A rare incidental encounter of unilateral choanal atresia during adenotonsillectomy in a 5-year-old child: A case report from Tanzania

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ABSTRACT

Unilateral choanal atresia refers to a congenital anomaly whereby a child is born with unilateral imperforate posterior nare. In most cases, the diagnosis is established several years after birth. The establishment of the diagnosis requires a high index of suspicion by clinicians. We report an unusual case of a 5-year-old girl who was diagnosed intraoperatively to have unilateral choanal atresia (right) during elective adenotonsillectomy after the failure of introducing a nasal catheter into the oropharynx and on digital palpation of the choanae intraorally. Endoscopic perforation of the membranous atretic choanal plate was done in the same sitting with adenotonsillectomy. The objective of this case report was thus to report an unusual case which went undiagnosed for 5 years and was diagnosed intraoperatively during elective adenotonsillectomy and also to review the few available pieces of the literature. To the best of our knowledge, this is so far the second reported case worldwide of unilateral choanal atresia incidentally diagnosed during adenotonsillectomy.

Key words: *Adenotonsillectomy, Choanal atresia, Tanzania, Unilateral*

Unilateral choanal atresia is an uncommon anomaly in which a baby is born with imperforate unilateral posterior nare. Such condition is due to the failure of a breakdown of the oronasal membrane at 38-weeks of the gestational age. It can be unilateral or bilateral and when it is unilateral, the right nostril is affected more than the left nostril at a ratio of 2:1 [1,2]. When congenital atresia of the choanae is unilateral, infants may go undiagnosed until unilateral rhinorrhea and nasal obstruction necessitate seeking medical care [2-5].

Unlike unilateral choanal atresia which may go unestablished for a couple of years after birth, bilateral choanal atresia presents with respiratory distress immediately after birth; therefore, it is an otorhinolaryngological emergency [4,6]. The reason, as to why children with bilateral choanal atresia presented with respiratory distress after birth, is because they are obligate nasal breathers up to 6 weeks of life due to the nature of their larynx where it appears to have a high position in the neck at the second to third cervical vertebrae, thus infants with such anomaly are diagnosed in the early postnatal period in nearly all cases [2,3,5].

The objective of this case report is to report an unusual case of a 5-year-old girl who was diagnosed intraoperatively to have unilateral choanal atresia (right) during elective adenotonsillectomy after the failure of introducing a nasal catheter into the oropharynx and on digital palpation of the choanae intraorally. Endoscopic perforation of the membranous atretic choanal plate was done in the same sitting with adenotonsillectomy and no stent was put *in situ* and on follow-up for 6 weeks the choana was patent.

CASE REPORT

A 5-year old girl presented to the otorhinolaryngology clinic at our hospital in Dar es Salaam, Tanzania, with a history of snoring and mouth breathing for about 6 months and an accompanied history of recurrent throat pain.

On physical examination during admission, she had good nutritional status. Vital signs were; blood pressure was 100/65 mmHg in the right arm supine position, respiratory rate was 14 breaths/min, pulse rate being 70 beats/min, and body temperature was 36.9°C.

On imaging, where X-ray of the nasopharynx lateral view was taken, maximum narrowing of the nasopharyngeal air column was found and this was consistent with obstructive adenoid hypertrophy. The final diagnosis was thus established to be obstructive adenotonsillitis (Fig. 1).

She underwent elective adenotonsillectomy as scheduled and intraoperatively, unilateral choanal atresia was found after the failure to introduce the nasal catheter into the oropharynx and on digital palpation of the choana, it was found to be imperforate and predominantly membranous in nature (Fig. 2). Nasal endoscopy

was done and the choana was noted to be imperforate. Surgical perforation of the atretic choana, widening of the choana, and removal of the posterior part of vomer without stenting were done endoscopically and on follow-up for 6 weeks, the choana was found to be patent (Fig. 3). To obtain the images during follow-up at the clinic, nasal endoscopy was done after the application of topical xylocaine spray. Long-term follow-up was not possible since the child migrated to a very far region away from where our clinic was located.

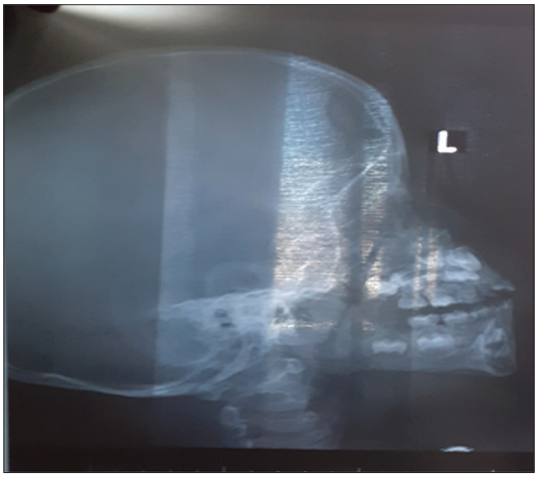

Figure 1: Pre-operative X-ray of the nasopharynx lateral view showing obstructive adenoid

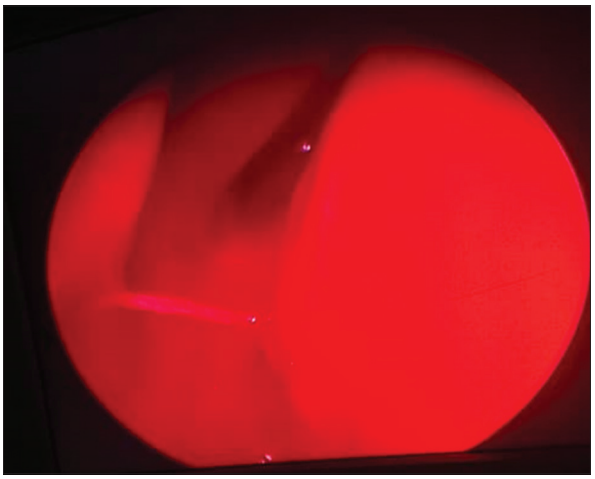

Figure 2: Endoscopic appearance of the atretic choana intraoperatively prior endoscopic atretic plate release

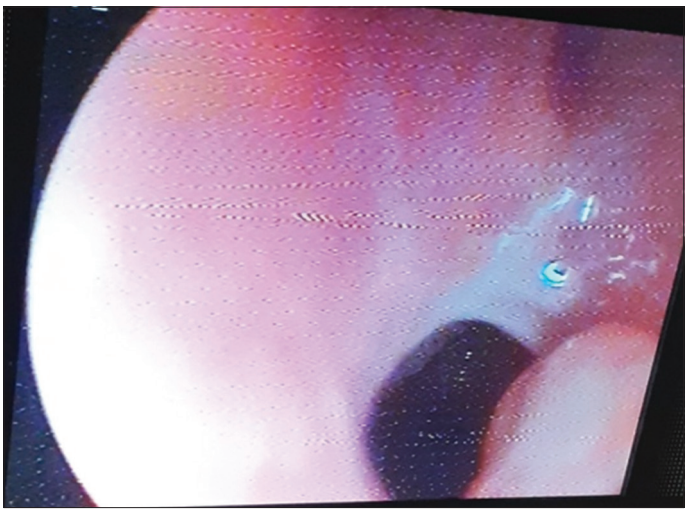

Figure 3: Endoscopic appearance of the choana after 6 weeks of follow-up post-endoscopic atretic plate release

DISCUSSION

Unilateral choanal atresia is not as common as bilateral choanal atresia and <1/2 of patients with choanal atresia have other associated anomalies such as coloboma of the eye, heart defects, retardation of growth and/or development, genital and/or urinary defects, and ear anomalies (CHARGE) syndrome [2,5,7]. Management of patients with choanal atresia and presenting with other associated anomalies such as CHARGE syndrome poses challenges to otorhinolaryngologists, thus requires a multidisciplinary approach to execute the treatment.

The atretic plate responsible for imperforate posterior nares may be bony (90%), membranous (5%), or mixed (5%), and computed tomography (CT) scan is recommended to establish the diagnosis and also as a guide during surgical perforation of the atretic choanae [2,3,8-13]. In terms of sex predilection, female predominance has been reported in the occurrence of choanal atresia with a male to female ratio being 1:2. The case described in this case report is a female child thus conforming to the known epidemiology of choanal atresia [2,8,14].

Unilateral choanal atresia is much more common than bilateral atresia [1,2,4-7,9,12-15]. Both anomalies may be associated with other anomalies such as congenital heart disease, external ear malformation, colobomata of eyelid or iris, gonadal hypoplasia, mental retardation, and cleft palate [2,5,9,11]. The incidence of choanal atresia is reported to be 1 in 5000 to 1 in 8000 live births [2,5]. Unilateral atresia usually presents at the age of 4–6 years with unilateral nasal discharge [4]. Our patient was diagnosed at the age of 5 years incidentally during adenotonsillectomy and thus such age lies within the age range at which the majority of the cases of unilateral choanal atresia are diagnosed. Unilateral atresia is surgically treated at the age of 4–6 years [4]. The patient reported in this case report was operated at the age of 5 years following incidental encounter during adenotonsillectomy thus choanal atresia release and adenotonsillectomy were done in the same sitting.

The diagnosis of choanal atresia is based on the clinical findings coupled by nasal endoscopy and other traditional methods such as the use of a piece of thread and alternately putting close to each nostril and observe the flipping movement of such thread, introducing a nasal catheter into each nostril and visualize it at the oropharynx if has patent choanae, introducing methylene blue dye into each nostril and observe its presence at the oropharynx and finally using a mirror alternately where it is placed close to one nostril after the other while observing for the presence of fog on the mirror in presence of patent choanae. Moreover, a CT scan can indicate whether the atresia is bony or membranous and this also acts as a guide during surgical perforation of the atretic choanae [2,9].

Choanal atresia may be approached during surgery either transnasally or transpalatally. The transnasal approach is simpler and widely applicable in practice. It can be used for either membranous, mixed, or thin bony atresia but atresia may recur in up to 30% cases. Transpalatal approach requires extensive surgery but gives good access and help in creating a larger choana. Stenting of choanae is necessary after surgery to keep it patent and the different types of stents used are portex endotracheal tubes, Foley's catheters, and Silastic tubes [2,6,8,10,11]. Our patient was treated endoscopically by transnasal perforation of the atretic choana without stenting and on follow-up for 6 weeks, the choana was found to be patent. This appears to be similar to what has been reported in other case reports [10,11].

CONCLUSION

Unilateral choanal atresia usuall present in younger age group or in adulthood with symptoms such as nasal obstruction and nasal discharge. Our case report emphasizes the fact that even though unilateral choanal atresia is a poorly recognized cause of nasal obstruction but otorhinolaryngologists should bear in mind the significance of using nasal catheters bilaterally so as to ensure adequate exposure of the nasopharynx during adenotomy or adenotonsillectomy and also digital palpation of the choanae intraorally during such surgical procedure. Transnasal endoscopic release of the atretic choanal plate without stenting has shown excellent outcome in this case thus may be practicable even in children.

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