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**MORTALITY OF OESOPHAGEAL ATRESIA IN A LOW-INCOME SETTING:
ABOUT 21 CASES.
MORTALITE DE L'ATRESIE DE L'ŒSOPHAGE EN MILIEU DEFAVORISE : A
PROPOS DE 21 CAS.**

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Abstract

Introduction: Oesophageal atresia (OA) is an extreme neonatal surgical emergency which when present, heavily affects the neonate, in Cameroon and in Sub-Saharan Africa generally.

The aim of our study was to describe the epidemiological, diagnostic and therapeutic characteristics of deceased patients with oesophageal atresia in our context.

Methodology: We conducted an observational, descriptive, retrospective study over a period of 14 years, from the 1st January 2005 to the 28th February 2019. All paediatric patients who died from oesophageal atresia were included.

Survivors were excluded (n = 3). **Results:** Mortality from oesophageal atresia was 87.5% in our series. The average age at the time of presentation was 5.2 days. The notion of attempted feeding at birth was found in 72.2% (n = 13). No antenatal diagnosis was noted. The lung condition was preoccupying in 61% of cases (n = 11). The average operating time was 7 days. Death before any surgical procedure concerned 22.2% of newborns (n = 4). Posterolateral thoracotomy with fistulectomy and oeso-oesophageal anastomosis represented 22.2% of our series (n = 4) while a feeding gastro-

jejunostomy was done in 66.6% of cases. Two deceased patients had undergone a feeding gastro-jejunostomy followed by a posterolateral thoracotomy. **Conclusion:** Knowledge of these epidemiological, diagnostic and therapeutic characteristics is a prerequisite for considering an inversion of the mortality curve for oesophageal atresia in our context.

Key words: Mortality, oesophageal atresia, Yaoundé

Résumé

Introduction : L'atrésie de l'œsophage (AO) est une extrême urgence chirurgicale néonatale pour laquelle le nouveau-né paye un lourd tribut au Cameroun et en Afrique Sub-saharienne en général. Le but de notre travail était de décrire les caractéristiques épidémiologiques, diagnostiques et thérapeutiques des patients décédés porteurs d'une atrésie de l'œsophage dans notre contexte. **Méthodologie :** Nous avons mené une étude observationnelle, descriptive, rétrospective sur une période allant du 1^{er} janvier 2005 au 28 février 2019, soit 14 ans. Ont été inclus tous les patients pédiatriques décédés des suites d'une atrésie de l'œsophage. Ont été exclus les patients survivants (n=3) opérés d'une atrésie de l'œsophage. **Résultats :** La mortalité de l'atrésie de l'œsophage a été de 87,5% dans notre série. L'âge moyen au moment de la présentation était de 5,2 jours. La notion de tentative d'alimentation à la naissance était retrouvée dans 72,2% (n=13). Aucun cas de diagnostic anténatal

n'a été noté. L'état pulmonaire était préoccupant dans 61% des cas (n=11). Le délai opératoire moyen était de 7 jours. Le décès avant tout geste chirurgical concernait 22,2% des nouveau-nés (n=4). La thoracotomie postéro-latérale avec fistulectomie et anastomose oeso-œsophagienne représentait 22,2% de notre série (n=4), alors que la gastro-jéjunostomie d'alimentation représentait 66,6%. Deux patients décédés avaient été l'objet d'une gastro-jéjunostomie d'alimentation suivie d'une thoracotomie postéro-latérale. **Conclusion :** La connaissance de ces caractéristiques épidémiologiques, diagnostiques et thérapeutiques est un préalable pour envisager une inversion de la courbe de mortalité de l'atrésie de l'œsophage dans notre contexte.

Mots clés : Mortalité, Atrésie de l'œsophage, Yaoundé.

INTRODUCTION

Oesophageal atresia is defined as a congenital complete interruption, in its thoracic course of the segment of the digestive tract located between the pharynx and the stomach. It is an extreme neonatal surgical emergency which when present, heavily affects neonates in Cameroon and Sub-Saharan Africa generally, with mortality rates between 70% and 100% depending on the series [1-4]. These figures are inconsistent with those of the Western series, notably that of Gottrand and al. which reports less than 5% mortality in its series in France, and of Lilja et al. in Sweden which notes 0% mortality in isolated forms [5,6]. With the aim of reversing this trend, we initially wished to describe here the epidemiological, diagnostic and therapeutic profile of deceased patients with oesophageal atresia in our context.

PATIENTS AND METHOD

It was an observational, descriptive and retrospective study over a period from the 1st January 2005 to the 28th February 2019, i.e. 14 years and 2 months, in the Paediatric Surgery unit of the Yaoundé Gynaeco-Obstetric and Paediatric Hospital (YGOPH). Were included all the records of paediatric patients with oesophageal atresia whose outcome was death. Were excluded records of newborn babies who survived after the treatment of oesophageal atresia (n=3). The following parameters were of interest to us:

- At the epidemiological level: age, sex, birth weight, term, type of transport, notion of attempted feeding.
- At the diagnostic level: antenatal diagnosis, postnatal diagnostic delay, type according to Ladd, associated malformations, "gap", prognostic

classification according to Waterston and Spitz.

- At the therapeutic level: the operation delay, surgical strategy, surgical procedures performed and postoperative death delay.

RESULTS

Twenty-one files of newborns with oesophageal atresia were collected during our study. Mortality concerned 18 cases of them. The deceased newborns were 5.2 days old (1 to 14 days) at the time of presentation. Predominantly female, born at term (13 cases) with an average birth weight of 2830 gr (1950 and 3780 gr). Thirteen of them were transferred to YGOPH by non-medical means and fed at birth in 14 cases. Antenatal diagnosis was inexistent in our series. The mean postnatal diagnostic delay was 6 days (0 to 16 days). Acute bronchopneumonia was present in 13 cases. Oesophageal atresia type III according to Ladd and Gross classification was noted in all cases. Short “gap” was noted in 13 cases, rarely associated with a cardiovascular malformation in 3 cases (Table I).

Table I: Distribution of paediatric patients according to associated malformations.

Associated Malformations	(n)	(%)
None	13	72,2
Cardiovascular system	3	16,6
Digestive system	1	5,5
Locomotor system	1	5,5

Eighteen are of group I according to Spitz and nine of group C according to Waterston (Table II and III).

Table II: Distribution of paediatric patients according to Waterston classification.

Waterston classification	(n)	(%)
Group A	2	11,2
Group B	07	38,8
Group C	9	50

Table III: Distribution of paediatric patients according to Spitz classification.

Spitz classification	(n)	(%)
Group I	16	88,8
Group II	2	11,2

The average operation delay was 7 days (2 to 19 days). Thoracoscopy was not used. A feeding gastrostomy prior to curative surgery was the adopted strategy in 12 cases in our series. Two deceased patients had undergone a feeding gastro-jejunoscopy followed by a posterolateral thoracotomy (Table IV).

Table VI: Distribution of deceased neonates depending on the procedure performed.

Surgical procedure	(n)	(%)
Feeding gastrostomy	12	66,6
Posterolateral thoracotomy	4	22,2
Thoracotomy + feeding gastrostomy	2	11,2

Four patients were the subject of posterolateral thoracotomy with fistulectomy and oeso-oesophageal anastomosis. Death before any surgery concerned 4 babies in our series. The

average delay in postoperative death was 8.3 days (0 to 26 days).

DISCUSSION

The average age at admission was 5.2 days, ranging from 1 to 14 days of life. These results are similar to those presented by Ndoye and al. in a study carried out in Dakar in 2017 [7]. The authors found an average age of 5.1 days with extremes from 0 to 21 days. Our population was predominantly female, which is consistent with the results of the study carried out by Mouafo et al. in 2010 in two hospitals in the city of Yaoundé [8]. In contrary to those of Sinha and al. in India who found nearly 92% of boys in their series [9]. This is explained by the female predominance of the population. The average birth weight was 2830g with extremes ranging from 1950g and 3780g and this was in accordance with that found in the sub-Saharan series [7,8,10,11]. The study population consisted of 13 term babies, just like in the study by Vukadin and al. on the analysis of mortality in patients with OA in Belgrade [12]. It should be noted that Spitz found no statistically significant association between prematurity and oesophageal atresia [13]. In our series, thirteen newborns were transferred to YGOPH by non-medical means, which is superior to the percentage in a study by Ndour and al. in 2006 in which 60% of patients did not benefit from medical transport [10]. This could be explained by the low socioeconomic level of the patients and the absence of an organized medical transport system in Cameroon. Finally, 14 newborns were fed at birth, comparably to Tandon and al who found a notion of feeding in 70% of cases, and this was associated with a significant increase in mortality due to the worsening of bronchopneumonia [14]. These similarities can be explained on one hand by the lack of sensitization on systematic oesophageal patency assessment using a nasogastric tube in the delivery room and on the other hand the encouragement of immediate breastfeeding after birth. In our

series, we did not find any case of diagnosis made during pregnancy, which is consistent with the results of Ndoye and al. in 2016 in Dakar and Mbaye and al. in 2015, but contrasting with the results of Gottrand and al. who in a study done in France in 2012 on the antenatal diagnosis of oesophageal atresia, showed that in 66.6% of cases, the diagnosis of OA was suspected before birth [4,5,7]. Antenatal diagnosis allows referral of patients to a hospital capable of providing adequate care for the disease within twenty-four hours of birth. The recurrent and predominant absence of prenatal follow-up in our current practice, due either to the lack of education or to the lack of financial means associated with a significant lack of qualified personnel able to detect the signs of the affection during an ultrasound explain these significant differences. The average diagnostic delay of 6 days (0 and 16 days) is identical to that of Wandaogo in 2009 at the Ouagadougou University Hospital in Burkina-Faso [11]. This is much higher than that of developed countries where treatment rarely exceeds 48 hours [6,11,15]. The shortness of this delay can be explained by a careful clinical examination of the newborn in the delivery room and systematic verification of oesophageal permeability in the West. In addition, several factors explain the delay in consultation, particularly the low educational level of parents [4]. Indeed, UNICEF reporting the situation of children in the world in 2004, showed that the understanding of health problems, in particular the early use of care, increases alongside with the mother's level of education; populations tend to consult traditional healers first and only go to health centers after a failure [16,17]. Finally, children spend a long time in health centers and secondary hospitals. The delay in diagnosis explains the picture of bronchopneumonia on admission in most children due to unsuccessful feeding attempts. In fact, 11 of newborns have bronchopneumonia. These figures are higher than those of the study by Mbaye and

al, in which 48% of patients were suffering from severe bronchopneumopathy, of whom half died before any surgical treatment [4]. At the conclusion of a study on the prognostic criteria of oesophageal atresia, Waterston and al. established a statistically significant relationship between pulmonary involvement and the prognosis, the latter being poor in severe lung lesions [18]. Similarly, in 2008, Tandon and al. found a survival rate of 82% for patients without preoperative pulmonary disease, 72% in the case of moderate lesions and only 20% if the lesions were severe [14]. Ladd and Waterston's type III oesophageal atresia was the only one found in our series, in accordance with the data in the literature [13]. We found 5 cases associated with malformations, most frequently cardiac (3 cases), which coincides with Spitz' findings [13]. The distribution of the population according to Waterston was 9 cases in group C, 7 cases in group B and 2 cases in group A with consequently 6%, 68% and 95% chance of survival, respectively. This distribution is probably explained by the repeated and relentless feeding attempts before consultation. The distance between the two ends of the esophagus, also called the gap, was inferior or equal to 3 vertebrae in 11 cases, results lower than those of Powell and al. who found a long gap in 10% of AO [19]. Similarly, the long gap was associated with higher mortality in the study by Li and al., due to the significant tension and the higher risk of anastomotic leak than in the presence of a short gap [20].

Four patients died before any surgical procedure. This result attests on one hand of the severity of the clinical pictures on admission and on the other hand of the insufficiency of the means of care, notably the absence of a paediatric intensive care unit, a finding also noted by Ndour and al. [10]. The mean operative delay was 7 days with extremes of 2 and 19 days of life. This delay is close to the one established by

Ndoye and al in 2017 in Dakar, i.e. averagely 8.4 days with extremes of 0 and 21 days [7]. These figures are quite opposite to those in the Western series in which care is achieved within 24 to 48 hours of birth [5, 13, 15]. The delay in treatment in our context is explained on one hand by the delay in diagnosis, but on the other hand by the difficulties that patients' families have to face the often-expensive costs of surgery and preoperative resuscitation. The most widely used surgical strategy was the 2-step strategy (12 cases) which implies performing a feeding gastrostomy before curative surgery. This strategy differs from those found in the literature in which almost 80% of patients undergo a thoracotomy straightaway [12,14]. This is explained by the fact that the indication for gastrostomy is conventionally reserved for types I and II of OA, rarely found in practice [13]. However, in our setting, gastrostomy is performed because it offers an alternative, considering the deplorable pulmonary state and the low weight of our patients. However, only 2 newborns benefited from gastrostomy followed by thoracotomy. The postoperative mortality delay was 8.3 days (0 and 26 days) after the operation, figures identical to those of Ndour and al. who found a delay of 9.1 days after the operation [10]. The mortality rate was 87.5%, a rate significantly higher than that found in the African series in which it varies between 60 and 75% [4,7,11]. This result contrasts with European series in which the mortality is almost zero [5,6,15]. It is however lower than that found by Leis and al. in Kabul, which is close to 100% [21].

CONCLUSION

Knowledge of the epidemiological, diagnostic and therapeutic profile of deceased patients with oesophageal atresia is a prerequisite for considering a short term inversion of the mortality curve of this condition in our context.

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