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Review Article

Trends in the publication of infantile hypertrophic pyloric stenosis in Africa: A systematic review

Olakayode Olaolu Ogundoyin[1], Akinlabi Emmanuel Ajao[1]

[1]Department of Surgery, College of Medicine, University of Ibadan and University College Hospital, Ibadan, Oyo, Nigeria.

***Corresponding author:**
Olakayode Olaolu Ogundoyin, Department of Surgery, College of Medicine, University of Ibadan and University College Hospital, Ibadan, Oyo, Nigeria.

kayogundoyin@gmail.com

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ABSTRACT

There are still global variations in the epidemiology of infantile hypertrophic pyloric stenosis, although the clinical presentations may be similar. Outcome of management, however, may depend on the degree of evolution of management of the anomaly. This review aimed at evaluating the trends of reporting of infantile hypertrophic pyloric stenosis from Africa. An evaluation of all publications from Africa on infantile hypertrophic pyloric stenosis focusing on epidemiology, evolution of management of the anomaly was carried out. Literature search of all publications from Africa on Infantile hypertrophic pyloric stenosis was conducted from January 1, 1951, to December 31, 2018. The articles were sourced from the databases of African Index Medicus, OvidSP, PubMed, African Journal Online, and Google Scholar. Extracted from these publications were information on the type of article, trend of reporting, the country of publication, demographic details of the patients, number of cases, clinical presentation, pre-operative management, type of surgical approach, and the outcome of management. Overall, 40 articles were published from 11 countries. Of these, 16 (40.0%) were published in the first 35 years (Group A, 1951–1985) and 24 (60.0%) published in the later 33 years (Group B, 1986–2018). Case reports 8 (20.0%) and case series 5 (12.5%) were predominant in Group A, whereas retrospective studies 12 (30.0%) predominated in Group B. The countries of publication included Nigeria (27.5%), South Africa (15.0%), Egypt (12.5%), Tanzania (10.0%), and Zimbabwe (10.0%). A total of 811 patients diagnosed and managed for infantile hypertrophic pyloric stenosis (IHPS) were reported. Their ages ranged from 1 day to 1 year with an incidence that ranged from 1 in 550 to 12.9 in 1000. There were 621 boys and 114 girls (M:F – 5.5:1). All the patients were breastfed with an average birth rank incidence of 42.4% among firstborns, 19.5% in second borns, 15.2% in third borns, 13.2% among fourth borns, and 10.0% among fifth borns and beyond. Associated congenital anomalies were reported in 5 (12.5%) studies with an incidence of 6.9–20% occurring in a total of 28 patients. All but 3 (7.5%) studies reported that open surgery was adopted to perform Ramstedt's pyloromyotomy on the patients. Reported post-operative complications include mucosal perforation in 8 (20.0%) studies, surgical site infection in 7 (17.5%), gastroduodenal tear 2 (5.0%), and hemorrhage and incisional hernia in 1 (2.5%) study each. Mortality was reported in 26 (65.0%) studies with a range of 1.8–50% and a mean mortality rate of 5.2%. There has been a change in the trend of reporting IHPS in Africa over the years, with increasing comparative studies on the modalities of management compared to case reports and series. Still very limited work has been done in the aspect of genetics and etiology of IHPS among Africans. There is a need to increase funding in this regard and to encourage multi-center collaborations in the study of this relatively rare condition.

Keywords: Africa, Hypertrophic, Infantile, Pyloric stenosis

INTRODUCTION

Infantile hypertrophic pyloric stenosis (IHPS) is an anomaly whose etiology is not clear, but there is global variation in its epidemiology. It is characterized by progressive hypertrophy of the

circular muscles of the pylorus with consequent obstruction of the gastric outflow, mostly in neonates and infants under the age of 1 year.[1] The resulting gastric outlet obstruction produces a clinical manifestation of postprandial, projectile, non-bilious vomiting with associated palpable epigastric abdominal mass.[2-4] IHPS is highly prevalent in the Caucasian population with an incidence of 5/1000 newborns[5-7] in comparison to the African population in which it is rarer.[8] Other well-documented risk factors include a male preponderance (especially firstborn male) with a 4–5 times higher risk than females, young maternal age, and positive family history of IHPS.[5,6] Clinical diagnosis is usually made following a palpable mass in the epigastrium during feeding; however, this may sometimes be difficult to appreciate. Ultrasonography of the abdomen and contrast study of the upper gastrointestinal tract have proved very valuable in identifying the olive-shaped mass in the epigastrium.[1] Following adequate pre-operative resuscitation to correct fluid and electrolyte abnormalities, Ramstedt's Pyloromyotomy[9] is often the surgical treatment of choice with a good post-operative outcome.

The relatively rare nature of this anomaly in the African population has made literatures from Africa to be very limited on the epidemiology, etiopathogenesis, management, and clinical outcome of IHPS. This systematic review sought to evaluate the trends of reporting IHPS from all publications emanating from various centers in Africa.

MATERIAL AND METHODS

The guidelines provided by the 2009 statement of the Preferred Reporting Items for Systematic Reviews and Meta-analysis[10] were used to carry out a systematic review of all literatures on IHPS emanating from Africa within the last seven decades from January 1951 to December 2018. Searches were conducted of all articles in the databases of African Index Medicus, OvidSP, PubMed, and African Journal Online added with Google Scholar Search using "Infantile hypertrophic pyloric stenosis" AND "Africa" OR "Congenital hypertrophic pyloric stenosis" AND "Africa." Full evaluation of each article was performed to identify those that reported IHPS in Africa. The relevance of these articles was examined and inclusion criteria were case reports, case series, and original articles. Excluded were review articles, commentaries, letters to the editor, and any other publication that did not provide adequate patients' information. Articles that were published in languages other than English language were translated to English language using Google translator. Recorded from these publications were information on the type of article, trend of reporting, the country of publication, demographic details of the patients, number of cases, clinical presentation, pre-operative management, type of surgical approach, and the outcome of management. The trend of reporting was examined by comparing the publications in the first 35 years of the study (1951–1985) with the ones published in the following 33 years of the study (1986–2018). Analysis of data was performed using Microsoft Excel Spreadsheet 2010. Categorical variables were summarized using frequencies and proportions, while continuous variables were summarized using the mean.

RESULTS

Following the database search for IHPS in Africa, a total of 62 publications were obtained with 59 publications meeting the criteria for the study after screening. A total of 40 articles were evaluated fully for the study after 19 duplicates were excluded [Figure 1]. Of these, 16 (40.0%) articles were published within the first three and a half decades of the study[11-26] [Table 1a] from 1951 to 1982 (Group A) and 24 (60.0%) were published in the last three and a half decades [Table 1b] from 1983 to 2018 (Group B).[9,27-49] There were 14 (35.0%) case reports,[12,14-17,19,21,24,32-34,43-45] of which eight (20.0%) were published within the first three and a half decades[12,14-17,19,21,24] and six (15.0%) articles were published later.[32-34,43-45] Seven (17.5%) articles were case series,[11,18,20,22,23,36,41] all but two (5.0%)[36,41] were published within the first three and a half decades, 15 (37.5%) articles were retrospective studies[9,13,25-31,35,37,39,40,46,48] with three (7.5%) published in the first three and a half decades[13,25,26] of the study, and 11 (27.5%) in the last three and a half decades.[9,27-31,35,37,39,40,46,48] There were four (10.0%) prospective studies,[38,42,47,49] all of which were published in the last three and a half decades of the study. Eleven (27.5%) articles

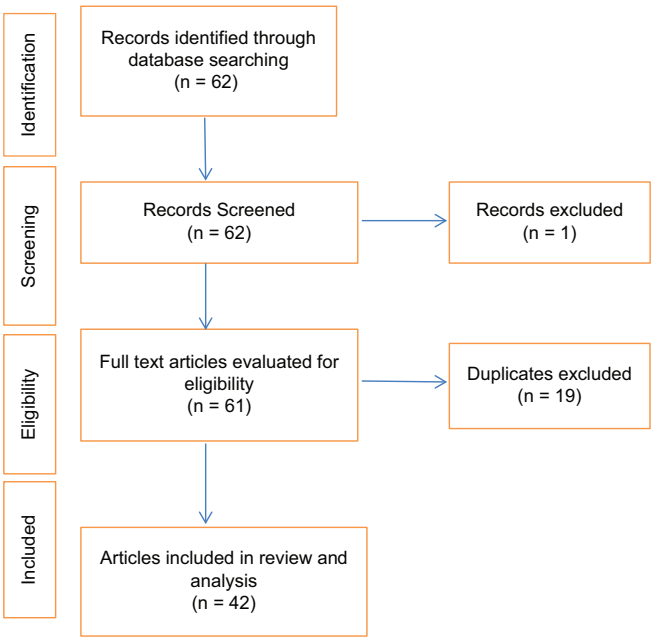

Figure 1: Flowchart for article selection for the study.

Table 1a: Type study and country of publication from 1951 to 1982 (Group A).

Authors	Year	Type	Country	Number of patients
Pitcher	1953	Case Series	Nigeria	2
Marks	1955	Retrospective Study	Zimbabwe	25
Shepherd-Wilson and Gelfand	1955	Case Report	Zimbabwe	1
Griffiths	1956	Case Report	South Africa	1
Luder	1956	Case Report	Uganda	1
Hamilton	1957	Case Report	Uganda	1
Menezes and Thethravusamy	1957	Case Report	Tanzania	1
Scragg	1958	Case Series	South Africa	3
Boroda	1960	Case Report	Uganda	1
Swan	1961	Case Series	Nigeria	10
Grave	1961	Case Report	Zimbabwe	1
Hammar and Forbes	1964	Case Series	Zimbabwe	3
Audu	1964	Case Series	Nigeria	5
Javett et al.	1973	Case Report	South Africa	1
Hassan and Bayomi	1975	Retrospective Study	Sudan	17
Johnson and Adekunle	1976	Retrospective Study	Nigeria	31

emanated from Nigeria,[11,20,23,26,31,33-35,43,45,48] 6 (15.0%) from South Africa,[15,18,24,29,37,39] 5 (12.5%) from Egypt,[32,38,42,47,49] 4 (10.0%) each from Tanzania[9,17,28,44] and Zimbabwe,[12,13,21,22] 3 (7.5%) from Uganda,[14,16,19] 2 (5.0%) each from Cameroon,[41,46] and Ethiopia[27,40] whereas 1 (2.5%) each were published from Republic of Benin,[36] Ghana,[30] and Sudan.[25]

A total of 811 patients diagnosed and managed for IHPS were reported with an age range of 1 day–1 year and an incidence that ranged from 1 in 550 to 12.9 in 1000. Sex incidence was reported by 35 studies (87.5%) and there were 621 boys and 114 girls (M:F – 5.5:1). All the patients were breastfed and the average birth rank incidence was 42.4% among firstborn, 19.5% in second, 15.2% in third, 13.2% among fourth, and 10.0% among fifth born and beyond. Vomiting was the most common clinical presentation reported by 20 (50.0%) articles, this is followed by visible peristalsis 19 (47.5%) and palpable tumor reported by 17 (42.5%) articles. Twelve (30.0%) articles reported electrolyte problems, of these, only one article (2.5%) had normal serum electrolytes. The reported electrolyte derangements included hypokalemia and metabolic acidosis reported by 7 (17.5%) articles each, hypochloremia by 6 (15.0%), and hyponatremia by 5 (12.5%).

Associated congenital anomalies were reported in five (12.5%) studies, with an incidence of 6.9–20% occurring in a total of 28 patients. Inguinal hernias were the most common associated anomalies (11 patients) reported by all the studies. Others included congenital heart defects (5 patients), neural tube defects, undescended testis, Down's syndrome, and craniosynostosis in 2 patients each, recto-vaginal fistula, cleft palate, and Meckel's diverticulum in one patient each. Thirty-five (87.5%) studies reported the operative procedures performed on the patients. Of these, all but three (7.1%) studies reported that open surgery was adopted to perform the pyloromyotomy. The remaining three studies reported laparoscopic pyloromyotomy using endo umbilical approach.[39,47,49] Of the open surgical approach, right upper transverse approach was most commonly used, other approaches included circumumbilical (3, 7.5%), supraumbilical (3, 7.5%), and upper midline (1, 2.5%). Mucosal perforation was reported by 8 (20.0%) studies with a range of 2.5–10.9%. Other post-operative complications reported included surgical site infection in 7 (17.5%) studies, gastroduodenal tear in 2 (5.0%), hemorrhage and incisional hernia in 1 (2.5%) study each.

Mortality was reported in 26 (65.0%) studies with a range of 1.8–50% and a mean mortality rate of 5.2%. The mean duration of hospital stay ranged from 2 to 13 days.

DISCUSSION

Trend of publications

A review of publications revealed a marked increase in the number of publications in the latter years, with the second group accounting for 60% of all publications used in this review. The increase is believed to result from increased awareness about the anomaly resulting in change of attitude of people across the continent about childhood surgical diseases and their treatment,[50] increasing number of specialist surgeons dedicated to the management of the surgical neonates and infants, as well as improvement in the diagnosis and treatment of the anomaly and changing trends in the attitude of reporting cases. Case reports and case series accounted for 52.5% of the publications, with majority these reports published in the first three and a half decades of the review. The relatively rare nature of this anomaly in Africans compared to Caucasians and Asians may account for this. Of the remaining 47.5%, a considerable proportion of the publications, especially the retrospective studies,[9,13,25-31,35,37,39,40,46,48] were dedicated to describing the pattern of presentation and outcome of management of the anomaly, whereas the prospective studies[38,42,47,49] tried to assess the outcome and usefulness of newer methods of managing hypertrophic pyloric stenosis.

Table 1b: Type of study and country of publications from 1983 to 2018 (Group B).

Authors	Year	Type of study	Country	Number of patients
Lemessa	1990	Retrospective Study	Ethiopia	40
Carneiro	1991	Retrospective Study	Tanzania	15
Emmink *et al.*	1992	Retrospective Study	South Africa	62
Tandoh and Hesse	1992	Retrospective Study	Ghana	84
Nmadu	1992	Retrospective Study	Nigeria	20
Gad *et al.*	2003	Case Report	Egypt	1
Okafor *et al.*	2004	Case Report	Nigeria	1
Egri-Okwaji *et al.*	2005	Case Report	Nigeria	1
Osifo and Evbuomwan	2008	Retrospective Study	Nigeria	57
Fiogbe *et al.*	2009	Case Series	Benin Republic	2
Banieghbal	2009	Retrospective Study	South Africa	32
Eltayeb and Othman	2011	Prospective Study	Egypt	40
Saula and Hadley	2011	Retrospective Study	South Africa	63
Tadesse and Gadisa	2014	Retrospective Study	Ethiopia	55
Tambo *et al.*	2014	Case Series	Cameroon	2
Chalya *et al.*	2015	Retrospective Study	Tanzania	102
Nofal *et al.*	2016	Prospective Study	Egypt	20
Ogunlesi *et al.*	2016	Case Report	Nigeria	1
Bizzocchi and Metz	2016	Case Report	Tanzania	1
Seyi-Olajide *et al.*	2017	Case Report	Nigeria	1
Ndongo *et al.*	2018	Retrospective Study	Cameroon	21
Elnaggar *et al.*	2018	Prospective Study	Egypt	20
Ezomike *et al.*	2018	Retrospective Study	Nigeria	26
Mohamed *et al.*	2018	Prospective Study	Egypt	40

Epidemiology

The prevalence of IHPS varies from one region of the world to the other and from one ethnic/racial group to another. It is a disease that is very common in Caucasians but less common among Africans and Asians, with reported incidence that ranged from 0.39/1000 newborns among the Taiwanese in Asia.[1] to 5/1000 newborns among the Caucasians.[5-7,51,52] There is a similar variation in the reported incidence of IHPS in the publications from Africa which ranged from 1/5500 to 12.9/1000 children.[27,28,40] Of the three publications that reported the incidence of IHPS, the reported incidence of 1/5500 live births from Tanzania[28] is considerably lower than the observed trends in other parts of the world.[1,3,8,50,53-56] This further suggests that IHPS is relatively less prevalent in the African population.

Risk factors

The etiology of IHPS is unknown; however, many risk factors have been suggested. Studies have reported interaction between genetic and environmental factors in the etiology of IHPS.[56,57] Environmental risk factors include infant breastfeeding pattern,[58] Helicobacter pylori infection,[59] maternal smoking,[60] and exposure to drugs.[61-63] Other factors which may be genetic include positive family history, twins, and maternal age. Reports on the relationship of breastfeeding to the incidence of IHPS have shown an inverse relationship, with some studies reporting a decrease in incidence with increasing frequency of breastfeeding,[64,65] others reported increased incidence with a decrease in the frequency of breastfeeding.[66] An outlook on the breastfeeding practices in Africa revealed that over 95% of infants are breastfed exclusively.[67-69] In a study of the effect of exclusive breastfeeding on the incidence of IHPS in Nigeria, the incidence of IHPS fell steadily from the late 1980s to the early 1990s when exclusive breastfeeding was introduced in the country.[35] This supports the earlier reports that breastfeeding may have a protective effect on the development of IHPS and this may account for the relatively lower incidence of IHPS in the African population,[26,33,70] where breastfeeding is widely practiced. The reported cumulative gender incidence reported in African publications is similar to the reported incidence in the Caucasians and Asians with IHPS more predominant in boys than girls.[1,4,5,71,72] Since the first report that linked the birth rank to IHPS as a risk factor,[73] many series have also discovered that the risk is highest with firstborn infant and declines with increasing birth order;[53,58,66,74-75] an observation that was also reported by the publications from Africa.[9,13,23,27,28,30,39,40,46,48]

Associated congenital anomalies have not been commonly reported in patients with IHPS supporting the belief that IHPS is more likely an acquired condition.

Key aspects of the study

Although only one case report tried to identify the etiology of IHPS at the genetic level,[32] it will be interesting if the relatively rare incidence of IHPS in Africans compared to Caucasians and Asians could stimulate extensive research on the etiology of the disease in the African continent.

In high-income countries (HIC), management of IHPS evolved from medical management to surgical management. Initially, IHPS was treated medically using antispasmodics such as atropine and scopolamine, whereas surgical management was adopted for cases with failed medical management and complicated cases.[44] Surgical management is now the gold standard in the management of IHPS. It has also evolved from open surgery to more cosmetically acceptable laparoscopic surgery that allows for quick recovery and early discharge from the hospital. The continent has witnessed slow progress in the evolution of management of IHPS as some centers still report medical management of pyloric stenosis. This probably may result from a lack of trained personnel or inadequate facilities for the surgical management of the patients.[36,44] Open surgery with varying surgical approaches is still very common in the continent, especially sub-Saharan Africa where many centers cannot boast of availability of pediatric laparoscopic facilities with inadequate experience in laparoscopic pyloromyotomy. The proximity to Europe of the few centers[39,47,49] which published studies on laparoscopic pyloromyotomy may account for the recent reports on laparoscopic management from the continent.

The reported morbidity and mortality of IHPS from the continent are still very high compared to outcomes from HICs.[11,18,46,48] Late presentation, lack of requisite facilities for diagnosis, non-availability of appropriate intravenous fluid, and inadequate resuscitation may account for this.

Limitations of study

The publication of articles in journals that are not indexed a very common phenomenon in the continent which reduces the global visibility of these articles is a major limiting factor. Furthermore, articles that did not use the term "Africa" may have been inadvertently excluded from the search.

CONCLUSION

The studies indicate that there is a gradual departure of publications from descriptive studies to comparative studies on newer modality of management of IHPS after the awareness about IHPS had been established. There were no publications on the etiology of IHPS to evaluate the interplay of genetics and environmental factors in Africans. Therefore, adequate funding should be provided by stakeholders to intensify research in this area as it relates to Africans. There is also the need to introduce more recent methods of management and to assess the outcome of these in Africa. Emphasis should also be made to publish these articles in indexed journals for more visibility and accessibility.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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