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A Case Report on Ectopic Intranasal Lateral Incisor Presenting as Chronic Rhinosinusitis in a 25-Year-Old Woman

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ABSTRACT

Objective: To discuss a rare case of ectopic intranasal eruption of a tooth presenting as chronic rhinosinusitis focusing on the cause of the delayed diagnosis and emphasizing the role of otorhinolaryngologists in the management of this disease entity.

Methods:

Design: Case Report
Setting: Tertiary Government Training Hospital
Patient: One

Results: A 25-year-old, partially edentulous woman initially presented with a four-year history of foul-smelling right nasal discharge accompanied by nasal congestion, nasal pain, and epistaxis. Several general physicians prescribed unrecalled antibiotics and pain medications which provided only temporary relief of the symptoms. Due to the persistence of the foul-smelling discharge which was recalcitrant to medications, culture and sensitivity testing at our institution revealed moderate growth of *pseudomonas aeruginosa* and computed tomography scans of the paranasal sinuses showed a right intranasal tooth, thickening of the inferior turbinate and septal deviation to the right. Following endoscopic removal of the intranasal tooth and septoplasty, there was resolution of symptoms and no complications.

Conclusion: Ectopic intranasal eruption of tooth has not yet been reported in the Philippines. Delays in diagnosing and starting proper management can lead to morbidity and serious complications such as development of antibiotic resistance. A high index of suspicion by an ENT-HNS physician with thorough nasal physical examination could have aided in the early diagnosis and management of the intranasal tooth.

Keywords: *unilateral rhinorrhea; ectopic lateral incisor; aberrant tooth; intranasal tooth; ectopic eruption of tooth; chronic nasal discharge; foul-smelling rhinorrhea*

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Ectopic eruption of tooth is a rare phenomenon with an incidence of 0.1-1% of the population.[1] The most common sites of ectopic eruption are the palate and the maxillary sinus due to their proximity to the alveolus; [2,3] requiring the expertise of an otorhinolaryngologist – head and neck surgeon. Due to the paucity of cases reported in literature, its etiology and pathophysiology remain controversial. To our knowledge, there is no published local case report based on a search of HERDIN Plus, the Western Pacific Region Index Medicus (WPRIM), the Directory of Open Access Journals (DOAJ) and MEDLINE (PubMed and PubMed Central) using the search terms "ectopic eruption of tooth," "foul-smelling rhinorrhea," "aberrant tooth," "intranasal tooth," and "ectopic lateral incisor." This report aims to discuss a rare case of ectopic intranasal eruption of a tooth presenting as chronic rhinosinusitis, focusing on the diagnostic delay, and emphasizing the role of otorhinolaryngologists in the management of this disease entity.

CASE REPORT

A 25-year-old partially edentulous woman was seen at our institution due to foul-smelling right nasal discharge with nasal congestion, epistaxis, and nasal pain for four years. Nine years prior to consult, the patient had extraction of carious teeth—maxillary right lateral incisor, bilateral central incisors, bilateral 1st premolars and left 2nd premolar. *(Figure 1)* Prior to this, she claimed to have had complete dentition, and was asymptomatic.

Four years prior to consult, she had foul-smelling right nasal discharge with nasal congestion, epistaxis and nasal pain which caused a lot of social discomfort. During the span of four years, she was repeatedly treated for bacterial rhinosinusitis with different broad-spectrum antibiotics by four different general practitioners, but the foul-smelling nasal discharge did not resolve.

The patient then consulted at our outpatient department. On nasal endoscopy, there was a whitish, hard, immobile mass surrounded by hyperemic mucosa and greenish crusts and discharge on the right nasal cavity floor. *(Figure 2)* The mass abutted the right inferior turbinate. Right-sided septal deviation was also noted. The rest of the physical examination findings were unremarkable. She had no history of any congenital craniofacial development anomaly such as cleft palate and denied sustaining any facial trauma or foreign body impaction in her nasal cavity. Due to the persistence of the foul-smelling discharge which was recalcitrant to medications, culture and sensitivity testing was done which revealed moderate growth of *Pseudomonas aeruginosa*. A computed tomography scan of the paranasal sinuses revealed a right intranasal tooth and thickening of the inferior turbinate with septal deviation to the right. *(Figure 3)* The patient underwent endoscopic extraction of the intranasal tooth and septoplasty. Intraoperatively, the right nasal floor mucosa was incised to reveal the entirety of the tooth. The tooth was noted to abut on the inferior edge of the right inferior turbinate. Grossly, the extracted specimen resembled a right lateral incisor in crown morphology and cuspal architecture. *(Figure 4)* Histopathologic findings were also consistent with that of a tooth. *(Figure 5)* There was no oronasal fistula seen on inspection of the palate, and no immediate post-operative complications.

On follow-up one week after surgery, the nasal discharge and congestion had resolved, and our patient remained asymptomatic on later consults. *(Figure 6)*

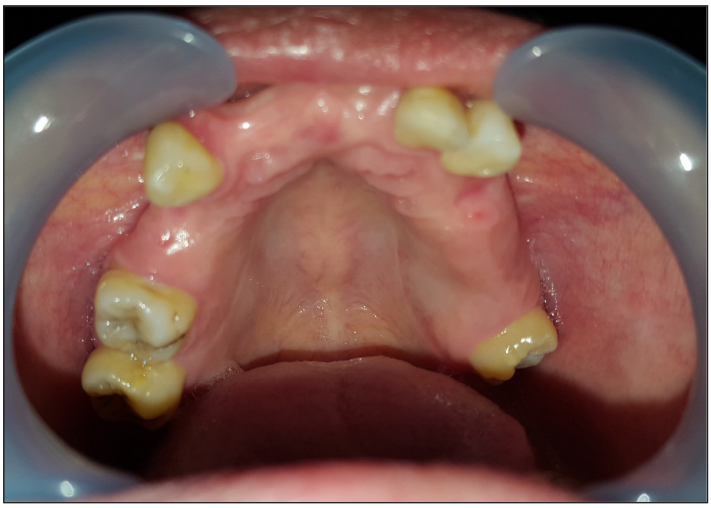

Figure 1. Patient's maxillary dentition at the time of consultation

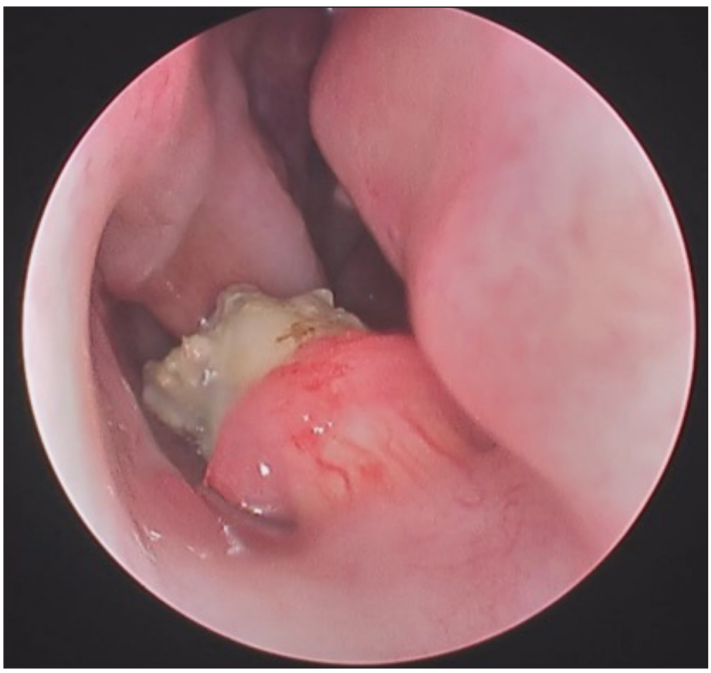

Figure 2. Nasal endoscopy showing a whitish, hard, immobile lesion on a hyperemic base in the right nasal cavity floor

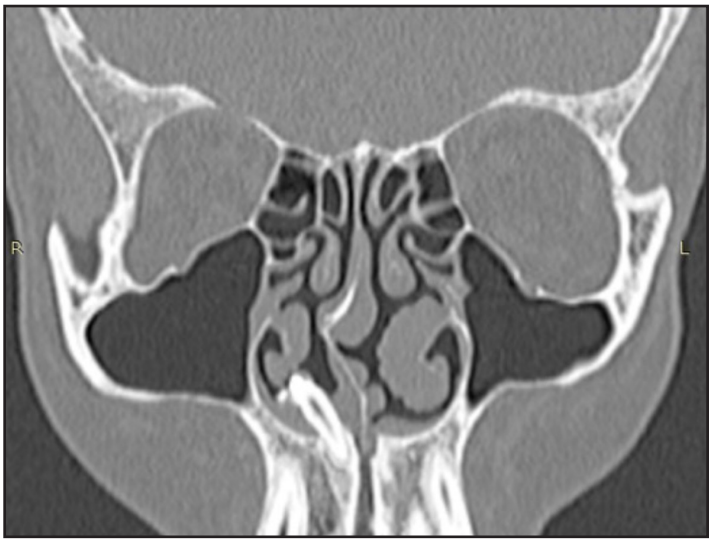

Figure 3. Plain CT scan of the paranasal sinuses, coronal view, revealing a hyperdense lesion on the right nasal cavity floor abutting on the right inferior turbinate with right-sided septal deviation noted

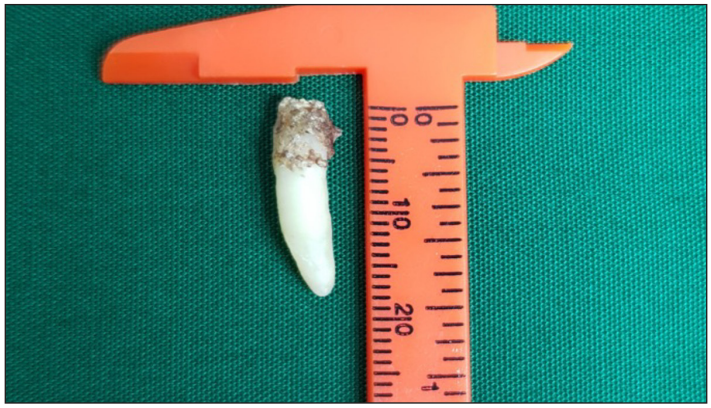

Figure 4. Specimen retrieved following endoscopic extraction. The crown morphology and cuspal architecture are macroscopically those of a lateral incisor

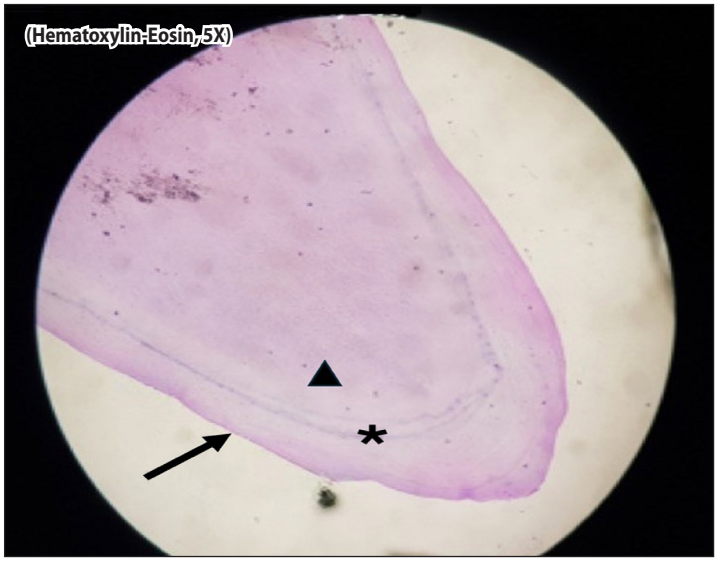

Figure 5. Histopathologic slide, hematoxylin-eosin, scanner view (5X) showing dentin (arrow), enamel (asterisk), and pulp (triangle), consistent with a tooth

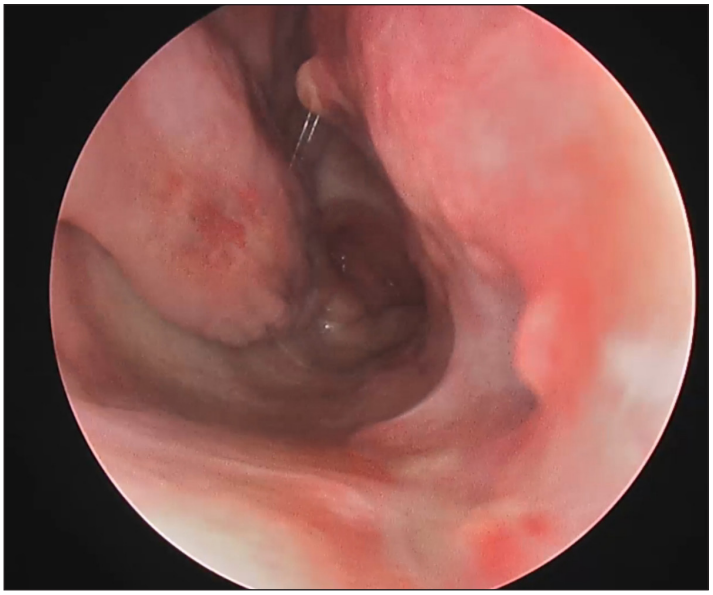

Figure 6. One-week post-operative nasal endoscopy revealing non-hyperemic right nasal cavity mucosa with correction of septal deviation and no nasal discharge

DISCUSSION

Our patient presented with chronic foul-smelling nasal discharge for four years. Diseases that may present similarly include chronic rhinosinusitis, rhinolith, fungal rhinosinusitis, infections like tuberculosis and syphilis, and other benign or malignant tumors of the nasal cavity.[1] Since on physical examination, the tooth was covered partially with nasal mucosa on the nasal floor with greenish crusts and discharge, a neoplasm can also be part of the differential diagnosis. Because of the varied etiologies of chronic nasal discharge, there is a tendency to disregard less common, but equally important differential diagnosis. This was the cause of delay in diagnosis and injudicious use of antibiotics in our patient.

The cause of ectopic tooth growth is still not well-understood. Some authors proposed theories of its pathogenesis: genetic predisposition, developmental abnormalities including cleft palate, rhinogenic or odontogenic infection, displacement due to trauma or cysts, persistent deciduous teeth, or exceptionally dense bone.[4, 5] None of these were present in our patient and not likely the cause of the ectopic intranasal eruption of tooth. The more plausible explanation could be crowding of dentition or the presence of supernumerary tooth since our patient previously had complete dentition but underwent exodontia due to dental caries. Although supernumerary teeth are said to be common in the maxilla and palate,[5] their eruption in the nasal cavity without passing through the maxillary antrum is rare. In our case, etiology is still unclear because we depended on the recollection of the patient about her allegedly complete dentition prior to exodontia. In terms of clinical

manifestations, reported cases of ectopic intranasal tooth eruption had similar signs and symptoms as our patient such as unilateral nasal obstruction, serous or purulent rhinorrhea, nasal pain and epistaxis.[4,6,7] Other manifestations include septal abscess and oronasal fistulas[4,6,7] while others still are asymptomatic and only detected incidentally on imaging.[8,9] It is noticeable that in our patient, the duration from appearance of symptoms to diagnosis took four years while being exposed to multiple courses of broad-spectrum antibiotics as opposed to patients from case reports wherein the duration ranged from 2-6 months.[10,11] This is probably due to prompt referral to or consultation with an ENT specialist, compared to our patient who was only able to consult a specialist after four years. The diagnosis was also missed during her dental consults because the signs and symptoms of the patient were primarily nasal in origin. Prompt detection and management must be done to avoid complications such as antibiotic resistance, abscess, and thrombosis in the cavernous sinus due to its location in the danger triangle of the face. In our case, the patient compliantly took several broad-spectrum antibiotics over four years in the hope of getting rid of her primary symptom of foul-smelling nasal discharge. Nasal discharge cultures revealed moderate growth of *pseudomonas aeruginosa*—a gram-negative aerobic bacilli which is a known opportunistic pathogen. Usually, colonization by such has been implicated in chronic rhinosinusitis in immunocompromised patients, those who already underwent surgery, or those who have been repeatedly treated with antibiotics.[12] It is possible that this is due to prolonged exposure to broad-spectrum antibiotics, adversely affecting our patient with possible antibiotic resistance.

The diagnosis of an intranasal tooth can be easily made based on clinical and radiologic findings. Since the patient was initially managed for chronic rhinosinusitis (CRS), based on Philippine Clinical Practice Guidelines, nasal endoscopy should have been the first-line confirmatory test in diagnosis.[13] However, in our case, the patient consulted general practitioners who did anterior rhinoscopy only. Imaging modalities such as computed tomography scans are useful in visualizing the location and depth of implantation of the tooth in the bone to aid in its surgical removal. Once identified, its management is quite simple and straightforward—tooth extraction. The most used approach which is considered safe and effective is endoscopic extraction because of best visualization, good illumination, more precise dissection, and improved post-operative course.[6,9] Other approaches that may also be used include the transnasal and transpalatal.[14]

In conclusion, ectopic intranasal eruption of tooth, being a rare disease entity, presents as a diagnostic dilemma when it presents similarly to a wide spectrum of diseases with differing managements. In an adult patient presenting with unilateral foul-smelling rhinorrhea, we must not only consider the more common disease entities such as infections and tumors, rather, a thorough physical examination including nasal endoscopy must be performed for proper visualization and evaluation. Physicians should adhere to clinical practice guidelines and utilize proper referral systems.

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