Inverted Supernumerary Intranasal Teeth as Unusual Indications of Endoscopic Surgery

Remo Accorona¹, (MD); Giovanni Colombo², (MD); Marco Ferrari³, (MD); *Enrico Fazio⁴, (MD); Andrea Bolzoni-Villaret³, (MD)

Abstract
Introduction:
Supernumerary teeth are frequently reported in dental clinical practice; however, eruption in nasal cavities and paranasal sinuses is an extremely rare clinical entity.

Case Report:
We report two cases with a history of recurrent nasal discharge and obstruction. In both cases, clinical and radiological findings confirmed the presence of an inverted supernumerary tooth erupted in the sinonasal cavities (i.e., the right nasal fossa and left maxillary sinus, respectively). We managed the cases with transnasal endoscopic approach. A survey of the English literature identified 69 documented cases with intranasal supernumerary teeth within January 1st, 1886 to December 31st, 2017.

Conclusion:
Inverted supernumerary teeth should be considered among the potential causes of unilateral nasal obstruction and rhinosinusitis and included in differential diagnoses among the fibro-osseous lesions of the sinonasal cavities.

Keywords:
Endoscopic sinonasal surgery, Inverted supernumerary tooth, Rhinosinusitis.

Received date: 10 Mar 2019
Accepted date: 28 Nov 2019

Please cite this article as: Accorona R, Colombo G, Ferrari M, Fazio E, Bolzoni-Villaret A. Inverted Supernumerary Intranasal Teeth as Unusual Indications of Endoscopic Surgery. Iran J Otorhinolaryngol.2020:32(3): 181-186.
Doi:10.22038/ijorl.2019.38918.2284

¹Division of Otorhinolaryngology, “San Maurizio” Hospital, Bolzano, Italy.
²Department of Otorhinolaryngology, Humanitas Clinical and Research Center, Rozzano, Milano, Italy.
³Department of Otorhinolaryngology and Head and Neck Surgery, University of Brescia, Italy.
⁴Department of Otorhinolaryngology, University of Insubria, Varese, Italy.
*Corresponding Author:
Division of Otorhinolaryngology, Department of Biotechnology and Life Sciences
University of Insubria, ASST Sette Laghi, Ospedale di Circolo e Fondazione Macchi, Via Guicciardini 9 Varese, 21100 Italy
E-mail: enricofazio@hotmail.it
Introduction

Although supernumerary teeth are frequently reported in dental clinical practice, eruption in sinonasal cavities is a rare clinical entity, occurring in only 0.1-1% of the general population (1). Clinical presentation includes nasal obstruction, chronic inflammation with persistent discharge and crusting, nasal bleeding, septal abscess, and oral-nasal fistula.

In addition to the sinonasal cavities, other possible sites of eruption are mandibular condyle and coronoid process (1). An accurate clinical and radiological assessment is essential for the establishment of the diagnosis and determination of the surgical approach. We present two cases of symptomatic inverted sinonasal teeth treated with an endoscopic transnasal approach.

Case 1

A 64-year-old man was referred to the Department of Otorhinolaryngology-Head and Neck Surgery of the University of Brescia, Italy, with a long-standing history of recurrent right nasal obstruction and discharge unresponsive to systemic therapy with antibiotic and corticosteroid. No other relevant clinical data were reported.

Noncontrast-enhanced computed tomography (CT) showed the presence of a calcified mass in contact with the floor of the right nasal fossa, approximately 1.3 × 0.5 cm in size (Fig. 1). The mass was described as between the inferior turbinate and nasal septum surrounded by inflammatory tissue. The examination with a 0° rigid endoscope identified a calcific element originating from the floor of the right nasal fossa, approximately 1.3 × 0.5 cm in size (Fig. 1). The mass was described as between the inferior turbinate and nasal septum surrounded by inflammatory tissue. The examination with a 0° rigid endoscope identified a calcific element originating from the floor of the right nasal fossa, surrounded by granulation tissue (Fig. 1). Differential diagnoses included the observation of a rhinolith and an inverted supernumerary tooth.

Case 2

A 28-year-old lady was referred to the Department of Otorhinolaryngology of the Humanitas Clinical and Research Center with a clinical history of recurrent left nasal obstruction, discharge, cacosmia, and pain in the left maxillary region. The patient was frequently treated with the administration of systemic antibiotics and corticosteroids at other institutions. The patient did not report any clinical history of facial trauma or surgery of cleft lip or palate. Clinical examination with a 30° rigid endoscope identified polypoid tissue and purulent discharge coming from the left middle meatus. Noncontrast-enhanced CT showed the presence of an inverted tooth erupted in the left maxillary sinus (Fig. 2) concomitant with diffuse rhinosinusitis. The patient underwent a functional endoscopic endonasal surgical procedure under general anesthesia after the preparation with systemic antibiotics and corticosteroids. The tooth was removed via a wide middle antrostomy (Fig. 2). The dissection was difficult due to the presence of calcific material and chronic inflammatory tissue surrounding the tooth.
Inverted Supernumerary Intranasal Teeth

**Fig 2:** a) Sagittal CT scan showed supernumerary tooth erupted inside the left maxillary sinus, surrounded by inflammatory tissue. b) Intraoperative nasal endoscopic view with 45° rigid scope showed the supernumerary dental element (*) on the floor of the left maxillary sinus through wide middle antrostomy. c) Disengagement of the supernumerary tooth from the floor of the left maxillary sinus after dissection of its insertion. d) Extraction of the tooth through the left nostril.

The macroscopic analysis showed a cuboidal dental element, and the diagnosis of the inverted supernumerary tooth was confirmed by histologic examination. The patient was discharged the day after the surgery. She reported complete relief of the symptoms during postoperative care and showed complete healing of the surgical field 2 months after the surgery.

**Discussion**

Supernumerary teeth are described as dental elements in excess of the normal formula. The prevalence of supernumerary teeth in the Caucasian population is within the range of 1-3% (2). If we only consider the inverted supernumerary teeth erupted in sinonasal cavities, the occurrence is reported within 0.1-1% of the general population (1,3,4).

More than 90% of supernumerary teeth occur between maxillary central incisors, and in that situation, they are called mesiodens (2,5). However, according to the literature, a wide variety of supernumerary dental elements, including canine, upper and lower second premolar, and impacted third molars (6-9), were reported. Out of these supernumerary dental elements, only 25% of the cases are inverted and erupted (10,11). Macroscopically, an inverted supernumerary tooth has the crown pointing upward; however, the root apex points toward the alveolar crest.

The first case reported in the present study was an inverted mesiodens. The second patient presented a rarer situation; according to the site, the tooth was an inverted supernumerary molar. The etiology remains unclear, especially in the absence of a clinical history of surgery, trauma, Gardner’s syndrome, or anatomic malformations, such as cleft lip and palate. Three theories have been postulated about this developmental glitch. The first and earliest theory of phylogenic reversion by Von proposed that supernumerary teeth could be the phylogenetic appearances of our extinct ancestors since they possessed three maxillary incisors; however (12), this theory has currently been discarded because it does not justify the presence of nonmesiodens supernumerary elements (2).

The second theory suggests that there could be a split in the tooth bud to create two different teeth (13). The third and the most widely accepted is the hyperactivity theory which postulates that supernumerary teeth can occur due to the hyperactivity of the dental lamina resulting in an additional tooth bud (2,3). Accordingly, a superior supernumerary tooth should grow into the floor of the sinonasal cavity and eventually erupt (14).

Few authors have speculated that supernumerary teeth might originate from a third tooth bed arising from the dental lamina near the permanent teeth (15,16). However, they are usually detected as a single element in the ectopic position, and based on this reasoning, most authors do not confirm the previous theory (14).

A survey of the English literature allowed the identification of 2,717 articles with the search term “supernumerary tooth” in PubMed (i.e., National Library of Medicine) and EMBASE (i.e., Ovid). A further selection of the search term “inverted supernumerary tooth” reduced the search to 59 papers. The final selection included 69 documented cases of an inverted intranasal tooth, 71 cases with the consideration of the 2 present cases, within January 1st, 1886 to December 31st, 2017.

Table 1 tabulates a summary of the results. Considering the second half of the 20th century, There have been reported 71 cases, including our series; only 4 cases dated back to before 1958.
Table 1: Included cases (n=71) of inverted supernumerary intranasal tooth according to year of publication and number of teeth per patient

| First author                  | Year | Number of cases |
|-------------------------------|------|----------------|
| Marshall (17)                 | 1886 | 1              |
| Long (26)                     | 1924 | 1              |
| Abercrombie (27)              | 1925 | 1              |
| Endicott (28)                 | 1934 | 1              |
| Rao (29)                      | 1958 | 1              |
| Quinn (21)                    | 1959 | 1              |
| Bahn (30)                     | 1966 | 1              |
| Hiranandani (24)              | 1968 | 1              |
| Chopra (31)                   | 1969 | 1              |
| Kohli (25)                    | 1970 | 1              |
| Martinson (32)                | 1972 | 12             |
| Savundranayagan (33)          | 1972 | 1              |
| Arora (34)                    | 1973 | 1              |
| Sood (35)                     | 1975 | 2              |
| Hong (36)                     | 1976 | 1              |
| Sivrastava (37)               | 1977 | 1              |
| Thawley (15)                  | 1979 | 1              |
| Smith (18)                    | 1981 | 2              |
| Johnson (38)                  | 1981 | 1              |
| Dayal (39)                    | 1981 | 1              |
| Wood (40)                     | 1984 | 1              |
| Spencer (41)                  | 1985 | 1              |
| King (42)                     | 1987 | 1              |
| Murty (14)                    | 1988 | 1              |
| Ogisi (22)                    | 1988 | 1              |
| Carver (43)                   | 1990 | 1              |
| Pracy (44)                    | 1992 | 1              |
| Yeung (19)                    | 1996 | 1              |
| Nastri (45)                   | 1996 | 1              |
| Lee (20)                      | 2001 | 13             |
| Kim (46)                      | 2003 | 1              |
| Kuroda (7)                    | 2003 | 1              |
| Lin (47)                      | 2004 | 3              |
| Sokolov (48)                  | 2004 | 2              |
| Lee (49)                      | 2006 | 1              |
| Kirmeier (8)                  | 2009 | 1              |
| Sanei-Moghaddam (1)           | 2009 | 1              |
| Iwai (16)                     | 2012 | 1              |
| Noleto (9)                    | 2013 | 1              |
| Ogane (50)                    | 2017 | 1              |
| Koçak (23)                    | 2017 | 1              |
| Personal data                 |      | 2              |
| **Total: 71**                 |      |                |

According to Kuroda et al. (7), Albins reported the first case in 1754, and other cases were identified within the end of the 19th and beginning of the 20th century (7). Marshall in 1886 firstly described the typical symptoms (i.e., nasal obstruction, fetid rhinorrhea, and strong headache) in a patient who blew a tooth autonomously out from the nostril (17). Smith et al. reported two cases and identified other 27 well-documented cases (18). In 1996, Yeung and Lee reviewed the literature and added 12 cases to the previous report (19). Lee reported his series of 13 intranasal teeth and underlined the advantages of endoscopic surgical management (20). A more recent review of the literature by Kirmeier et al. documented 25 cases with inverted supernumerary teeth in 23 patients within 1959 to 2008; nevertheless, the observations of Lee are not reported by Kirmeier et al. (8,20). Most authors reported a
Inverted Supernumerary Intranasal Teeth

single intranasal tooth per patient, as the cases of the present study, with a high prevalence of a supernumerary mesiodens. Conversely, the cases with multiple elements, to the best of our knowledge, are anecdotic (9, 16, 21-23).

In normal dental clinical practice, a supernumerary tooth can be diagnosed as a cause of the misalignment of a tooth, dilacerations of a permanent tooth, and cyst formation, such as a dentigerous cyst. However, in the rare occurrence of an eruption in the sinonasal cavities, the prevailing symptoms often lead the patient to refer to an ear, nose, and throat department. The most common complain are nasal discharge, unilateral nasal obstruction, nasal bleeding, and facial pain (8, 9, 16, 17, 23-25).

Clinically, both the cases of the present study showed unilateral nasal obstruction and persistent fetid rhinorrhea. Rare complications, including the abscess of the nasal septum, deformity of the nasal pyramid, oral-nasal fistula, and nasal septal perforation, were also reported (8, 9). The differential diagnoses include rhinoliths, exostoses, fungal infection, radiopaque foreign bodies, and tumors of bone and cartilage (8, 9, 16, 23-25), and CT scan confirmed the diagnosis.

With the improvement of endoscopic surgery and refinement of functional techniques, the nasal pathway has become the natural way of a tooth extraction with prevalent sinonasal growth. In endoscopic surgery, it is possible to have a clear visualization of the insertion, avoidance of injury to surrounding mucosa, and precise dissection with the preservation of surrounding structures (20). Finally, it also allows treating concomitant pathological conditions, such as chronic rhinosinusitis and septal deviation. The possible complications of endoscopic surgery are related to the extent of the procedure. In the management of an intranasal tooth, theoretically, the most aggressive procedure should be a wide antrostomy for an intramaxillary tooth. Except for the risk of postoperative bleeding, the main discomfort for the patient should be the necessity of nasal medications to avoid crusting and bad healing of the surgical bed.

Conclusion

Inverted supernumerary teeth are rare clinical entities; however, they should be considered among the potential causes of unilateral rhinosinusitis and included in the differential diagnoses among the fibro-osseous lesions of the sinonasal cavities. In this regard, the endoscopic approach is nowadays the treatment of choice.

References

1. Sanei-Moghaddam A, Hyde N, Williamson P. Endoscopic removal of supernumerary tooth from the nasal cavity in an adult. Br J Oral Maxillofac Surg 2009;47:484-5.
2. Jangid K, Varghese SS, Jayakumar ND. Ectopic supernumerary tooth at the anterior nasal spine. A Developmental glitch. J Clin Diagn Res 2015;9:1-2.
3. Primosch RE. Anterior supernumerary teeth assessment and surgical intervention in children. Pediat Dent 1981;3:204-14.
4. Mason C, Azam N, Holt RD, Rule DC. A retrospective study of unerupted maxillary incisors associated with supernumerary teeth. Br J Oral Maxillofac Surg 2000;38:6.
5. Rajab LD, Hamdan MA. Supernumerary teeth: Review of the literature and survey of 152 cases. Int J Paediatr Dent 2002;12:244-54.
6. Sykaras SN. Mesiodens in primary and permanent dentitions. Oral Surg 1975;39:870-4.
7. Kuroda H; Tsutsumi K, Tomisawa H, Koizuka I. A case of inverted tooth in the nasal cavity. Auris Nasus Larynx 2003;127-9.
8. Kirmeyer R, Truschnegg A, Payer M, Maljyk J, Daghighi S, Jakse N. The supernumerary nasal tooth. Int J Oral Maxillofac Surg 2009;38:1219-22.
9. Noleto JW, Prado R, Rocha JF, Da Costa MA, Barbosa CU, Toscano MD. Intranasal inverted tooth: A rare cause of a persistent rhinosinusitis. Indian J Dent Res 2013;24:762-4.
10. Mc Kibben DR, Brearley LJ. Radiographic determination of the prevalence of selected dental anomalies in children. J Dent Child 1971;38:390-8.
11. Kumar A, Namdev R, Bakshi L, Dutta S. Supernumerary teeth: Report of four unusual cases. Contemp Clin Dent 2012;3:71-7.
12. Von AT. Anterior maxillary supernumerary teeth: a clinical and radiographic study. Aust Dent J 1992; 37:189-95.
13. Russell KA, Magdalena AF. Mesiodens-Diagnosis and management of a common supernumerary tooth. J Can Dent Assoc 2003; 69:362-66.
14. Muryt PS, Hazarika P, Hebbar GK. Supernumerary nasal teeth. Ear Nose Throat J 1988; 67:128-9.
15. Thawley SE, LaFerriere KA. Supernumerary nasal tooth. Laryngoscope 1977;87:1770-73.
16. Iwai T, Aoki N, Yamashita Y, Omura S, Matsui Y, Maegawa J. et al. Supernumerary intranasal teeth. J Oral Maxillofac Surg 2012;70:1030-4.
17. Marshall JS. A superior wisdom tooth discharged from the nasal passages with remarks. JAMA 1886;7:516-17.
18. Smith RA, Gordon NC, DeLuchi SF. Intranasal Teeth. Report of two cases and review of the literature. Oral Surg Oral Med Oral Pathol 1979;47:120-2.
19. Yeung KH, Lee KH. Intranasal tooth in a patient with a cleft lip and alveolus. Cleft Palate Craniofac J 1996;33:157-9.
20. Lee FP. Endoscopic extraction of an Intranasal tooth: a review of 13 cases. Laryngoscope 2001;111:1027-31.
21. Quinn JH, Lewis M. Bilateral inverted supernumerary central incisors penetrating nasal cavity: report of case. J Oral Surg Anesth Hosp Dent Serv 1959;17:61-2.
22. Ogisi FO, Odita JC. Ectopic nasal dentition associated with squamous cell carcinoma of palate in a 12-year-old boy. Br J Oral Maxillofac Surg 1988;26:58-61.
23. Koçak HE, Özdamar K, Bilgi B. A rare cause of intranasal mass: bilateral ectopic nasal teeth. Iran J Otolarhinolaryngol 2017;29:287.
24. Hiranandani LH, Melgiri RD. Supernumerary tooth in the floor of the nose. J Laryngol Otol 1970;82:845-8.
25. Kohli GS, Verma PL. Ectopic supernumerary tooth in the nasal cavity. J Laryngol Otol 1970;84:537-8.
26. Long CJ. Supernumerary tooth in the nose. Dental Cosmos 1924;66:360.
27. Abercrombie PH. Eruption of a canine tooth into the nasal fossa attended by rhinitis caseosa. J Laryngol Otol 1925;40:586-9.
28. Endicott CL. A case of a supernumerary incisor erupting into the nose. Br Dent J 1934;56:385-6.
29. Rao AB. Aberrant canine tooth in the nose. Laryngol 1958;67:370-1.
30. Bahn SL. Supernumerary tooth in the right nostril associated with a median palatine cyst. Report of a case. Oral Surg Oral Med Oral Pathol 1966;21:409-13.
31. Chopra SS, Joshi MR. Mesiodens erupted in the nasal cavity. Report of a case. Oral Surg Oral Med Oral Pathol 1969;28:856-8.
32. Martinson FD, Cockshott WP. Ectopic nasal dentition. Clin Radiol 1972;23:451-4.
33. Savudranayagan A. A migratory third molar erupting into lower border of the orbit causing blindness in the left eye. Aust Dent J 1972;17:418-20.
34. Arora MM, Grewal BS, Coomar HS. Dentigerous cyst from supernumerary tooth in the nasal cavity. J Indian Dent Assoc 1973;45:85-6.
35. Sood VP, Kakar PK. Intranasal tooth. Eye Ear Nose Throat Mon 1975;54:25-9.
36. Hong CY. Ectopic nasal tooth. Med J Malaysia 1976;30:239-40.
37. Srivastava RP, Pradhan AC, Yadav VNS. Tooth in nasal cavity: a case report of cleft lip and palate. J Indian Dent Assoc 1977;49:145-6.
38. Johnson AP. A case of an intranasal canine tooth. J Laryngol Otol 1981;95:1277-9.
39. Dayal PK, Dewan SK, Bihani VK, Dave CJ. Eruption of a tooth into nasal cavity due to osteomyelitis. J Laryngol Otol 1981;95:509-12.
40. Wood TE, Webster E. Intranasal eruption of a permanent central incisor: a case report. Natl Dent Assoc J 1984;41:29-30.
41. Spencer MG, Couldery MG. Nasal tooth. J Laryngol Otol 1985;99:1147-50.
42. King NM, Lee AMP. An intranasal tooth in a patient with cleft lip and palate: report of case. J Am Dent Assoc 1987;114:475-8.
43. Carver DD, Peterson S, Owens T. Intranasal teeth: a case report. Oral Surg Oral Med Oral Pathol 1990;70:804-5.
44. Pracy JP, Williams HO, Montgomery PQ. Nasal teeth. J Laryngol Otol 1992;106:366-7.
45. Nastrî AL, Smith AC. The nasal tooth. Case report. Aust Dent J 1996;41:176-7.
46. Kim DH, Kim JM, Chae SW, Hwang SJ, Lee SH, Lee HM. Endoscopic removal of an intranasal ectopic tooth. Int J Pediatr Otorhinolaryngol 2003;67:79-81.
47. Lin IH, Hwang CF, Su CY, Kao YF, Peng JP. Intranasal tooth: report of three cases. Chang Gung Med J 2004;27:385-9.
48. Sokolov M, Jecker P, Roth Y. Nasal teeth associated with rhinosinusitis. Rhinology 2004;42:167-70.
49. Lee JH. A nasal tooth associated with septal perforation: A rare occurrence. Eur Arch Otorhinolaryngol 2006;263:1055-6.
50. Ogane S, Watanabe A, Takano N, Shibahara T. Case of inverted supernumerary tooth in nasal cavity. Bull Tokyo Dent Coll 2017;58:255-8.