

Otitis media with effusion in Nigerian children with cleft palate: incidence and risk factors

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Abstract

Otitis media with effusion is common in children with cleft palate, and the aim of this study was to find out its incidence and risk factors in Nigerians. We prospectively studied 84 patients (42 with cleft palate and 42 control subjects); 27 were male and 15 female, who were age and sex matched with control subjects. The extent and size of the clefts were measured using a dental cast and Vernier calipers, and the otitis media was diagnosed with otoscopy and tympanometry. The mean (SD) age of the groups was 11 (7) months (range 1–33). Twelve children in the cleft group had otitis media compared with three in the control group. Infants and boys were more likely to be affected. There was a significant association between age ($p=0.02$), sex ($p=0.01$), and size of cleft ($p=0.00$). However, only the size of the cleft was confirmed to be an independent predictor, with children who had extremely wide clefts being more likely to develop otitis media than those with narrow clefts (OR = 8.71, 95%CI = 1.07 to 70.5). We conclude that the incidence of otitis media with effusion was higher among children with cleft palate than among those who did not have a cleft. Infants had a higher incidence than older children, and boys had a higher incidence than girls. Age, sex, and the size of the palatal cleft were significantly associated with otitis media, but not the extent of clefting.

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Introduction

Otitis media (OM) with effusion is common in infants and young children, and is debilitating if not recognised and managed early.¹ The Clinical Practice Guidelines on OM with effusion that were published in 2004 defined it as the accumulation of fluid in the middle ear without any signs or symptoms of acute infection,² and its pathogenesis is largely linked to dysfunction of the eustachian tube.³ Anatomical and physiological factors, with or without a pathological element, were reported to be the causes of such dysfunction.⁴ Symptoms of OM with effusion are insidious, so that if its diagnosis

is delayed it may ultimately result in complications such as speech defects and hearing loss.⁵

The association between cleft palate and OM with effusion is well documented.^{6–8} OM is reported to be almost universal among infants with cleft palate,⁴ which has led some authors to advocate early insertion of ventilation tubes in all children born with the condition.⁹ OM with effusion in cleft palate is caused by abnormal insertion of tensor palatini muscles and the shape of eustachian tube.¹⁰

It is difficult to calculate the incidence of OM, as it may not always be recognised or reported.¹¹ Its incidence in patients with cleft palate was reported to have ethnic variations, with that among Asians being much lower than among Europeans.¹² While much has been written about the incidence in other populations,^{5,11–13} none seems to have been reported among Africans. Risk factors in children who do not have clefts have been well documented,¹² but we know

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of no reports among children with clefts, so a report on the risk factors in the patients with clefts may help to eliminate delay in diagnosis.¹⁴ Our aim, therefore, was to establish the incidence and risk factors of OM with effusion in Nigerian children with clefts at our tertiary health care centre.

Subjects and methods

We designed and implemented a controlled, prospective, cohort study, for which ethics approval was obtained before the study started. The guardians or parents of all participants also gave written informed consent. All consecutive children with cleft palate alone who first presented to our clinic between March 2013 and April 2018 were recruited, and information on sample size was taken from a previous study.¹⁵ An age- and sex-matched control group was selected at random from those who attended for conditions unrelated to cleft palate.

Subjects were excluded if they had a history of: exposure to smoking, prolonged bottle feeding, attendance at the day care centre, recurrent acute otitis media, adenoidal hyperplasia, or upper respiratory tract infection, or if the cleft was syndromic. The clefts were then categorised according to their extent as described by Jensen et al.¹⁶ grade 1 = submucous or bifid uvula, grade 2 = soft palate cleft, grade 3 = soft palate and <1/3 hard palate, and grade 4 = complete cleft palate. We further subdivided them based on the size of the palatal cleft according to Badach's classification: narrow cleft = defect ≤ 1.5 cm; wide cleft = defect >2 cm; and extremely wide cleft = palatal defect $>60\%$ of the width of the entire palate or the width of both palatal shelves less than the width of the palatal defect.¹⁷ The size of the cleft was measured on a dental cast (Fig. 1) using calibrated self-retaining Vernier calipers (Tresna Ltd). The cleft was measured in millimetres using the inter-tuberosity points.¹⁸ The instrument was adjusted to an accuracy of 0.05 mm. All measurements were made twice at an interval of two days by a single observer (BEE) and the mean values used for analysis. To assess intra-observer agreement, we used the single measurement, absolute-agreement, 2-way mixed effect model to calculate the intra-class correlation coefficient (ICC). The definition of OM with effusion was based on detection of fluid in the middle ear behind an intact tympanic membrane without active inflammation,² and concurrent otoscopy of both ears was used to detect fluid while tympanometry was used to confirm OM with effusion.² Otological evaluation and assessment were made in the Department of Otorhinolaryngology.

We recorded age, sex, extent of cleft, size of cleft, and presence of OM with effusion. The primary predictive variable was size of the cleft, while the secondary predictive variables were age, sex, and extent of the cleft. The outcome variable was OM with effusion.

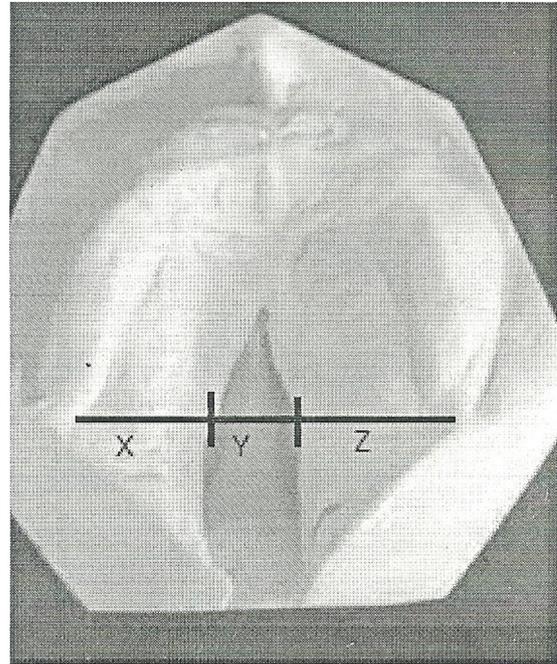


Fig. 1. Measurement of size of grade 3 palatal cleft. Y = size of cleft (19.5 mm).

Statistical analysis

In the descriptive analysis, continuous data were summarised as mean (SD) and range, and categorical data were summarised as frequency. Normality and homogeneity of variance of continuous data were checked with the Shapiro–Wilk and Levene's tests, respectively. In bivariate analysis, the significance of differences between continuous variables was assessed with an independent *t* test, while that between categorical variables was assessed with the chi squared test for homogeneity or Fisher's exact test where applicable. In multivariate analysis, associations between predictive and outcome variables were tested with the chi-squared test for independence, while predictions were made with binomial logistic regression. All analyses were done with the help of the SPSS software package (version 17, SPSS Inc), and probabilities of less than 0.05 were accepted as significant.

Results

A total of 84 patients (42 in each group) were studied. The mean (SD) age was 11 (7) months, range 1–33.

Twelve of the 42 patients developed OM with effusion as did three in the control group ($p=0.00$). The infants (aged 1–12 months) had the highest incidence of OM while the older children (25–36 months) had the least in both groups (Table 1). The boys and those in the control group had a higher incidence of OM than the girls (Table 2).

Table 1

The incidence of otitis media (OM) with effusion in patients and controls according to age.

Age (months)	Patients (n = 42)		Controls (n = 42)	
	OM present	OM absent	OM present	OM absent
1–12	7	4	2	18
13–24	4	13	1	12
25–36	1	13	0	9
≥37	0	0	0	0
Total	12	30	3	39

Table 2

The incidence of otitis media (OM) with effusion in patients and controls according to sex.

Sex	Patients (n = 42)		Controls (n = 42)	
	OM present	OM absent	OM present	OM absent
Male	9	18	3	24
Female	3	12	0	15
Total	12	30	3	39

Table 3 shows the univariate analysis between OM and the risk factors. The age, sex, and size of palatal clefts significantly influenced the development of OM, while the extent of the palatal clefts among the patients did not ($p = 0.17$).

In the multivariate analysis, the size of the palatal cleft was confirmed as an independent predictor of OM ($p = 0.02$), unlike age ($p = 0.13$), or sex ($p = 0.28$). The children with extremely wide palatal clefts were more likely to develop OM than those with narrow clefts (OR = 8.21. 95% C.I = 1.02 to 70.6).

There was almost perfect intra-observer reliability among the two measurements with an ICC value of 0.96, 95% CI 0.92 to 0.99 for mean measures. This showed that reliability of the variables chosen to identify the extremities of the cleft was good.

Table 3

Patients' characteristics and otitis media (OM) with effusion: univariate analysis.

Variables	OM		p value	OR (95% CI)
	Present (n = 12)	Absent (n = 30)		
Age (months):				
>12	3	17	0.02	3.17 (1.18 to 8.52)
≤ 12	9	13		
Sex:				
Male	8	20		
Female	4	10	0.01	3.85 (1.34 to 11.0)
Extent of cleft:				
Grade 1	1	3		
Grade 2	2	15	0.17	2.60 (0.57 to 11.9)
Grade 3	5	5		
Grade 4	4	7		
Size of cleft:				
Narrow	1	22		
Wide	4	7	0.00	13.22 (1.7 to 90.2)
Extremely wide	7	1		

Discussion

Since the incidence of OM with effusion in patients with cleft palate was reported in 1969 by Paradise et al⁸ reports from other parts of the world have shown that it ranges from 75%–95% (Table 4).^{11–19} Our data show that in Nigeria the incidence might be lower than that predicted by other global studies,^{2,8,7,6} but it is difficult to calculate the incidence, and reported incidences vary.¹¹ Because the symptoms of OM are insidious,⁵ some patients may not present to an otorhinolaryngologist, and the condition may also go unnoticed by oral and maxillofacial surgeons and the parents. Chen et al¹² reported that although the incidence of cleft palate is higher in Asians than in Europeans, the incidence of OM among children with cleft palate is the reverse, because Oriental people have significantly small maxillas and mandibles, more severe mandibular retrognathism, increased total and upper facial height, and steeper and shorter anterior cranial bases than Europeans. Like the lower incidence of cleft palate among black populations reported by previous studies,^{13,18} our study seems to show that the lowest incidence of OM in children with cleft palate is among black people. The likely reasons could be related to the smaller incidence of cleft palate in our environment,¹³ together with the increased mandibular height, and increased depth and height of the mastoid in black infants.^{20,21}

Children with cleft palate are known to be much more likely to develop OM than normal children and they develop it at an early age,^{12,16} which was supported by our findings (Table 1). The abnormal insertion of the tensor and levator palatini muscles, immature immune system, failure of the eustachian tube to close, abnormal compliance of the eustachian tube, increased propensity for middle ear infections, and reduced depth and height of the mastoid in these patients could be contributory factors.¹⁰ We found a higher incidence in boys than girls, which confirms the results of pre-

Table 4
Worldwide incidence of otitis media with effusion and cleft palate.

First author and reference	Period of study (years)	Country	Type of study	Duration of study (years)	Sample size	Incidence
Kwan ⁵	2011	China	Retrospective	10	104	76.1
Flynn ¹⁰	2009	US	Controlled prospective	5	22	74.7
Sheahan ¹¹	2002	Ireland	Prospective	5	104	95.2
Chen ¹²	2012	Taiwan Spain	Prospective	5	319	71.9
Valtonen ¹³	2005	Finland	Prospective	10	51	98.0
Sancho ¹⁴	1997		Retrospective	5	40	84.8
Present study	2018	Nigeria	Controlled prospective	5	42	29.0

vious studies.^{13,15} The increased hyperactivity in boys may have adverse effects on the functionality of the eustachian tube, as it was thought that boys are more hyperactive than girls.²¹

We found that infants aged less than 12 months, boys, and those with extremely wide clefts tend to develop OM, but this association was not confirmed by the extent of clefting (Table 3). Multivariate analysis confirmed an independent association between size of cleft and OM, but this was not the same with the age and sex of the patient, which indicates that as the patients grow older from infancy, OM tends to resolve.^{14,17}

Tympanometry is a dynamic and objective test that provides information about the compliance or mobility of the tympanum-ossicular system as a result of changes in the air pressure in the external auditory canal. The use of tympanometry in children and adults is well-established, but its use for infants is still controversial.^{22,23} Publications recommend 1000 Hz tympanometry for infants aged less than 6 months, which has a greater sensitivity for the correct identification of middle ear dysfunction in this group.²³ There are a number of difficulties associated with the tympanometry test in infants.²³ Diagnostic problems may result from the presence of wax in the ear canal, lack of cooperation, incorrect placement of the tympanometry probe in the ear, the narrow tympanic-ossicular system, and the poor sensitivity of conventional tympanometry (227 Hz).²³ To achieve reliable readings in infants less than 6 months old, we used 1000-Hz tympanometry, while conventional tympanometry (226-Hz) was used for children over 6 months.²³ Multiple or repeated tests were done for infants who were uncooperative to obtain more reliable records.

The main reason for concern about persistent OM with effusion in children with or without cleft palate is the potentially adverse effect of reduced acuity of hearing, speech, and language, as well as mental development long after the OM has resolved and the hearing has returned to normal.²⁴ Though the effects of surgical or conservative management, or both, on OM still remain controversial, there is a belief that early management of OM with insertion of grommets at the time of palatoplasty is beneficial, despite the complications that have been reported.²⁴ The multidisciplinary team protocol, therefore, still remains the best management for this group of patients. Because the resources for speech assessment in this part of the world are regarded as a luxury, and

because the follow-up required for accurate assessment of speech is long-term, we were unable to collect this information.

Although we did not assess the outcomes of hearing and speech, to the best of our knowledge this paper is one of the first to establish the incidence of OM with effusion in patients with cleft palate in a black population, and also among the first to list the confounding factors of OM in patients with cleft palate globally. However, these risk factors and a follow-up assessment of the outcomes of speech and hearing need to be validated by further studies with larger samples.

Conclusion

We found that the incidence of OM with effusion in the cleft palate among Nigerians with and without cleft palate were 12/42 and 3/42, respectively. The age, sex, and the size of the cleft of the patients were significantly associated with OM in patients with cleft palate, while the extent of the cleft was not significantly associated.

Conflict of interest

We have no conflicts of interest.

Ethics Statement/Confirmation of patients' permission

The approval of the institution's Ethics and Research Committee was obtained, and signed informed consents were given by guardians or parents of the participants.

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