LARYNGEAL PAPILLOMATA IN CHILDREN

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PAPILLOMATA are the commonest tumours of the larynx in children and although they are benign in their histological appearance, they are malignant in their rapidity of growth, their tendency to recur, their ominous sequelæ, and in many cases, their lethal termination. Jackson[1] states that these tumours are the commonest of all benign growths of the larynx. According to Crowe and Breitstein,[2] the mortality rate from papillomata of the larynx in children under five years of age exceeds that of laryngeal carcinoma in adults. In children these growths tend to be multiple and may arise from the vocal cords or the mucosa of the larynx, trachea or epiglottis. As in carcinoma of the larynx or bronchogenic carcinoma, diagnosis must be early if treatment is to prove effective.

The incidence of laryngeal papillomata in children seems to vary. Some authors report it to be approximately one case per 1,000 patients, but Ferguson and Scott[13] in reporting 15 cases over a period of nine years state the incidence to be approximately one case in 6,000 clinic children. Jackson[1] reports having treated 200 cases over a period of many years. The onset is, it appears, most frequently between the ages of fifteen months and four years, but may occur at any age. It is reported that approximately 20% of these cases are congenital. The sex incidence is apparently not definitely established nor agreed upon by authors, some authors stating it is more common in girls and others that it is more common in boys. It is interesting to note that all the cases reported by Crowe and Breitstein[2] were boys.

The specific etiology of the papilloma still remains a mystery although many theories have been advanced. In a review of the literature there is no suggestion that environment may be a factor in this benign lesion. These tumours when seen through the laryngoscope present a characteristic picture. In children, they are usually multiple, glistening, grayish-pink in colour, mulberry or cauliflower-shaped on a short stalk with a fine nodular surface, and of firm consistency distinctly papillary in structure.

The vocal cords are the most frequent site of origin, but if extensive, the anterior commissure and epiglottis are usually involved. That they may extend to the trachea and bronchi is shown by Patterson,[8] who reported a case in which bronchoscopic removal from the trachea and left bronchus was necessary. Holding[3] reported a case of extension of the growth beyond the bifurcation of the trachea, resulting in death. Aerial metastasis to the lungs does occur and was reported by Hitz and Oesterlin.[5] In their case, autopsy showed the presence of implantation metastases in the smaller bronchi and actual growth in the alveoli

The histological characteristics are a branching connective tissue stroma with abundant proliferation of squamous epithelial cells forming downward-extending pegs which do not break through the basement membrane. The cells are mature, well-differentiated epithelial cells. Mitosis, concentric cell groups and some irregularity in cellular arrangement may occasionally be seen. Keratinization of the outer layer is usually present in which varying stages of nuclear degeneration are seen. The evidence of the presence of inclusion bodies in the epithelial cells as reported in the literature has not been substantiated by Ferguson and Scott[13] in their series of cases.

The symptomatology of papillomata of the larynx is similar to that usually present in all neoplastic conditions of the larynx. Hoarseness, huskiness, harshness or some alteration of voice, even to complete aphonia, may be present. When the growth becomes obstructive dyspnœa and stridulous breathing are added to the above symptoms. Progressive hoarseness of the voice or cry is the most common early presenting symptom. Cough is not usually a feature. Asphyxia results if these lesions completely obstruct the glottic airway.

The diagnosis can only be made by a careful history and direct laryngoscopy. Age is no contraindication and no anæsthetic should be used in children. Biopsy of course should be performed in all cases. As Jackson[1] states, "The day of inferential diagnosis of laryngeal disease is past." Lateral roentgenograms of the neck, particularly laminographs, are helpful. An infant or child with any of the aforementioned symptoms presents a diagnostic problem and such laryngeal lesions as some other benign tumour, syphilis, tuberculosis, foreign body or congenital anomaly must be ruled out. This can only be done by early direct laryngoscopy.

This type of growth in children is very diffi-

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cult to cure but emphasis must be placed on conservative treatment because the lesion is histopathologically benign and is self-limiting in the majority of cases. An external operation such as thyrotomy or laryngo-fissure is never justified in the treatment of papillomata of the larynx in children because of the resultant laryngeal stenosis. Imfeld[12] reporting on the postoperative control in children states that these growths are both clinically and phonetically more unfavourable in their behaviour than any other benign growth and those occurring in the first three years of life have a graver prognosis because of their frequent recurrences and anatomical extensions.

It is well established that there is a great tendency of these growths to recur following surgical removal, but they usually disappear at puberty or before, regardless of treatment. Broyles[9] has called this active period the "juvenile phase", during which the growth repullulates until the process is burned out. In this latter stage complete removal usually effects a cure.

The average duration of the disease as reported by Ferguson and Scott[13] was three years. The age incidence has been mentioned previously, but McDougal and Wright[4] recorded a case in which the diagnosis of multiple papillomata of the larynx was not established until the patient was 19 years of age and apparently they had been present for six years. In reviewing the literature, the most remarkable case was that presented by Lejeune[10] in which the papillomata persisted for 21 years and terminated fatally with the development of an epidermoid carcinoma of the larynx. This patient had had all methods of treatment available, including 97 surgical procedures.

The treatment of papillomata of the larynx in children has proved universally unsatisfactory as evidenced by the many therapeutic measures. No single method has proved to be the panacea for this condition. Chemical caustics such as silver nitrate, nitric acid and many more have been used in the past but they have been of no benefit, nor is subcutaneous testosterone satisfactory.

New[16] cites the expectant treatment of McKenzie who in 1901 advocated tracheotomy on all cases instead of treating the papillomata and thus provide an airway until the growths had burned themselves out. Most authors agree that tracheotomy is not necessary unless there are signs of obstruction but it would appear that it is necessary in a high percentage of cases. Ferguson and Scott[13] tracheotomized nine of their fifteen cases and Crowe and Breitstein[2] ten of their eleven reported cases. Unless these cases can be kept under close surveillance drastic sequelæ may follow.

Fulguration or electro-coagulation of the growth has been practised by some clinics, but most authors feel that this is a radical form of treatment because of the resultant damage to the vocal cords and laryngeal cartilage. Jackson[1] states that it is fatal to cauterize the base of these

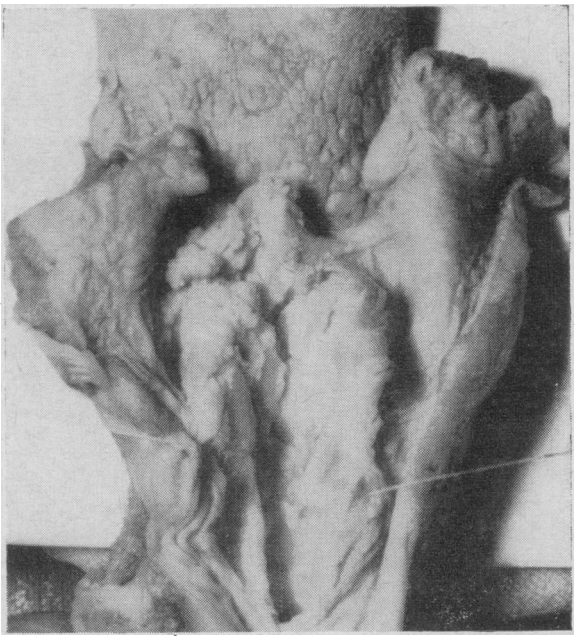

Fig. 1

growths when the origin is from the surface epithelium.

The use of x-ray or radium in the treatment of these lesions has been successful in the hands of several writers. New,[16] reporting in 1921, using extra laryngeal and endolaryngeal radium had 11 out of 17 cases entirely free from papillomata following this treatment. He attributed his success to the use of the radium under direct vision and keeping it in motion while in the glottis. Foster also concludes from his series of cases that most, if not all, primary cases of papillomata of childhood are amenable to proper radiotherapy. Hollinger,[15] as well as many other authors, feels that such treatment, while removing the papilloma, will prevent further growth of the larynx and its probable stenosis or destruction will develop.

Broyles[9] in 1941 reported five cases of papilloma of the larynx in children treated locally by an œstrogenic hormone. He used amniotin. An estimated 1,000 international units was applied at each treatment. He reports that the characteristics of the growth were changed from the infantile to the adult type. To effect a cure required a six month period of treatment. He maintained also that this made surgical treatment more satisfactory. Broyles'[9] methods have not proved so successful in the hands of other writers. Zaline[14] of Liverpool reported considerable success in three cases with the use of dimenformon.

It would appear from the literature that the method of choice in the treatment of laryngeal papillomata in children is repeated endolaryngeal excision using biting forceps flush with the adjacent mucosal surface. Excisions are carried out as often as their recurrence demands. Whether one uses suspension laryngoscopy or not is a matter of individual choice. Ferguson and Scott[13] perform most of their excisions under general anæsthesia.

I present the following case because it is the only one I have seen at this age, and there are so many conflicting opinions in the literature, as well as in this case, as to the most effective method of treatment.

CASE REPORT

S.F., a white female child, age 2 years, was referred by a pædiatrician because of aphonia, dyspnœa and cyanosis and some vomiting. These symptoms had been present since the child was 6 months of age and were becoming progressively worse. On admission to hospital, physical examination showed her to be a pale, thin, cyanotic child who was obviously quite ill. Her temperature was 100° per rectum, pulse rate was 140 and respirations were 42. There was aphonia with some inspiratory stridor and chest retraction. There were diminished breath sounds throughout the chest.

Direct laryngoscopy showed multiple cauliflower growths throughout the whole larynx. The glottic chink was patent but no vocal cord detail could be made out. No attempt was made at removal at this time due to the patient's extreme dyspnœa and cyanosis. The following day, under intratracheal anæsthesia, a tracheotomy was done and numerous papillomata removed with cup forceps. There was considerable hæmorrhage during the procedure. The initial pathological report was that of papillomatous epidermoid carcinoma. In view of this, a barium examination of the œsophagus was done which showed displacement of the trachea and œsophagus to the left suggesting extrinsic involvement, and it was decided to commence radiotherapy at once. I could find no record in the literature of a carcinoma of the larynx in so young a child, so corresponded with Dr. Hollinger[15] of Chicago and Dr. New[16] of the Mayo Clinic, sending further specimens to the former for his opinion. Benign papilloma was the final diagnosis so radiotherapy was discontinued after eleven treatments. There had been no change in the character or extent of the growth.

Two months after admission the child developed measles and bilateral bronchopneumonia and was isolated, during which time she received no treatment. She made an uneventful recovery under the usual chemotherapeutic and antibiotic therapy.

Following the above episode, four further surgical removals of the tumour were performed at weekly intervals but each time the growth appeared the same, probably a little more profuse. Direct application of amniotin in oil as recommended by Broyles[9] was done as well as spraying the larynx with the above preparation between treatments. I felt I was fighting a losing battle but resolved to continue surgical excision as often as possible. It was quite obvious that these papillomata were in the juvenile phase as described by Broyles.[9] There was subglottic extension of the lesion three months following admission requiring bronchoscopic removal of these tumours on two occasions.

All the treatment she had received proved to no avail. Her tumours continued to increase in extent, her airway through the tracheotomy tube was proving very adequate and there was no sign of any extension below the tube level or around the tracheotomy wound. However, six months after admission, one morning she was found dead in bed. There was no sign of there having been any struggle to suggest asphyxia.

Postmortem examination ordered by the coroner showed involvement of the epiglottis and extension for 1.5 cm. down the œsophagus and for 2 cm. below the vocal cords into the trachea. The lungs were filled with a thick viscid mucus but showed no sign of any metastatic tumour involvement. Death was, in all likelihood due to asphyxia and although not stated by the pathologist, was probably due to collapse of the larynx and trachea or sudden blocking of the tracheotomy tube from the papillomatous growth.

SUMMARY

A review of the literature and a case of papillomatosis of the larynx with fatal termination are presented.

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