



Juvenile Laryngeal Papillomatosis in Benin: Epidemiological, Diagnostic, Therapeutic and Evolutionary Aspects

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Abstract

Introduction: To describe the epidemiological profile of patients with laryngeal papillomatosis, to identify diagnostic aspects and evolution after treatment.

Method: This was a retrospective study over 16 years, from May 1995 to April 2011, in the ear nose throat in Hubert Koutoukou Mags, National Teaching Hospital of Cotonou. It was based on descriptive analysis of medical record together with operation report of patients.

Results: We have sampled 18 patients, 1.12 cases/year. The sample included 10 males and 8 females with a frequency of 0.55 in favor of males. The average age was 6.95 year old and children in school or about to be in school were 14. The time-frame between the beginning of symptoms and consultation was 1.17 years. During the endoscopy, the lesions in grains or clusters sometimes obstructive acting as seat on the larynx were confirmed at pathologic check-up.

The treatment consisted of peeling all of them with an average of 4.5 (1-23) associated with the local whitewashing by nifedipine in 5 cases. A tracheotomy rescue had to be performed urgently (12 cases). The monitoring was effective in 14 cases over an average of 14.85 months (3 months-175 months). Ten (10) patients had both normal voice and breathing. In 3 cases the patients had suprasternal draw supine and intermittent dysphonia after decannulation. In a case the weaning could not be achieved. The complications were related to the peeling, to tracheotomy, and to the presence of the cannula.

Conclusion: Laryngeal papillomatosis is a common pathology in children. It is easy to diagnose this pathology but this raises recurrent therapeutic problems such as education disruption and budget burden. The functional and vital prognosis can be involved, hence the need for early consultation.

Keywords: Juvenile laryngeal papillomatosis; epidemiology; diagnosis; Treatment; Complications; diagnostic aspects; and to determine the evolutionary aspects after treatment.

Introduction

Laryngeal papillomatosis is a benign squamous papillary tumor proliferation developed at the expense of the laryngeal mucosa in 90% of cases [1]. Broadly observed in children, though relatively rare it is the most common benign tumors of the larynx. Prevalence of the juvenile laryngeal papillomatosis is 0.7 to 3 [2,3]. The pathogenesis is poorly known evoking intricate theories that are caused by hormones, viral and traumatic. The latter is the only one to date that allows the isolation of the human papillomavirus (HPV) type HPV6 and HPV11. Currently, there is no cure which makes treatment difficult. The clinical course is unpredictable: evolutionary pursuit with a risk of asphyxiation and death, often enucleated healing recurrence with or without an extension to various parts of the respiratory tree. The degeneration is possible but rare and occurs most often in the adulthood [4]. The aim of this study was to describe the epidemiological profile of laryngeal papillomatosis, to identify

Method

This is a retrospective study we conducted based on patients' medical record in ear nose throat department at Hubert Koutoukou Mags National Teaching Hospital of Cotonou from May 1995 to April 2011. The study covered patients with ages ranging between 0 and 17 year old and for whom the diagnosis of laryngeal papillomatosis was done based on anamnestic, clinical and paraclinical arguments. The examination indicated firstly a chronic dysphonia evolving for more than three weeks before then secondarily a dyspnea, and at indirect or direct laryngoscopy clusters blackberries aspect in supraglottic, glottic or glotto-subglottic stage. We have reviewed some patients at dyspnea stages as classified by Pineau and Chevalier Jackson (Table 1).

A rescue tracheotomy with an operation of a cannula was essential when the subject had a stage 3 dyspnea of this classification. All patients underwent surgical treatment of papillomas with forceps

peeling on bi-weekly average. The pathological examination after biopsy confirmed the diagnosis of laryngeal papillomatosis. This was followed by a painting with mitomycin six where the lesion was exuberant or recurrent. When the larynx was obstructive then the decannulation was made after weaning by gradual light reduction of the material in use. The parameters studied were frequency, sex, age, consultation period, reasons for consultation with or without associated signs, the location of lesions, treatment and evolution.

	Stage 1	Stage 2	Stage 3	Stage 4
Draw	Discrete	Vegetate	Major Diffuse	Ice
State of integument	Normal	Vulvular	++ Cyanosis	Cyanosis
Pulse rate	-/+ Normal	+/+ Increased	Increased	Collapsed
Atelectasis	+/- Normal	+/- Increased	Increased	Collapsed
State respiratory way	o/ Normal	Aghast	Anguish	++ +/Dyspnea

Table 1: Classification of Pineau and Chevalier Jackson.

Results

Over the 16 years study period, 18 patients were consulted with a rate of 1.12 cases per year. The sample included 10 males/18 with a frequency of 0.55 and an average age of 6.95 years (21 months-16 years), 14 children were in school or at age to be.

		Number (frequency)
Aspects lesions	Grains of papillomas	02 (11%)
	papilloma clusters	03 (17%)
	Fonds**	13 (72%)
Localisation	Supraglottis+epiglottis**	05 (28%)
	Glottis	04 (22%)
	Glottis+subglottis	06 (33%)
	Supra-glottis+glottis+subglottis	03 (17%)
Total		18 (100%)

*Over 13 presenting a fixed aspect 10 lesions were obstructive.

**the localisation related to the first 2 stages of the larynx, one case spread over to the oropharynx

Table 2: Distribution of patients by endoscopic examination.

Clinically, the consultation period was 1.17 years (1 week-5 years), 1/3 consulted for dysphonia crawling, another 1/3 dysphonia association and dyspnea without signs of decompensation and the remaining one 1/3 for dyspnea stage 3. Table 2 summarizes the distribution of patients according to the results of the endoscopic review. Clamp peeling with an average of 4.6 (1-23) was treatment opted for in our series. Mitomycin was applied 1 to 4 times by brushing after surgery in 3 patients. In an emergency, a tracheotomy rescue was necessary in 12 cases and in total during the study period.

this act was performed 15 times with an average of 1.25 (1-5). Pathological examination of samples has proved laryngeal papillomatosis. 04 patients underwent additional consultations in emergency way. In a case there were 2 readmissions and 1 readmission in 3 cases.

The evolution was assessed in 14 patients over an average of 14.85 months (3 months-175 months) on the quality of the voice, breathing and the occurrence of complications (Table 3). Ten (10) patients had both normal breathing and voice. We noted the impossibility of decannulation in 1 case and in 3 cases the patients had supraglottal draw supine and intermittent dyspnoea after decannulation.

		Number (frequency)
Peeling	functions complications	08/18 (44%)
	changing mucosa dysplasia	01/18 (6%)
Tracheotomy	Emergency total-cervical-thoracic	02/12 (17%)
	Voiced cervical scar	01/12 (8%)
cannula	larynx Prosthesis	01/12 (8%)
	pericannular Purulent secretions	01/12 (8%)
	Tracheal-bronch fistula	01/12 (8%)
Granuloma		03/12 (25%)

Table 3: Distribution of patients according to the complications. All 18 patients were peeling -12 underwent emergency tracheotomy and wore a cannula.

Discussion

We consulted 18 patients over 16 years with a rate of 1.12 cases per year. The sample included 10 males with a frequency of 0.55 in favour of males and an average age of 6.95 years. The consultation period was 1.17 years (1 week-5 years), 1/3 consulted for dysphonia crawling, another 1/3 dysphonia association and dyspnea without signs of decompensation and the remaining one third for dyspnea stage 3. Clamp peeling with a mean of 4.6 (1-23) was the treatment opted for in our series. Mitomycin was applied 1 to 4 times by brushing after surgery in 3 patients. In an emergency, a tracheotomy rescue was necessary in 12 cases and in total during the study period, this act was performed 15 times with an average of 1.25 (1-5). The evolution was assessed in 14 patients over an average of 14.85 months (3 months-175 months) on the quality of the voice, breathing and the occurrence of complications.

Epidemiologically, laryngeal papillomatosis is a benign and occasional disease with variable incidence is 0.2 to 0.7 cases per 100,000 population [5,6]. The number of new cases per year of the juvenile laryngeal papillomatosis is 0.7 to 4 [9] while it was 1.12 in our series. If the male predominance is for Ndizey [3], laryngeal papillomatosis is more common in children and regardless of gender with an average age ranging from 6 to 10 years [7,8].

The diagnosis is often delayed because of the long use of consultation up to 5 years. Indeed, dysphonia first sign of laryngeal papillomatosis may be laryngitis or an asthma status difficult diagnosis [9,10]. This is not the case in the series of Trunac [11], where 90% of cases were consulted at the stage of dysphonia. In our series, patients were

consulted in 7/3 of cases at the stage of dyspnea. This is a laryngeal cypnea faster or slower type inspiratory bradypnea with draw and wheezing.

The review to nasofibroscopy or direct laryngoscopy under A/G addresses clinical assessment by objectifying in a mobile larynx lesions pink or greyish as grains clusters, sometimes florid, obstructive or not. The lesions extended to the 3 floors of the larynx and aryepiglottic in one case. As for the tracheobronchial tree, it has not been explored due to limited technical facilities in our work environment.

The pathological examination was performed in all cases to confirm the benign lesions.

Theoretically, the delay in consultation, sometimes multistage extension of the lesions and the obstructive nature of lesions required a tracheotomy first in 12/13 cases. This gesture would be saving an aggravating factor as it would favor the extension of lesions throughout the respiratory tract [1,12,13]. Peeling was the only surgical treatment for all patients because the gesture is simple, easily accessible and inexpensive.

In evolutionarily, 14/15 patients were reviewed. Recurrences were frequent with the need to resume peelings every 2 weeks. Apart from these controls set, 4 patients were received emergency in an acute of cypnea stage 3. During the monitoring period, the average peelings was 4.6 (1-23). The complications were related to emergency tracheotomy and residence of the cannula. The facial-cervical-thoracic emphysema can be explained by tight stitches on both sides of the incision instead of tracheotomy or too small tracheostomy tube allowing fuser upstream and downstream of the site. As for tracheal stenosis, it produces in contact with the tracheostomy tube of secretions which at long term pass over inferior and goes down to the lower airways. Similarly mini friction of the cannula can create irritation, then a naso-tracheal fistula and a granuloma. The sequelae of peeling are firstly a bad voice quality probably due to a lack of contraction of the vocal cords after fibrosis or poor healing of the laryngeal mucosa and also observed the occurrence of dysplasia one patient by the mucosa redesign.

Conclusion

Laryngeal papillomatosis is a common pathology of the child. It is fairly easy to diagnose but poses therapeutic difficulties. The

development is toward recurrence and eduction disruption of the child. The functional and vital prognosis can be involved, hence an early consultation is needed.

References

1. Conness G, Herzig B, Roguel E, Gombin J, Pautier J. (2005) Chirurgie des tumeurs bénignes de larynx. Elsevier Paris.
2. Szeceki B, Williams W, Deslovers C, Van Boven M, Nansen L, et al. (1998) Treatment of severe laryngeal papillomatosis with intracavitary injections of interferon α 2a. *J Clin Virol* 34: 319-325.
3. Christensen TH, Jørgensen K, Christensen A. (1984) Juvenile papillomatosis of the larynx: 45 years follow-up from county of Denmark. *Acta Otolaryngol* 112: 37-39.
4. Brandt A (1958) Juvenile laryngeal papillomatosis. An epidemiological study from the Copenhagen region. *Acta Otolaryngol* 116: 367-371.
5. Ondrouts G, Galiba J, Kausse B, Flume F (2002) Laryngeal papillomatosis: value of early diagnosis, apropos of 7 cases diagnosed at the University Hospital Center in Kinshasa (Congo). *Med Trop (Mars)* 62: 163-165.
6. Ndiaye M, Ndiaye IG, Hane ODA, Tall A, Diatta BK, et al. (2009) Papillomatose laryngée de l'enfant. *FR ORL* 94: 299-302.
7. Tahirou SK, Kouyba-Togole F, Mohamed AA, Keita HA, Soko HB, et al. (2002) Juvenile papillomatosis in Mali: A propos of 19 cases collected at the Gabriel Touré Hospital of Bamako. *Bull Soc Fr Jol For* 55: 21-24.
8. Maliki D, Nouri H, Zied J, Kechi Y, Aïmeche I, et al. (2012) La papillomatose laryngée de l'enfant : aspects épidémiologiques, thérapeutiques et évolutifs. *Journal de pédiatrie et de pédiatrie* 28: 237-241.
9. Bloof B, Ouhis K, Ndiaye I, Diop FM, Diop IS (1988) Laryngeal papillomatosis: report of 27 cases. *J Ghar Med* 34: 102-106.
10. Mohamed A, Timbo SK, Konte-Cugala P (1996) Papillomatose du larynx: étiologie et propos de 6 cas récents. *Med Afr Nouv* 43: 650-653.
11. Tsouvas I, Devars P, Petit J, Partmann D, Papaxanthos M, et al. (1987) [Result of the treatment of juvenile papillomatosis of the larynx. Apropos of 138 cases]. *Rev Laryngol Otol Rhinol (Bour)* 108: 331-334.
12. Bauman NM, Smith BJ (1964) Recurrent respiratory papillomatosis. *Pediatr Clin North Am* 11: 1385-1401.
13. Colclough L, Pages C, Assier S, Guimond P, Nassy P (1988) [Development of laryngeal papillomatosis in children. Apropos of 17 cases]. *Arch Fr Pédiat* 45: 387-392.