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Case report

Unilateral choanal atresia and a co-existent long-standing medium-sized ipsilateral rhinolith in a 15-year old boy: Case report and literature review

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ABSTRACT

Introduction and importance: Unilateral choanal atresia is a congenital anomaly where a newborn baby is born with a unilateral imperforate posterior nare. In most of the time the diagnosis may be missed for years after birth. A rhinolith is an entity formed by gradual deposition and coating of different salts of calcium and magnesium over an endogenous or exogenous nidus in the nasal cavity. Coexistence of a rhinolith and choanal atresia is a very rare encounter in clinical practice and to the best of our knowledge this is perhaps the first documented case in Tanzania.

Case presentation: We present a 15-year old boy who was attended at our department with a longstanding history of left sided non-foul smelling nasal discharge which was noticed first when he was 5 years old but at the age of 13 years, he presented with ipsilateral nose bleeding and episodic foul smelling nasal discharge. He was attended at various peripheral health facilities without relief.

Clinical discussion: The patient underwent left sided nasal endoscopy where unilateral choanal atresia and a rhinolith were found. Transnasal endoscopic choanal atresia release and rhinolith removal were done under general anaesthesia in operating room. Postoperatively, he was kept on a nasal decongestant, a broad-spectrum antibiotic, intranasal corticosteroid and an analgesic.

Conclusion: Clinicians must have a high index of suspicion so as to establish the diagnosis of unilateral choanal atresia in patients with persistent unilateral non-foul smelling discharge and also nasal foreign bodies in those with foul smelling nasal discharge.

1. Introduction

Unilateral choanal atresia is a rare congenital anomaly where a newborn baby is born with a unilateral imperforate posterior nare [1,2]. Such anomaly occurs due to a persistent oronasal membrane when it fails to break down at 38 weeks of gestational age [2]. The right nostril is commonly affected more than the left at a ratio of 2:1 in unilateral choanal atresia [2–5].

When congenital choanal atresia is unilateral, infants may go undiagnosed until when unilateral rhinorrhea and nasal obstruction necessitate seeking medical care [6]. Unlike unilateral choanal atresia that may go undiagnosed for a couple of years its counterpart named bilateral choanal atresia in most cases is diagnosed immediately after birth since the affected newborns presents with respiratory distress immediately after birth and thus a neonatal otorhinolaryngological emergency [1]. The reason as to why neonates with bilateral choanal atresia presents with a state of respiratory distress immediately after birth is due to their behavior of being obligate nasal breathers up to 6 weeks of life as they tend to have a larynx with a high position in the neck from the second to third cervical vertebrae [2].

Rhinolith is an entity formed by gradual deposition and coating of different salts of calcium and magnesium over an endogenous or exogenous nidus in the nasal cavity [7–10]. The type, size and duration of the rhinolith lead to multiple types of presentation. The insidiousness and gradual development leads to gradual symptoms developing years following rhinoliths formation and therefore causing persistent or recurrent nasal infections. The predominant clinical features are unilateral nasal obstruction and foul smelling nasal discharge [11]. Other features for rhinoliths include cacosmia, epistaxis, headache, facial pain and epiphora [12–14].

Regarding the location of nasal foreign bodies, the commonest location is the floor of the nose at the junction between anterior and

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posterior nares [15,16]. Under normal circumstances, nasal foreign bodies tend to be positioned anteriorly but due to normal physiological action, movement posteriorly occurs and thus a favorable location [13]. Deposition of salts responsible for formation of rhinoliths may be attributed by certain local inflammatory reactions around the focal nidus for rhinoliths that leads to deposition of salts of calcium phosphate, magnesium, calcium carbonate, and aluminium. Such changes around focal nidus make it hard in texture and the size of the rhinolith increases gradually [12,13]. In this report we describe a rare case of unilateral choanal atresia and a co-existent medium sized rhinolith in a 15-year old boy that was managed by transnasal endoscopic choanal atresia release and foreign body removal. The work has been reported in line with the SCARE 2020 [17].

2. Case presentation

A 15-year old secondary school student presented to our otorhinolaryngology clinic with a longstanding history of left sided non-foul smelling nasal discharge which was noticed first when he was 5 years old but at the age of 13 years, he presented with ipsilateral nose bleeding and episodic foul smelling nasal discharge. Multiple visits to peripheral health facilities in Southern Tanzania assured the mother that nasal obstruction and nasal discharge would subside with time because they were due to allergic rhinosinusitis. He was kept on repeated doses of antihistamines, oral corticosteroids, intranasal corticosteroids, decongestants and antibiotics without permanent relief. His mother denied prenatal cigarette smoking, alcohol or caffeine consumption and there was also no reported maternal history suggestive of prenatal thyrotoxicosis. There was also no family history of similar illness. He opted self-referral for further management upon no permanent resolution of the complaints.

Upon nasal endoscopy, a friable mucosal lining of the left nasal cavity was noted and some granulation tissues at the mid part of the nasal cavity were found. Further advancement of the rigid scope revealed an atretic left choana (Fig. 1). Computerized tomography scan (CT scan) of the paranasal sinuses with contrast was ordered and an atretic left choana was found and was predominantly bony in nature (Fig. 2). Physical examination revealed no presence of any associated congenital anomalies.

Results from laboratory tests revealed the following; negative HIV serology, elevated erythrocyte sedimentation rate (30/h) and hemoglobin of 12 g/dl.

The patient was planned for transnasal endoscopic choanal atresia release under general anaesthesia and intraoperatively a coexisting rhinolith surrounded by abundant granulation tissues was encountered. Such procedure was performed successfully where intraoperatively the rhinolith was removed (Fig. 3) and also choanal atresia release was done by taking down the vomer and the atretic choanal plate (Fig. 4). No stent was kept in situ. Postoperatively, he was kept on intravenous ceftriaxone 1 g once daily for 3 days, intravenous paracetamol 1 g 8-hourly for 3 days and ephedrine (1 %) nasal drops 8-hourly for 5 days. Upon discharge, he was prescribed tablets amoxicillin/clavulanic acid 625 mg 12-hourly for 7 days, tablets paracetamol 1 g 8-hourly for 7 days and fluticasone furoate nasal spray 12-houly for 30 days. He was also advised to continue with ephedrine (1 %) nasal drops for 5 days. He was kept under close postoperative follow up on weekly basis for the first month and then monthly for 3 months. He later on absconded from further postoperative follow up.

He was kept under follow up for three months with complete healing of the neochanae (Fig. 5).

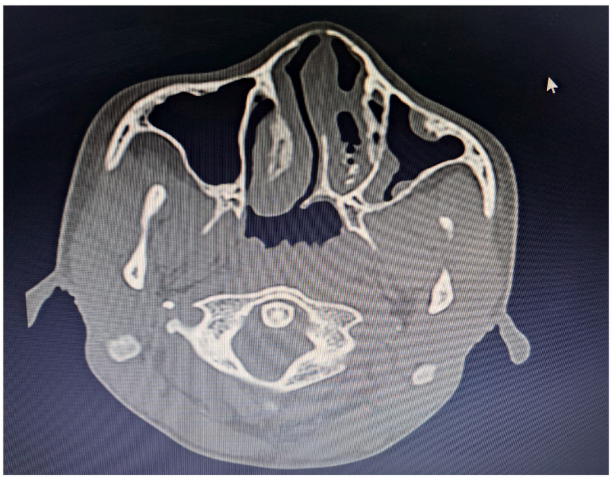

Fig. 2. Axial view of CT scan of the paranasal sinuses showing the predominant bony left atretic choana.

3. Discussion

Unilateral choanal atresia is not as common as bilateral choanal atresia and less than half of patients with unilateral atretic choana have other associated anomalies such as coloboma of the eye, heart defects, growth retardation, genital and/or urinary defects, ear anomalies (CHARGE Syndrome). Such patients with choanal atresia and other congenital anomalies require multidisciplinary approach in terms of their management [1,18].

In patients with choanal atresia, the atretic plate may be bony (90

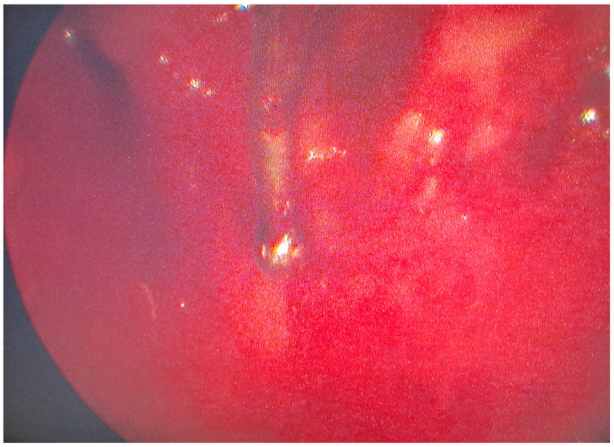

Fig. 1. Endoscopic appearance of the left atretic choana (metallic probe showing the atretic site).

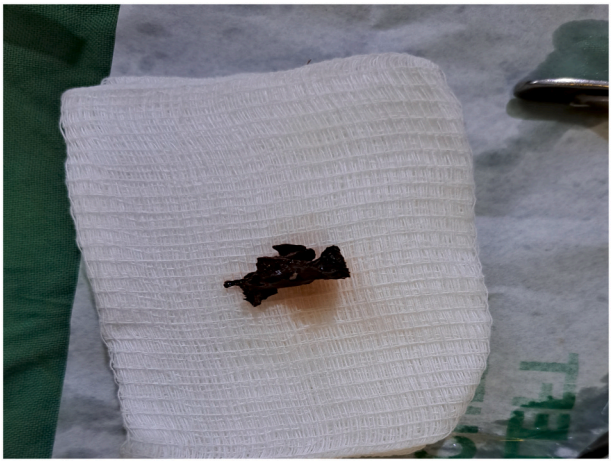

Fig. 3. Rhinolith post removal from the left nasal cavity.

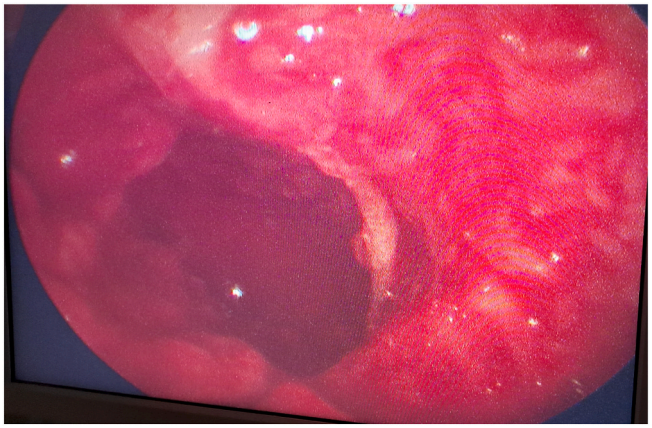

Fig. 4. Endoscopic appearance of the patent choana post release of the atretic plate.

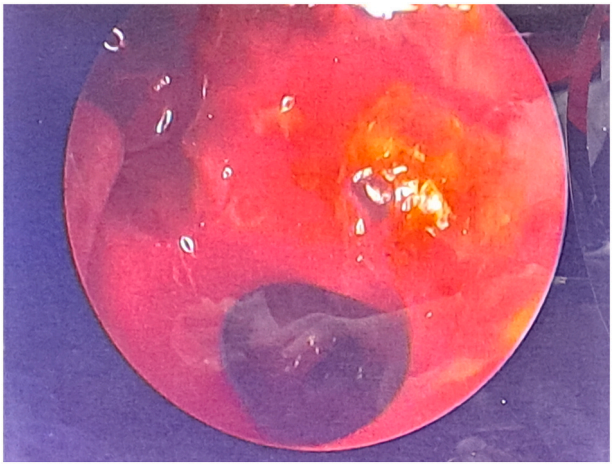

Fig. 5. Endoscopic appearance of the patent choana after three-months of follow up.

%), membranous (5 %), or mixed (5 %) [19,20]. Computed tomography (CT) scan is recommended to establish the diagnosis and also as a guide during surgical perforation of the atretic choana though this may be challenging in resource limited setting [2]. The atretic choanal plate in the case we are hereby reporting was purely bony in nature. Regarding sex predilection of choanal atresia, female preponderance has been reported with male to female ratio being 1:2 [19,21]. The case described in this case report is a 15-year old boy thus not conforming to the known epidemiology of choanal atresia. Incidence of choanal atresia has been reported to be 1 in 5000 to 1 in 8000 live births [2,22].

Most cases of unilateral choanal atresia may go undiagnosed until old age especially if there is no high index of suspicion in patients with recurrent hospital visits due to persistent unilateral nasal obstruction. There are various bed-side tests that are of help to establish the diagnosis of choanal atresia during infancy such as use of methylene blue dye, nasal catheter, use of mirrors by observing fog on the mirror and use of a piece of thread alternately in both nostrils where one observe flipping action in case the tested nostril is patent. Both anterior and posterior rhinoscopy are mandatory to avoid missing such patients since if left undiagnosed till later ages it tends to impair the quality of ones' life through frequent hospital visits [2].

There are various surgical techniques in management of patients with choanal atresia such as transnasal resection or transpalatal resection. Restenosis that required revision surgery was reported in 18 % of patients who underwent transnasal resection and on the other hand possible complications of transpalatal resection include palatal flap necrosis or fistula [2,23].

There are controversies existing over the use of stenting as well as use of fibroblast inhibitors (mitomycin-c). Preventing postoperative cicatrization and stenosis are the commonest reasons for stenting. Materials used for stenting are modified endotracheal tube, silastic stents and Teflon stents. Stents are regularly instilled with saline and suctioned to prevent crusting and blockage [23]. Traditionally the inserted stents are left in-situ for 6–8 weeks for re-epithelization of the neochoana so as to prevent stenosis [24]. Stents are associated with various risks such as patients' discomfort, septal or columellar necrosis, nasal and paranasal infection and intranasal synechiae. Due to morbidities associated with stenting, several studies argued postoperative stenting to be not compulsory [25,26]. Our case was managed by endoscopic resection of the atretic plate without stenting and no stenosis was observed at 3-months upon follow up.

On the other hand, nasal foreign bodies tend to evoke a chronic inflammatory response with deposition of mineral salts mainly calcium and magnesium that increases gradually with time and may be lodged for years [13,27]. This appears to correlate with what was found in our case report where the rhinolith was retained for 2 years. Symptomatology is usually minor by the time of entry of a foreign body into the nose and in most cases patients may forget the event. Thereafter there is a variable latent period during when the nasal foreign body develops and enlarges [11].

Typical clinical features of the rhinolith are unilateral nasal obstruction and a foul-smelling nasal discharge where the discharge is often purulent and fetid and may be blood stained. Other features include epistaxis, nasal or facial swelling, anosmia, epiphora and headache [11–13]. Our case had a similar symptomatology characterized by unilateral nasal obstruction and foul smelling nasal discharge for two years with occasional episodes of epistaxis though went undiagnosed for such duration due to remoteness of where the patient was residing and with limited access to otorhinolaryngologists.

Regarding treatment of rhinoliths, they are removed anteriorly with the help of topical local anaesthesia for pain control in most of the cases and this is upon rhinoscopy that is the main stay in establishing such diagnosis [11]. In our case report the foreign body was removed under general anaesthesia in operating room since it co-existed with unilateral choanal atresia and on the other hand it was not easily visualized probably due to surrounding granulation tissues having enveloped the foreign body in the nasal cavity. Generally, simple removal of a rhinolith with the help of topical local anaesthesia remains to be the treatment of choice though endoscopic removal is also suitable if the equipment permit [11,28].

4. Conclusion

A high index of suspicion by clinicians is necessary in establishing the diagnosis of unilateral choanal atresia otherwise it may be missed until adulthood. Transnasal endoscopic resection of the atretic plate remains to be the treatment of choice due to favorable outcome compared to transpalatal approach and in our case transnasal endoscopic release of the atretic choanal plate without stenting has shown excellent outcome. Any child presenting with unilateral foul smelling nasal discharge must be considered to have a nasal foreign body until proven otherwise and similarly, clinicians must have a high index of suspicion so as to establish the diagnosis of a rhinolith based on symptomatology and a history of foreign body introduction into the nose.

Consent

Written informed consent was obtained from the patients' biological mother for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Ethical approval

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CRediT authorship contribution statement

ZSA-Conceptualization, writing original draft of the manuscript
AAK-Conceptualization and reviewing the prepared original draft of the manuscript.

Conflicts of interest

The authors report no conflict of interest.

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