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CASE SERIES

Paediatric Bronchiectasis in a Resource-Constrained Centre: A Case Series

Kuti BP^{*1}, Jallow M², Jobarteh ML²

¹Department of Paediatrics and Child Health, Obafemi Awolowo University, Ile-Ife, Nigeria

²Department of Paediatrics, Edward Francis Small Teaching Hospital, Banjul, The Gambia

***Correspondence: Professor BP Kuti, Department of Paediatrics and Child Health, Obafemi Awolowo University, Ile-Ife, Nigeria. E-mail: bkuti@oauife.edu.ng ; ORCID - <https://orcid.org/0000-0002-4778-4362>.**

Summary

Bronchiectasis denotes fixed and often irreversible dilatation of the bronchial wall caused by persistent inflammation and infection. Children with bronchiectasis in developing countries suffer recurrent hospitalisation and poor quality of life and usually succumb to the condition. These children are poorly recognised, misdiagnosed, and under-reported. We report five cases of computerised tomography-diagnosed paediatric bronchiectasis managed at the Edward Francis Small Teaching Hospital (EFSTH), Banjul, The Gambia. The ages of the children at presentation ranged from 3 to 14 years, and they all had recurrent chest infections. Four out of the five cases had Pulmonary Tuberculosis, among whom two cases had HIV/TB co-infections, and the only non-TB, non-HIV case had features of Down syndrome with congenital heart lesions. All the cases had growth impairment; digital clubbing was observed in four, and low peripheral oxygen saturation in room air in three of the five children. These cases are reported to increase the index of suspicion among clinicians working in resource-limited settings to consider the diagnosis of bronchiectasis in children with recurrent chest infections. Early diagnosis and prompt management, including appropriate follow-up care, will improve the quality of life of these children and ensure better management outcomes.

Keywords: *Bronchiectasis, Post-TB lung disease, Recurrent chest infection, Resource-poor setting, Tuberculosis.*

Introduction

Bronchiectasis is a term used to describe a respiratory disorder characterised by fixed and often irreversible dilatation and thickening of the bronchi wall, often from recurrent infections and inflammation leading to progressive deterioration of lung functions. ^[1] It is usually the final common pathway for several lung pathologies that cause persistent inflammation, infection and obstruction. ^[2] The aetiological

factors associated with childhood bronchiectasis range from congenital anomalies of the bronchi, including the yellow nail syndrome, Campbell-William syndrome, and trachea-bronchomegaly, to genetic conditions like cystic fibrosis and recurrent or persistent chest infections. ^[1, 2]

In developed countries, cystic fibrosis (CF) is a leading cause of bronchiectasis and conditions like primary immune deficiency and defects in mucociliary clearance mechanisms have also

been associated with the bronchiectasis. [3, 4] In developing countries, however, chronic infections, including tuberculosis, recurrent chest infections like pneumonia and foreign body aspiration, predominate as the leading causes of bronchiectasis. [2] Chest computerised tomographic (CT) scan has replaced bronchography as the main investigative modality for diagnosing bronchiectasis. [1] Bronchiectasis is present when the internal luminal diameter of the bronchus is greater than the adjacent blood vessel. Bronchial wall thickness may also be present, often called 'peribronchial thickening' as bronchial wall thickness is equal to or larger than the diameter of the adjacent vessel. [1] Reid's histopathologic classification of bronchiectasis into three phenotypes in 1950, is still very relevant regarding clinical outcomes. [5] These phenotypes include the tubular type, characterised by smooth dilation of the bronchi; the varicose type, characterised by dilated bronchi with multiple indentations; and the cystic type, in which dilated bronchi terminate in blind-ending sacs. [5]

The pathogenetic mechanisms underlying bronchiectasis are still not fully understood. It is generally believed that bronchiectasis arises from poorly regulated and orchestrated responses to inflammatory processes initiated by respiratory pathogens. [1, 6, 7] This perpetuates the vicious cycle of inflammation, tissue destruction, obstruction and further tissue destruction, as proposed by Cole in 1986. [6-9] Prompt and adequate treatment of respiratory infections, including tuberculosis, is essential for preventing bronchiectasis. [1-3] Additionally, individuals with recurrent infections should be carefully investigated, and underlying aetiological factors associated with the disease should be treated to halt the vicious cycle. Consequently, the prevalence of non-cystic fibrosis bronchiectasis in developed countries is reducing. For instance, a

prevalence of one in 6000 paediatric population was reported in Auckland, New Zealand, [3] while 0.5 per 100 000 was reported from the UK and 2.3 per 100 000 from Ireland. [4] There is, however, a paucity of reports from developing countries, particularly sub-Saharan Africa, where the challenges of diagnosing, managing and reporting the condition in children are enormous.[2]

We report five cases of paediatric bronchiectasis managed at the Edward Francis Small Teaching Hospital, Banjul, the Gambia - the only tertiary health facility in The Gambia. We highlight the clinical manifestations, radiological findings and the underlying aetiological/predisposing factors of these cases, which had hitherto remained undiagnosed. These case series may increase the index of suspicion of clinicians working in developing countries in suspecting and prompt diagnosis of bronchiectasis in children.

Case series

The salient clinical and radiologic findings in the five cases are highlighted in Table I. The ages of the children ranged from three to 14 years, and the duration of illness before presentation to the hospital ranged from one week to 5 months. Cough, progressively worsening difficulty in breathing and easy fatigability were the presentations common to all the children. Additionally, two (40.0%) of the cases presented with pedal oedema and four (80.0%) presented with digital clubbing. Three (60.0%) were hypoxaemic in room air at presentation, with the peripheral oxygen saturation (SPO₂) ranging from 37% to 99% in room air. All the children were malnourished, with two (40.0%) of them severely wasted (Table I).

Table I: Clinical and radiologic findings in children with bronchiectasis

Clinical and laboratory findings	Case 1	Case 2	Case 3	Case 4	Case 5
Age (years)	4	3	5	11	14
Sex	Male	Female	Male	Male	Female
Duration of illness before recent presentation	2 weeks	1 week	5 months	4 months	3 months
Weight for height (Z score)	< -2SD	<-4SD	<-3SD	<-3SD	<-2SD
Presence of oedema	Yes (Grade 3)	Yes (Grade 1)	No	No	No
Digital clubbing (Grade)	Yes (Grade 4)	Yes (Grade 3)	Yes (Grade 1)	Yes (Grade 1)	No
Peripheral Oxygen saturation in room air	37%	56%	99%	88%	95%
Chest X-ray findings	Diffuse Honeycomb appearance in both lung fields	Honeycomb appearance in the left lower lobe and opacities on the right	Homogenous opacity on the left middle and lower lung zones	Patchy perihilar opacities	Honeycomb appearance
Chest CT scan findings	Bilateral cystic & tubular bronchiectasis	Left lung bronchiectasis with right lung pneumonia and diffuse inflammatory interstitial oedema	Left apical tubular bronchiectasis	Bilateral cystic bronchiectasis	Tubular bronchiectasis
Possible aetiology(ies)	HIV-TB infection	co- Down syndrome with Recurrent Pneumonia and AVCD*	Pulmonary tuberculosis	Pulmonary tuberculosis	HIV-TB co-infection

*AVCD atrioventricular canal defects

Case 1 was confirmed HIV-1 infected two years before presentation and had been on antiretroviral treatment (ABC, 3TC, NVP). He was also in the fourth week of anti-tuberculosis treatment for smear-negative pulmonary tuberculosis (PTB). GeneXpert revealed MTB was not detected, but therapy for PTB was commenced because of chronic cough, weight loss, history of contact with individuals with chronic cough and chest X-ray highly suggestive of PTB. He had four hospital admissions in the past year on account of chest infections. Both parents were also on treatment for HIV. The chest X-ray at presentation is shown in Figure 1.

He was managed with intravenous cefuroxime and oral azithromycin. Antiretroviral and anti-TB medications were continued, and he was stable enough to be discharged home after three weeks of hospitalisation. He is currently being followed up at the Paediatric Chest Clinic and the Physiotherapy unit of the hospital.

Case 2 had features of Down syndrome, including global hypotonia, flat facial profile, prominent epicanthic folds, tongue protrusion, simian crease and low-set ears. She also had pectus carinatum, dull percussion notes with decreased breath sounds on the right lung field, and crepitations at the left lower lung zone.



Figure 1: Chest radiograph of Case 1 with bilateral honeycombing of the lung parenchyma

In the cardiovascular system, she had anterior bulging of the cardiac area, displaced apex beat to the fifth intercostal space anterior axillary line, a grade 3 pansystolic murmur loudest at the left lower sternal border. A chest radiograph revealed hyperinflated lung fields with bilateral bronchiectatic changes that were more prominent on the right. (Figure 2) Chest CT confirmed deformity of the thoracic skeleton (pectus carinatum). Additional reports from the chest CT included an increased volume of the right lung and a decrease on the left with hyperaeration. There is a honeycombing image in the left lung, keeping with bronchiectasis. There is consolidation with an air bronchogram in the posterior inferior segment of the inferior lobe of the right lung. Also, there is an interstitial pattern (diffuse ground glass opacity in the middle and inferior lobe of the right lung - inflammatory interstitial oedema). The heart was enlarged but there was no lymphadenopathy. Normal soft tissue outlines were also reported. Left lung bronchiectasis with right lung pneumonia and diffuse inflammatory interstitial oedema was diagnosed. Echocardiography revealed a large ventricular septal defect (VSD). Gastric lavage for GeneXpert and AFB and retroviral screening were all negative. She was managed with

intravenous antibiotics, oxygen therapy, diuretics and digoxin. She is presently being co-managed by the pulmonology and cardiology teams.

Case 3 had earlier been diagnosed with PTB and he was on the fifth month of anti-TB therapy. He was managed for right-sided pleural effusion secondary to pneumonia six months ago and had earlier been managed for diaphragmatic hernia in infancy. Retroviral screening was negative, and gastric lavage for AFB was also negative for MTB. Chest X-ray revealed homogenous opacity on the left middle and lower lung zones, while chest CT scan showed left lobar consolidation with pneumatoceles and bullae; there was also left apical bronchiectasis with pleural thickening. A diagnosis of acute exacerbation of bronchiectasis secondary to PTB was made. Anti-TB therapy was continued, in addition to antibiotics, including azithromycin. He is being followed up at the pulmonology clinic.

Case 4 has had a total of eight previous hospital admissions at peripheral private and public health facilities receiving care for respiratory conditions, including severe pneumonia and asthma. There was a history of contact with an

adult with chronic cough and weight loss. He has also tried various unorthodox and traditional

(complimentary) medicines with no significant improvement.

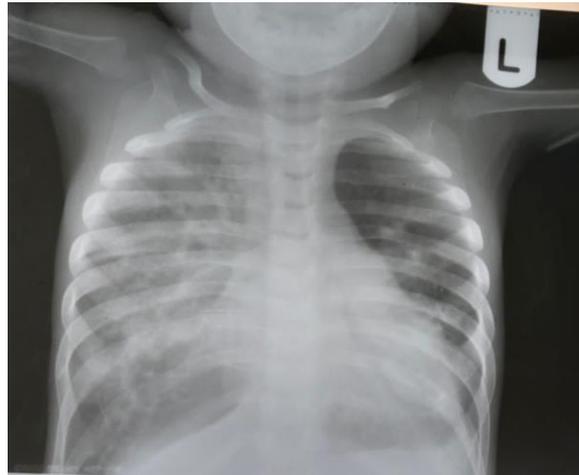


Figure 2: Chest radiograph of the three-year-old with Down syndrome, VSD and bronchiectasis

Respiratory examination revealed reduced chest expansion on the right, increased tactile fremitus and vocal resonance, and bronchial breath sound in the same region. There were, however, hyper resonant percussion sounds on the left. Other systemic examinations were essentially normal. Chest X-ray revealed patchy opacities on the left middle and lower lung zones and right perihilar opacities (Figure 3). Chest CT revealed left lung

consolidation with peripheral bilateral inflammatory consolidations. There was also diffused areas of cystic bronchiectasis bilaterally with right hilar lymph nodes enlargement suggestive of TB (Figure 4). Sputum AFB and Gene Xpert were negative for *Mycobacterium tuberculosis*. He was commenced on anti-TB drugs and is presently being followed up at the clinic with significant improvement.



Figure 3: Chest X-ray showing bilateral opacities

Case 5 was diagnosed to be HIV-infected and was previously treated for TB and presumed to have completed treatment four months prior to

the latest presentation. She was admitted with the diagnosis of right-sided lung consolidations with pulmonary fibrosis (Post TB syndrome) to

rule out relapsed PTB in a known HIV-infected patient. Chest X-ray showed right lung opacities with a honeycomb appearance but her sputum AFB was negative.

Chest CT scans revealed right middle and lower lung bronchiectasis. She was admitted and

treated with intravenous Cefotaxime, oral Cotrimoxazole, Azithromycin and anti-TB medications, while the HAART was also continued. She is on follow-up care at the Respiratory Clinic and doing well on her medication.



Figure 4: Chest CT of case 4 showing diffuse bilateral cystic bronchiectasis changes in the lung parenchyma

Discussion

This case series highlighted five cases of childhood bronchiectasis diagnosed with chest CT scans. The children's ages ranged from 3 to 14 years, and all the children had recurrent chest infections, including Tuberculosis (TB) disease. The children all had chronic lung disease, which had hitherto not been diagnosed, and the majority already had features of chronic hypoxaemia, including digital clubbing and growth impairment. This implies that recurrent chest infections in children should be appropriately investigated for underlying causes/effects for better outcomes.

Concerning the underlying possible aetiologies of the cases with bronchiectasis, within the limit of our investigative capacities, 80.0% of the children in our series had a present or past history of pulmonary tuberculosis either alone

(40.0%) or co-infected with HIV (40.0%). TB has been reported as the leading aetiological cause of paediatric bronchiectasis in developing countries. [2, 10] Children with TB, particularly when not promptly diagnosed and adequately treated, are likely to develop complications, including lung fibrosis and traction bronchiectasis. [10] Delayed diagnosis and a massive burden of TB in adults in the Gambia [11] and other developing countries continue to constitute a reservoir for paediatric TB with its resultant post-TB chronic lung disease, including bronchiectasis. Poor drug adherence in childhood TB has been recognised as another essential factor associated with the development of pulmonary complications in children with TB, as TB-HIV co-infection increased pill burden, makes the already difficult diagnosis of TB, more complicated and accelerates lung destruction, including fixed and dilated bronchi as found in children with bronchiectasis. [10] This implies that

children with TB-HIV co-infection should be closely monitored and supported to ensure drug adherence and better outcomes. HIV and TB-associated bronchiectasis has been described as being neutrophil-driven in which T-helper cells Type 1 (Th1) induced cytokines like IL-1, IL-6, GM-CSF and IFN- γ play significant roles. [10] These cytokines reflect the body's ability to mount immune responses to infectious agents, thereby perpetuating the vicious cycle of infection, inflammation, exudative airway obstruction and ultimately, airway destruction and fixed dilatation of the bronchi. [7-9]

The only non-TB, non-HIV case in this series had Down syndrome with a VSD, which predisposes the child to recurrent chest infection and recurrent hospitalisation. Cystic lung diseases, bronchiectasis and recurrent chest infections have been reported in children with Down syndrome with or without associated congenital heart diseases. [12] As Cole proposed in his vicious cycle hypothesis, recurrent chest infections lead to bronchial lumen obstruction from inflammatory exudates; a vicious cycle of obstructions and infections lead to fixed and irreversible dilation of the bronchi, hence the development of bronchiectasis. [6, 7-9] Unfortunately, cardiac repair of the VSD is not readily available in The Gambia and in many parts of low- and middle-income countries (LMICs), making these children live with unrepaired cardiac lesions with its attendant complications including recurrent chest infections and bronchiectasis. [13] Making appropriate cardiac interventions available and affordable for families of children with cardiac lesions may reduce the burden of consequent chronic lung disorders associated with such cardiac lesions.

The management of bronchiectasis in children is very challenging, more so in LMICs where facilities for definitive diagnosis and prognostication are not readily available. [2, 14-16]

The European Respiratory Society (ERS) and other bodies highlight the guidelines for investigating and managing bronchiectasis in children. [15-17] High-resolution CT scan, bronchoscopy, and bronchoalveolar lavage may be necessary for cellular and microbiological analyses to identify aetiologies and superimposed microbial agents that may cause exacerbations. [16, 17] Long-term antibiotics, especially azithromycin, have been reported to reduce airway and systemic inflammation in non-cystic fibrosis bronchiectasis. [18] Hence, all the children received azithromycin in addition to the specific treatment of the underlying aetiologies.

This case series reports the clinical manifestations, history, and possible aetiologies of paediatric bronchiectasis, which was diagnosed using a computerised tomographic scan of the chest in a resource-poor centre. This constitutes the strength of this report. However, we recognise that the use of bronchoscopy and bronchoalveolar lavages to characterise the aetiology further [16] and define the microbial agents responsible for this condition, were not done due to a lack of facilities. Also, primary immunodeficiency states were not searched for due to lack of facilities. Nonetheless, this report will add to the scanty information about paediatric bronchiectasis from a resource-poor centre. It is aimed at increasing the index of suspicion of clinicians in a similar set up to look for and promptly detect bronchiectasis for management, including adequate parental/caregiver and child counselling. The importance of chest imaging, particularly chest CT, in diagnosing bronchiectasis must be emphasised; hence, the availability of the facility in centres in developing countries will increase the recognition and characterisation of the disease in children.

Conclusion

All the children in this case series had recurrent chest symptoms requiring hospital visits. Tuberculosis and HIV are the major risk factors for developing bronchiectasis in the majority of the children reported, hence detailed investigations of children with recurrent chest infection including chest CT scan, Genexpert and retroviral screening are, therefore, important for prompt recognition of underlying chest pathology and possible aetiology for optimal management, especially in LMICs. We hope this case series will increase the index of suspicion of bronchiectasis in children with recurrent chest infections in developing countries.

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References

1. Dagli E. Non-CF bronchiectasis. In: Eber E and Midulla F (Editors) ERS Handbook Paediatric Respiratory Medicine. European Respiratory Society. 2013; 253-257.
2. Karadag B, Karakoc F, Ersu R, Kut A, Bakac S, Dagli E. Non-cystic fibrosis bronchiectasis in children: a problem in developing countries. *Respiration* 2005;72:233–238. <https://doi.org/10.1159/000085362>.
3. Edwards EA, Asher MI, Byrnes CA. Paediatric bronchiectasis in the twenty-first century: Experience of a tertiary children's hospital in New Zealand. *J Paediatr Child Health* 2003;39:111-117. <https://doi.org/10.1046/j.1440-1754.2003.00101.x>.
4. Brower KS, Del Vecchio MT, Aronoff SC. The etiologies of non-CF bronchiectasis in childhood: a systematic review of 989 subjects. *BMC Pediatr* 2014;14:299.10. <https://doi.org/1186/s12887-014-0299-y>.
5. Reid L. Reduction in bronchial subdivisions in bronchiectasis. *Thorax* 1950;5:223–247. <https://doi.org/10.1136/thx.5.3.233>.
6. Pizzutto SJ, Hare KM, Upham JW. Bronchiectasis in Children: Current Concepts in Immunology and Microbiology. *Front Pediatr* 2017;5:123. <https://doi.org/10.3389/fped.2017.00123>.
7. Cole PJ. Inflammation: a two-edged sword – the model of bronchiectasis. *Eur J Respir Dis Suppl* 1986;147:6–15.
8. King PT. The pathophysiology of bronchiectasis. *Int J COPD* 2009;4:411–419. <https://doi.org/10.1016/j.prrv.2010.10.011>.
9. Grimwood K. Airway microbiology and host defences in paediatric non-CF bronchiectasis. *Paediatr Respir Rev* 2011;12:111–1118. <https://doi.org/10.1016/j.prrv.2010.10.009>.
10. Masekela R, Anderson R, Moodley T, Kitchin OP, Risenga SM, Becker PJ, et al. HIV-related bronchiectasis in children: an emerging spectre in high tuberculosis burden areas. *Int J Tuberc Lung Dis* 16:114–119. <https://doi.org/10.5588/ijtld.11.0244>.
11. Owolabi OA, Jallow AO, Jallow M, Sowe G, Jallow R, Monica D. Genekaha MD, et al. Delay in the diagnosis of pulmonary tuberculosis in The Gambia, West Africa: A cross-sectional study. *Int J Infec Dis*

- 2020;101:102-106.
<https://doi.org/10.1016/j.jiid.2020.09.029>.
12. George M, Amodio J, Lee H. Cystic Lung Disease in Down Syndrome: A Case Report and Literature Review. *Case Rep Pediatr*. 2016;2016:4048501.
<https://doi.org/10.1155/2016/4048501>.
 13. Dearani JA, Neirotti R, Kohnke EJ, Sinha KK, Cabalka AK, Barnes RD, *et al*. Improving pediatric cardiac surgical care in developing countries: matching resources to needs. *Semin Thorac Cardiovasc Surg Pediatr Card Surg Ann* 2010;13:35-43.
<https://doi.org/10.1053/j.pcsu.2010.02.001>.
 14. Lavery A, Jaffe A, Cunningham S. Establishment of a web-based registry for rare (orphan) pediatric lung diseases in the United Kingdom: the BPOLD registry. *Pediatr Pulmonol* 2008;43:451-456.
<https://doi.org/10.1002/ppul.20783>.
 15. Zaid AA, Elnazir B, Grealley P. A decade of non-cystic fibrosis bronchiectasis 1996-2006. *Ir Med J* 2010;103:77-79.
 16. Pizzutto SJ, Grimwood K, Bauert P, Schutz KL, Yerkovich ST, Upham JW, *et al*. Bronchoscopy contributes to the clinical management of indigenous children newly diagnosed with bronchiectasis. *Pediatr Pulmonol* 2013; 48:67-73.
<https://doi.org/10.1002/ppul.22544>.
 17. Chang AB, Grimwood K, Boyd J, Fortescue R, Powell Z, Kantaret A. Management of children and adolescents with bronchiectasis: summary of the ERS clinical practice guideline. *Breathe* 2021;17:210105.
<https://doi.org/10.1183/20734735.0105-2021>.
 18. Chalmers JD, Smith MP, McHugh BJ, Doherty C, Govan JR, Hill AT. Short- and long-term antibiotic treatment reduces airway and systemic inflammation in non-cystic fibrosis bronchiectasis. *Am J Respir Crit Care Med* 2012;186:657-665.
<https://doi.org/10.1164/rccm.201203-0487OC>.



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