EPIGLOTTIDECTOMY FOR THE RELIEF OF CONGENITAL LARYNGEAL STRIDOR, WITH REPORT OF A CASE.*

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Congenital laryngeal stridor is a rare condition, characterized by its onset shortly after birth and by a peculiar inspiratory obstruction associated with a crowing sound. The stridor usually disappears during the second year of life. Dyspnea is usually absent, but there may be considerable inspiratory retraction of the lower thorax and at the sternal notch. In severe cases, asphyxial attacks may occur and may at times be so severe as to cause death.

Thus Reardon,¹ in reviewing 101 cases found that "in seven cases death seemed directly due to the stenosis." While Blackader and Muckleston² have found that one-sixth of the cases reported have died from affections of the respiratory system.

According to Thomson and Turner,³ the infantile epiglottis is characterized by its soft non-resistant texture and by more or less approximation of its lateral edges and of the ary-epiglottic folds. This gives the epiglottis a peculiar trough or gutter-like shape. As the child grows the epiglottis gradually becomes more rigid and elevated and tends to unfold, so that by the ninth year it begins to assume the form of the adult epiglottis.

In cases of congenital stridor most observers have found an exaggeration of the infantile type of epiglottis. In such cases, upon examination, the epiglottis can be seen to tilt backward, during inspiration, while its lateral borders and the aryfolds frequently come into contact. The upper aperture of the larynx may thus be reduced to slit-like orifice with a rhomboid opening just anterior to the arytenoids. The stridor is probably due to the vibration of the aryfolds and the borders of the epiglottis.

Post-mortem examination of the larynx from stridor cases shows that this deformity persists even after death, and it seems most likely that the malformed epiglottis is the cause of the symptoms. Jackson⁴ states that occasionally the epiglottis of normal infants has a tendency to roll inward during inspiration but considers the stridor cases a pathological exaggeration of this phenomenon.

Lees⁵ was among the first to demonstrate such a larynx. The illustration of this specimen shows the epiglottis markedly in-

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curvated with its lateral halves in contact, and with the laryngeal vestibule reduced to a mere slit. Variot⁶ demonstrated a similar specimen in 1898.

Judging from the illustration of Refslund's⁷ case the pathological anatomy was almost identical with Lees' case. The autopsy in Koplik's⁸ case, in a one-year-old child, which died of pneumonia showed a pronounced infolding of the epiglottis with its edges in contact. The lumen of the larynx seemed narrower than normal. An enlarged thymus was also found in this case. Variot⁹ has recently found some abnormalities in the muscles of the larynx of a child which had presented symptoms of stridor during life. The right crico-arytenoideus posticus muscle was entirely wanting, and there was slight atrophy of the vocal cord.

Report of Case: About five months ago a pronounced case of laryngeal stridor came under my care. The case is of special interest because the infant was born in the hospital, where it was carefully observed by Dr. Blackfan and Dr. Higgins, of the Pediatric Service. Further interest is added by the fact that the infant had an enlarged thymus to which the symptoms were at first attributed. The thymus was treated by Dr. Little with the X-ray and as a result diminished markedly in size, but the symptoms persisted.

The following abstract of the history prior to the time I saw the case is quoted from Dr. Blackfan's¹⁰ records: "A colored female infant weighed six pounds at birth. (Born in the Cincinnati General Hospital, March 29, 1921.) Immediately following delivery it was observed that there was difficulty in breathing. There was marked inspiratory stridor and cyanosis. The dyspnea was constant and from time to time it became paroxysmal; being increased by crying and nursing. The paroxysms would begin with a series of inspiratory efforts followed by a loud crowing inspiratory sound and then followed by a short expiratory effort. During this there was marked retraction of the supra and infra-clavicular spaces and the intercostal spaces. There was an anxious expression of the infant, the eyeball became prominent and there was extreme cyanosis. On several occasions the attacks were so severe as to require artificial respiration.

The physical examination was negative except for an area of increased dullness in the region of the thymus. The dullness extended one-and-a-quarter inches to the left and seven-eighths of an inch to the right of the mid-sternal line.

It was continuous below with the cardiac dullness. This area of dullness disappeared when the infant's head was held in extreme

dorsal flexion. It was also noticed that the symptoms were markedly relieved when this position was assumed. It was repeatedly observed that the symptoms could be induced and relieved by the position of the infant's head. When first seen it was believed from the increased area of dullness that the symptoms were due to an enlarged thymus. The possibility of congenital malformation of the larynx was however considered. In as much as the X-ray picture showed a shadow in the upper mediastinum indicative of an enlarged thymus a series of X-ray treatments were begun. Although no marked improvement in the symptoms followed this treatment the shadow in the X-ray entirely disappeared. In as much as the infant was being breast fed and the paroxysms were less frequent, the patient was discharged after one month," with instructions that it was to be brought to the dispensary at regular intervals.

Ten days later, on May 4, 1921, while in the dispensary the child suddenly became cyanotic, ceased breathing and required artificial respiration stimulation to restore it (Dr. Higgins). On the next day I noted the following: Voice normal, as manifested by crying. Respiration interrupted at times by laryngeal stridor with supra and infraclavicular retractions and girdle-like retraction of the lower ribs. On direct examination of the larynx, the free borders of the epiglottis are drawn together at each inspiration and come into contact; at the same time, the epiglottis is drawn backward into the larynx. The epiglottis appears to be flabby in consistence. The epiglottis closes like a book during inspiration. Arytenoids and vocal cords appear normal in structure and motion.

Epiglottidectomy: On May 6, 1921, without anesthesia, I suspended the patient, using a very short spatula made overnight especially for this case by Dr. Henry Freiberg. The epiglottis was exposed by the suspension laryngoscope. The tip of the epiglottis was seized with a small alligator forceps and a nasal snare was passed over the epiglottis, severing the free portion. There was practically no bleeding. It was rather soft and flabby but showed no gross anomalies. (Histological examination of the specimen by Dr. Austin showed nothing abnormal in structure.) The child was able to swallow in a normal manner on the same day and thereafter. The stridor, however, did not disappear immediately, but recurred at times for about twelve days after the operation, after which it was no longer observed. No asphyxial attacks occurred after the operation.

Five months after the operation the child breathes in a perfectly normal manner, when quiet, but shows a slight retraction

of the lower ribs when it cries loudly. The mother states that it has never had any difficulty in swallowing and she has not noticed any stridor except when the child has a cold. A very small epiglottic stump can be made out on direct laryngoscopy.

Comment: Since most observers agree that congenital stridor is due to a valve-like infolding of the epiglottis it seems to be a rational procedure to remove the epiglottis in all cases subject to asphyxial attacks. As is well known the epiglottis can readily be dispensed with in older patients and its removal apparently has no influence on deglutition even in infants. The persistence of some stridor for several days after the operation described above is difficult to explain, but may have been due to inflammatory reaction; or the flopping of what was left of the ary-epiglottic folds after the epiglottidectomy; or to some weakness in the laryngeal skeleton. At any rate the stridor disappeared much sooner than could have been expected in the natural course of the disease.

As an alternative to epiglottidectomy tracheotomy or intubation must be considered. Epiglottidectomy is a much simpler operation than tracheotomy with less risk to the patient, while intubation even if efficacious would require the wearing of a tube for many months.

So far as I can ascertain, epiglottidectomy has not hitherto been performed to relieve congenital laryngeal stridor. A review of the literature for the past twenty-five years apparently contains no record of such an operation for this condition. (American Institute of Medicine). This is more to be wondered at since Variot," in 1898, suggested that cutting away the ary-epiglottic folds might relieve this condition; and Reardon, op. cit. in 1907, suggested that the removal of the incurvated portion of the epiglottis probably would give relief if the removal of the epiglottic folds did not suffice.

BIBLIOGRAPHY.

1. REARDON, T. J.: Amer. Jour. of Med. Sciences, 1907, pp. 242-52.
2. BLACKADER and MUCKLESTON: Arch. of Pediat., 1909, v. 26, pp. 401-16.
3. THOMSON and TURNER: British Med. Jour., vol. 2, 1900.
4. JACKSON, CHEVALIER: Peroral Endoscopy, 189.
5. LEES, B. B.: Transact. of Path. Soc., London, 1883, vol. XXXIV, p. 19.
6. VARIOT, G.: Bull. et. Mem. Soc. Med. de Hop. de Par., 1898, 35, .vol. 40.
7. REFSLUND (quoted by Thomson and Turner), op. cit.
8. KOPLIK, H.: Arch. of Pediat., 1905, v. 22, pp. 881-89.
9. VARIOT, G.: Arch. de Med. enf., 1917, v. 20, pp. 530-34.
10. BLACKFAN, K. D., and LITTLE, K.: To appear in Amer. Jour. of Chil. Disease. 1921-22.