body. Ryerson¹⁰ first used it with success, and New and Clark³ have more recently used it with good results. Surgical removal of the tumor either intra-laryngeally or through laryngotomy is a distinctly dangerous operation. Laryngotomy seems to have been less dangerous than intra-laryngeal removal of the tumor.

However, an operation for the relief of dyspnea may be necessary in infants as an emergency measure. Intubation has been advocated by some, tracheotomy by others. Intubation was successfully performed by Levbarg¹⁰ and by New and Clark. On the other hand, the latter authors state that "intubation in such cases is not safe, because the trauma associated with such procedure is likely to produce severe hemorrhage." In the case herewith reported, it seems that an already severe constriction of the larynx was increased somewhat by hemorrhage into the tissue at the time of the attempted intubation, and the death probably somewhat hastened thereby. Mayer has, on various occasions, strongly advised against any intra-laryngeal operation. That tracheotomy, on the other hand, is not

free from danger is shown by the cases of Shurly¹⁰ and Phillips and Ruh,¹ in which the tumors extended into the upper part of the trachea (in the latter case, the tumor extended to the third tracheal ring). In Shurly's case, death followed incision of the tumor during tracheotomy. When tracheotomy must be performed it would seem that it should be as low as possible and with adequate preparation made in advance for the prompt control of a possible profuse hemorrhage. In the case I am reporting, I do not see how a diagnosis of angioma could have been made before the autopsy. Intubation was the logical treatment. The only possible lesson that could be learned is that in obscure cases one should be prepared to do a low tracheotomy immediately, should intubation fail to relieve the dyspnea.

SUMMARY.

1. Hemangioma of the larynx is rare, and especially rare in children.
2. Males are affected twice as frequently as females.
3. Two types should be recognized: (a) An adult type, distinctly rounded and projecting, often pedunculated, occurring almost always upon or above the vocal cords, and characterized clinically by hoarseness of voice with only occasional slight dyspnea. (b) An infantile type, sessile in attachment and usually rather flattened and diffuse, occurring always below the vocal cords and characterized clinically by serious interference with respiration.
4. Hemorrhage is the most serious complication; it may occur spontaneously, but is usually the result of operative interference. An intramural hemorrhage may occur, especially in the infantile type, and may cause sudden death by suffocation.
5. Radium treatment is indicated as the only safe method for the cure of the condition.
6. As an emergency measure for the relief of dyspnea, most authors state that tracheotomy is preferred rather than intubation, though both are dangerous. Tracheotomy should be as low as possible, and every preparation should be made for the checking of any profuse hemorrhage.

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