Patent nasopalatine ducts: an update of the literature and a series of new cases

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Abstract

Purpose The objective of this review is to present an update and summary of clinical findings of cases with a patent nasopalatine duct (NPD) reported in the literature from 1881 to 2016.

Methods Previous articles and reviews about patent NPDs were studied and copies of all original publications were obtained for data verification. Furthermore, a literature search was conducted. In addition, the study sample was complemented with four cases recently seen in our institution.

Results Ten out of 67 published cases were to be excluded for this analysis due to misinterpretation or misreporting in previous articles. Overall, 57 cases with NPD patency could be analyzed. Males outnumbered females in a ratio of 2:1. The mean age (when this information was available) was 34.1 ± 17.6 years (range 6–69 years). NPDs were located bilaterally (60%), unilaterally (20%) or centrally (20%). Complete or partial patency was reported in 73.9 and 26.1%, respectively. 74.1% of patients presented a variety of clinical signs and symptoms. The ability of the patient to produce a squeaky or whistling sound was the most frequent clinical finding (23.8%).

Conclusions Caution must be exercised when reading review articles about NPD patency since wrong data have been copied in several subsequent publications. Since

epidemiological data are missing with regard to patent NPDs, age and gender predilections are not warranted. Bilateral occurrence and full patency were prevailing features in the evaluated case reports of patent NPDs.

Keywords Patent nasopalatine duct · Nasopalatine canal · Vomeronasal organ · Case report

Introduction

Patency of the nasopalatine duct (NPD) in humans is a rarely described anomaly of the anterior maxilla. Niels Stensen (1638–1686), a Danish anatomist and physician, was the first to provide a thorough description of the NPD in 1683 [24]. The NPD is often mixed up with the nasopalatine (incisive) canal (NPC). The NPC is a prominent structure within the anterior maxilla and represents a passageway originating bilaterally from the anterior nasal floor, subsequently fusing to form a single canal terminating at the incisive foramen in the anterior palate. The NPC conveys the nasopalatine arteries and nerves from the lower and anterior portion of the nasal septum to the palatal mucosa posterior to the maxillary incisors. The NPC further contains veins and seromucous glands [28, 49].

During the fetal period, an epithelialized duct also runs through the NPC as an additional connection from the oral cavity to the so-called vomeronasal organ (VNO) [23, 39]. The VNO (also called Jacobson's organ) is located bilaterally at the bottom of the anterior nasal septum. The presence of the VNO in humans is still under debate, but has been documented histologically in cadavers [48, 56]. There is general agreement that the NPD degenerates and/or obliterates completely before birth [23, 39]. As a consequence, the canalicular link between the oral and nasal cavities is usually

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no longer present after birth. However, in some individuals, the NPD does not close and remains partially or completely open [54]—hence the term “patent NPD”.

The first description of patency of the NPD in humans is attributed to Leboucq in 1881 [25]. However, he described patent NPDs in two “foetus à terme”, meaning in late fetus stage, and not in living humans. Since then, many case reports have described the features of the NPD, but epidemiological data are missing. Moreira Falci et al. [33] published in 2013 a review article including case reports of patent NPDs from 1881 to 2010. The objective of the present article was (1) to verify and complement the data from that and previous reviews, (2) to add case reports published from 2010 to 2016, and (3) to present four new cases seen in our department within the last 6 years.

Material and method

Previous review articles about NPD were used as baseline information [11, 22, 33, 42]. An additional literature search (without language restriction) of publications indexed in the MEDLINE database was conducted in December 2016 using the following search terms:

- (patent OR palatal opening OR persistent) AND (nasopalatine duct OR nasopalatine canal OR incisive canal)
- (oronasal OR nasopalatine) AND fistula
- perforate nasopalatine communication.

Exclusion criteria were:

- reports of patent NPDs in unborn human specimens
- reports of anatomical structures misinterpreted as patent NPDs
- reports failed to describe method of NPD evaluation.

Furthermore, the information of four new cases examined in our department from 2011 to 2016 was included in the present analysis.

The following data was collected from the case reports: age and gender of patient, location (Fig. 1) and patency (Fig. 2) of NPD, clinical signs and/or symptoms, and any performed treatment. The data was tabulated (Table 1) and analyzed.

New cases

Case 1 (Fig. 3)

A 64-year-old medical practitioner was referred by his private dentist for extraction and subsequent implant

Fig. 1 Illustration of the typical locations of patent NPDs relative to the incisive papilla: **a** bilateral occurrence, **b** unilateral location, and **c** central location (posterior to the incisive papilla)

replacement of the maxillary right central incisor. This tooth presented with multiple findings: sinus tract on labial aspect, insufficient quality of root-canal filling, and root malformation with lateral radiolucency on mesial aspect. Further inspection revealed a unilateral opening on the left side of the incisive papilla. Three-dimensional radiography (CBCT) confirmed complete patency of the NPD with the inserted guttapercha point reaching the left nasal floor. The right central incisor was subsequently extracted and replaced with a dental implant but no specific treatment was necessary with regard to the NPD.

Case 2 (Fig. 4)

A 33-year-old female dentist in our postgraduate program attended one of the anatomy lectures by the first author of this article. She could produce a squeaky sound at will,

Fig. 2 Illustration of the patency of NPDs running through the NPC (anterolateral view of premaxilla): complete patency on left side, partial patency (cul-de-sac) on right side. *IF* incisive foramen, *NPC* nasopalatine canal, *cNPD* completely patent nasopalatine duct, *pNPD* partially patent nasopalatine duct, *NPN/A* nasopalatine nerve and artery, *VNO* vomeronasal organ

and furthermore, she was aware of passage of air or fluids from/to the anterior palate but did not realize that she had a patent NPD until the aforementioned event. The patient presented healthy maxillary incisors but bilateral openings were noticed at the incisive papilla. On the right side a guttapercha point could be placed into the opening to its full length whereas on the left side a guttapercha point could only be inserted partially. The intranasal inspection with an endoscope confirmed the through-and-through patency of the right NPD. The patient requested no further therapy.

Case 3 (Fig. 5)

The dental hygienist of a 69-year-old male noticed a mucosal opening at the left side of the incisive papilla. The patient presented no symptoms. All maxillary incisors reacted in a delayed but positive way to CO₂-snow testing and periodontal probing depths were ≤ 3 mm. A guttapercha point could be inserted into the opening to a depth of 9 mm. An incompletely patent, unilateral NPD was diagnosed and no further action was undertaken.

Case 4 (Fig. 6)

The private dentist of a 39-year-old patient had detected two small openings bilaterally to the incisive papilla. The patient, though without any symptoms, was referred to our department for the clarification of the findings. Guttapercha points

could be partially inserted without causing any bleeding or pain. The adjacent central and lateral incisors were clinically and radiographically inconspicuous. The patient and the referring dentist were informed about the diagnosis of partially patent NPDs. No further treatment was necessary.

Results

Since 1881, 45 publications (including this one) have presented a total of 67 case reports with NPD patency (Table 1). The number of case reports per article ranged from one to five. Meticulous analysis of the articles resulted in the exclusion of ten cases. One and the same case have been reported in two papers [4, 13]; hence the data were only included once. Five cases were excluded since the case description was incomplete, not verifiable or not related to NPDs (Table 2). We also excluded the two cases reported by Leboucq [25] since they were described in fetus. Furthermore, thorough reading of the article by Rawengel [41] resulted in the inclusion of only three cases, not five cases as mistakenly reported in previous reviews. As a consequence, 57 cases from 37 articles were finally analyzed (Tables 3, 4).

The age distribution ranged from newborn to 69 years. For 50 cases with precise age information (mean age 34.1 ± 17.6 years, median age 31 years), age decade groups are shown in Fig. 7. The gender of the patients was given in 54 cases with a male-to-female ratio of 2:1 (36 males, 18 females). The locations of the NPDs with regard to the incisive papilla (if reported) were bilateral (60%), unilateral (20%) or central (20%), respectively. Considering bilateral occurrence, a total of 88 NPDs were available for classification of their patency. Patency of the NPDs was complete in 73.9% and partial in 26.1%. Multiple clinical signs and symptoms were described in 74.1% of the cases if that information was available. The most frequent clinical finding was the ability of the patient to produce a squeaky or whistling sound (23.8%). Treatment, if provided at all, was mostly surgical excision and closure of the patent NPD. However, in the majority of cases (59.5%), no treatment was necessary or required by the patient.

Discussion

The present paper is an update of clinical case reports about patent NPDs. Four new cases are presented resulting in a total of 57 patients reported in the literature. The strength of this review is that all cases published in the literature were verified by obtaining copies of the original articles. Our literature analysis also demonstrates that misinformation was often copied by subsequent review articles without verification of previously published data.

Table 1 Case reports of patent NPDs published from 1881 to 2016

Author(s)	N of cases	Age	Gender	Location of NPD	Patency of NPD	Main clinical signs and symptoms	Treatment
◆ Leboucq [25]	2	Foetus	NA	Bilateral	2 × complete	NA	NA
		Foetus	NA	Unilateral (right)	1 × complete	NA	NA
◆ Potiquet [38]	2	NA	NA	NA	NA	NA	NA
		NA	NA	NA	NA	NA	NA
Rawengel [41]	5	Newborn	NA	Bilateral	2 × complete	NA	NA
		Newborn	NA	Bilateral	2 × complete	NA	NA
		◆ Newborn	NA	Bilateral	2 × complete	NA	NA
		◆ Newborn	NA	Bilateral	2 × complete	NA	NA
		Adult	NA	Unilateral (left)	1 × complete	NA	NA
◆ Meyer [32]	1	NA	NA	NA	NA	NA	NA
Fry [17]	2	62	Male	Central	1 × complete	Discomfort in anterior palate	Surgical excision
		62	Male	Central	1 × complete	Salty taste, discharge, periodical swelling	Surgical excision
◆ Grohs [20]	1	NA	NA	NA	1 × complete	NA	NA
Roper-Hall [44]	1	18	Male	Unilateral (right)	1 × complete	Chirping sound	NA
Osman Hill and Darlow [36]	1	Adult	Male	Bilateral	2 × complete	Squeaky noise, passage of air/fluid from nose to mouth	None
Rogers [43]	1	46	Male	Central	1 × partial	None	NA
MacGregor [30]	1	57	Male	Central	1 × complete	Discharge from right nostril on eating	Surgical closure
Buchner and Mlinek [8]	1	45	Male	Bilateral	2 × partial	None	NA
Goebel [18]	1	22	Female	Bilateral	2 × complete	Vacuum air from nose into mouth at will	None
Broome and Seymour [7]	3	23	Male	Bilateral	1 × complete 1 × partial	None	NA
		64	Male	Central	1 × complete	Suppuration, bad taste	NA
		31	Female	Bilateral	2 × complete	Swelling	NA
◆ Rapp and Winter [40]	1	4	Female	Central	NA	NA	NA
Allard et al. [3]	1	28	Male	Bilateral	2 × complete	Squeaky noise	NA
Farman et al. [16]	1	8	Male	Bilateral	1 × complete 1 × partial	None	None
Moskow [34]	1	20	Male	Bilateral	2 × complete	Drainage on palate, foul taste	NA
Signy and Rule [47]	2	6	Male	Central	1 × partial	Debris collection	NA
		Adult	Male	Unilateral (right)	1 × partial	None	NA
Lehman and Poulsen [27]	2	22	Male	Bilateral	2 × complete	Passage of air from nose, squeaky noise	None
		38	Female	Bilateral	2 × complete	Passage of air from nose, squeaky noise	None
Blackburn [6]	1	19	Female	Bilateral	2 × complete	Passage of air from nose to mouth	None
Cros et al. [12]	1	26	Male	Bilateral	2 × complete	Nasal secretion into mouth	None
Götzfried [19]	1	44	Female	Bilateral	2 × complete	Drainage on palate, foul taste	Instilling an alcoholic amino-acid prolamin solution
Hattori [21]	1	12	Male	Unilateral	1 × partial	Pain, secretion	NA

Table 1 (continued)

Author(s)	N of cases	Age	Gender	Location of NPD	Patency of NPD	Main clinical signs and symptoms	Treatment
Eppley and Delfino [15]	1	9	Male	Bilateral	2 × complete	Occasional passage of liquids into nose when drinking; whistling deformity	Excision of NPDs and surgical closure with rotational flap
Chapple and Ord [11]	4	29	Male	Bilateral	2 × complete	Patient could express air from nose into mouth making a “squeaking noise”	None
		31	Male	Central	1 × partial	Entrapment of food	Surgical closure with rotational flap
		45	Male	Bilateral	1 × complete 1 × partial	Discomfort and discharge	Surgical closure with palatal flap
		13	Female	Bilateral	1 × complete 1 × partial	Squeaky noise	Surgical closure planned
Shimura et al. [46]	1	20	Male	Central	1 × complete	Occasional passage of liquid into nose	Excision of NPD and surgical closure with rotational flap
Beckett and Gilmour [5]	1	39	Female	Bilateral	2 × partial	None	None
Chandler and Gray [10]	1	31	Male	Bilateral	1 × complete 1 × partial	Bad taste	None
Winslow et al. [57]	1	11	Male	Central	1 × complete	Passage of water/food to nose	Surgical closure
Moss et al. [35]	1	42	Male	Central	1 × partial	None	None
Knecht et al. [24]	1	24	Male	Central	1 × complete	Passage of air from mouth into nose, whistling sound	None
Lundner and Warunek [29]	1	13	Male	Bilateral	2 × complete	Patient could express water through right nostril; whistling noise	None
Catros et al. [9]	1	55	Male	Bilateral	1 × complete 1 × partial	Drainage of viscous substance causing a foul taste	Removal of entire NPD and surgical closure with palatal flap
Valstar and van den Akker [53]	1	34	Male	Unilateral (right)	1 × complete	Patient could produce squeaky sound	None
Rodrigues et al. [42]	1	15	Female	Unilateral (left)	1 × complete	Chirping noise during speech and swallowing	Excision of incisive papilla and content of the NPC with surgical flap closure
von Arx and Bornstein [54]	2	29	Female	Bilateral	2 × complete	None	None
		24	Male	Bilateral	2 × complete	Patient could express air from nose into mouth	None
Edwards et al. [14]	2	Early 20ies	Female	Unilateral (right)	1 × complete	Whistling sound, able to express fluid from mouth into nasal cavity	None
		Mid 50ies	Male	Unilateral (left)	1 × complete	None	None
Ebinger et al. [13]	1	Q65	Female	Unilateral (left)	1 × partial	Pain, foul taste, secretion	Chlorhexidine 0.1%, Ledermix®
Pithon [37]	1	13	Male	Bilateral	2 × complete	None	None
Schulze [45]	1	43	Female	Bilateral	2 × partial	Chronic inflammation	NA

Table 1 (continued)

Author(s)	N of cases	Age	Gender	Location of NPD	Patency of NPD	Main clinical signs and symptoms	Treatment
Lee et al. [26]	2	51	Female	NA	NA	Pain, inflammation	Surgical excision
		49	Female	NA	NA	Pain, inflammation, swelling	Surgical excision
Moreira Falci et al. [33]	1	15	Female	Bilateral	2 × complete	None	None
Bassetti et al. [4]	3	53	Female	Bilateral	2 × complete	Pain	Chlorhexidine 0.1%, Ledermix®
		41	Male	Bilateral	2 × complete	Squeaky sound	None
		♂♦ 65	Female	Unilateral (left)	1 × partial	Pain, foul taste, secretion	Chlorhexidine 0.1%, Ledermix®
Werder et al. [55]	1	53	Female	Bilateral	2 × complete	Pain	Excision of incisive papilla and content of the NPC with surgical flap closure
New cases (this paper)	4	64	Male	Unilateral (left)	1 × complete	None	None
		33	Female	Bilateral	1 × complete 1 × partial	Passage of air/fluid via NPD, squeaky sound	None
		69	Male	Unilateral (left)	1 × partial	None	None
		39	Male	Bilateral	2 × partial	None	None

NA information not available

♦Cases excluded in this review (but cases were included in previous reviews)

♂Same patient (reported in two different publications)

No epidemiological information is currently available with regard to the prevalence of NPD patency. Detection of persistent openings in the anterior palate are most frequently incidental findings since the area around the incisive papilla is generally not inspected during routine dental examination. Furthermore, mucosal openings may be hidden from direct visual view by overlying lateral ledges of the incisive papilla [14]. Abnormal symptoms such as the passage of fluids or air from the nose to the mouth and vice versa, or the ability of the patient to produce squeaky or whistling sounds, may prompt the patient to consult a dentist. Pain or signs of infection are rarely associated with a patent NPD and, therefore, are not the primary cause for seeking dental or medical counseling.

Some case reports have described the occurrence of patent NPDs after active orthodontic expansion of the premaxilla [15, 29, 37]. The authors speculated that widening of the palatal suture in the anterior maxilla had opened presumably closed NPDs. Others noted that expansion of the hard palate could cause these openings to move in a lateral direction relative to the stationary incisive papilla, making the openings visible at the lateral margins of the incisive papilla [14]. The narrow dimensions of the preexisting NPDs might have precluded clinical symptoms prior to orthodontic therapy.

A male to female ratio of 2:1 was found in the present review. Since epidemiological data do not exist, it is not

possible to warrant a gender predilection of NPD patency. One may speculate that males exhibit more lateral maxillary growth (widening of the palate) than females with subsequent opening of latent NPDs. With regard to the reported ages of the case presentations, no further conclusions can be drawn except that age does not seem to be a key factor for the detection of a patent NPD.

Since the NPD is related to the NPC, mucosal openings of the NPD are located immediately lateral or posterior to the incisive papilla. To our knowledge, a location of the NPD orifice anterior to the incisive papilla has never been reported. We may speculate that cases with a single mucosal opening (unilateral or central location of patent NPD) may have a second NPD that is not open to the oral cavity. Jacob et al. [22] have documented the consistency of nasal openings of NPDs but without patency through to the oral mucosa (cul-de-sac). Maximum depth of guttapercha point insertion reached 8 mm [22].

Techniques of evaluation of NPD patency include a variety of methods: the insertion of gutta-percha points with subsequent radiography is the most frequently applied technique. The use of wires or metallic probes has been criticized since these may forcefully perforate the tissues rather than glide through the (usually curved) NPD. Others have used dyes or contrast agents for the assessment of

Fig. 3 64-year-old male presenting with a sinus tract (**a**, arrow) and mesial radiolucency (**b**, arrow) associated with the right maxillary central incisor. The palatal view (**c**) shows the inserted gutta-percha point (asterisk) into the mucosal opening left to the incisive

papilla. The sagittal (**d**) and axial (**e**) CBCT images demonstrate that the inserted gutta-percha point (asterisk) runs inside the nasopalatine canal up to the nasal floor

duct patency. Evaluation of the duct patency has also been done with histology using serial sections [26, 41].

Persistence of NPDs is related with a vast variety of clinical signs and symptoms. However, only few patients would seek medical or dental consultation since pain is not a primary symptom of patent NPDs. The most frequently reported clinical signs are directly associated with the communication nature of the patent NPD, i.e. the ability of the patient to produce a whistling sound and the passage of air from the nose to the mouth. Most patients were aware of these signs but did not consider them as problematic, and thus would not consult a dental practitioner. In consideration of the absence or mildness of clinical signs and symptoms, most patients do not ask for (surgical) treatment. Reassurance and counseling are often sufficient once a diagnosis of patent NPD has been established unless the patient is really suffering. In the latter situation, surgical

excision of the content of the NPD with subsequent flap closure has become the standard approach.

In a rare study from an ENT perspective, university community volunteers were endoscopically examined to characterize the nasal opening(s) of the NPD [22]. The latter were detected in 94% of 221 nostrils, and the nasal NPD openings were located on average 19 ± 0.2 mm dorsal to the columella nasi and 2 ± 0.1 mm above the junction of the nasal floor and the septum.

In animals, the NPD may serve to deliver pheromones and social chemo-signals entering the oral cavity to the VNO [22]. The latter authors summarized three general anatomical relations between the VNO and the NPD in mammals: (1) the VNO opens directly into the nasal cavity with the NPD close by (rodents and bats), (2) the VNO opens directly into the NPD (cats), and (3) the VNO opens directly into the oral cavity (cattle). In humans, the existence and function of

Fig. 4 33-year-old female presenting with bilateral openings (**a**, arrows) next to the incisive papilla. Guttapercha points (**b**, asterisk) were inserted into both mucosal openings. The occlusal radiograph (**c**) exhibits the inserted guttapercha points (asterisk)—on the right side to full length and on the left side only partial. The endoscopic

inspection shows the right palatal opening (**d**, arrow) with the guttapercha (**e**, asterisk) point being inserted. The endonasal endoscopy (**f**) exhibits the guttapercha point (asterisk) reaching the right nasal cavity through the patent NPDP

the NPDP and VNO remain unascertained. However, recent cadaver studies including histology and immunohistochemistry confirmed the presence of the VNO in humans [48, 56].

The clinical relevance of the patent NPDP is that most clinicians do not know this anomaly or are not familiar with its clinical and anatomical findings. There is a certain risk that patent NPDPs may be misinterpreted as sinus tracts originating from neighboring teeth with subsequently unnecessary conventional or surgical endodontic (re-) treatment [35]. Nasopalatine duct pathosis can mimic endodontic (apical) lesions, and thus should be considered in a differential diagnosis when the signs and symptoms consist of either a radiolucent area, a palatal swelling, or a palatal exudate in the region of the anterior maxillary teeth [7]. Epithelial remnants of the nasopalatine duct have also been associated with the development of the nasopalatine duct cyst [1, 2, 31]. The latter is a “true” pathology and the cyst requires surgical enucleation [50, 52]. When NPDP

cysts are misinterpreted as periapical lesions, unnecessary endodontic treatments may be initiated [51]. In most NPDP cyst cases, the anterior teeth react positively to pulp sensitivity testing, but correct diagnosis might be difficult when pulp sensitivity testing is negative or when endodontic treatment has already been performed in the incisor region. In this context, Lee et al. [26] published an interesting article including two case reports of patent NPDPs that radiographically appeared as ovoid cystic lesions in the anterior maxilla. However, histological analysis of the enucleated tissue showed no features of cyst formation. In contrast, ducts aligned in a linear fashion and lined with ciliated columnar epithelium were present. In addition, immunohistochemistry demonstrated the proliferating potential of the ductal epithelial cells and their capacity to induce inflammatory reactions. The article emphasizes the risk of histological misinterpretation of patent NPDPs as nasopalatine duct cysts.

Fig. 5 A unilateral opening (a, arrow) left to the incisive papilla was observed in a 69-year-old male. A gutta-percha point (b, asterisk) could be introduced and the corresponding periapical radiograph (c) shows the direction of the inserted gutta-percha point (asterisk)

Fig. 6 A 39-year-old male demonstrated bilateral mucosal openings (a, arrows) at the incisive papilla. Two gutta-percha points (b, asterisk) could be partially inserted. The periapical radiograph (c) exhibits the length and course of the introduced gutta-percha points (asterisk)

Patency of an NPD may also simulate a presumable oronasal communication upon the nose-blow-test after the removal of posterior maxillary teeth [53]. Thus, the presence of a patent NPD is considered a diagnostic pitfall [25]. Some authors have challenged the term “patent nasopalatine duct” since, by definition, a duct is an epithelium-lined tube that conveys fluid or other secretions, and it was proposed

to name this variant “developmental oronasal fistula of the incisive papilla” [14].

In conclusion, the present review provides a comprehensive update of the literature with regard to patency of NPDs in humans. The majority of patients were males. Patent NPDs were commonly located bilateral to the incisive papilla, and patency was mainly complete. The majority of

Table 2 Summary of cases that had been included in previous review articles but were excluded in the present literature update

Author(s)/year of original article	First erroneous interpretation in article by	Reason for exclusion	Comment
Leboucq [25]	Rawengel [41]	Leboucq [25] writes on page 391 in his article: “Parmi vingt-huit fœtus à terme que j’ai observés, j’ai trouvé sur deux le conduit perméable: une fois des deux côtés, une fois à droite seulement”	Thus the cases mentioned in later articles and attributed to Leboucq are not newborns but fetus, and hence cannot be included in a review including only cases after birth
Potiquet [38]	Jacob et al. [22]	The article by Potiquet [38] describes in detail the “Jacobson’s canal” located above the “Jacobson’s organ” within the nasal septum and its exploration by rhinoscopy: “Jacobson’s canal” is not the NPD!	Jacob et al. [22] were the first to include erroneously two cases from the article by Potiquet [38]; subsequent articles have just copied that information
Rawengel [41]	Farman et al. [16]	Table II on page 521 of the original article by Rawengel [41] summarizes the histological findings of ten newborn and four adult cadaveric premaxillae. However, only two newborn specimens had bilateral patent NPDs and one adult specimen had a unilateral patent NPD	The erroneous data have probably been taken from Table I on page 521 of the original article, but those data refer to ducts and not to patients
Meyer [32]	MacGregor [30]	Meyer [32] describes in his paper in detail median anterior maxillary cysts and speculates about their origin: on page 1867 he writes “I have seen only one case of what appeared to be a patent incisive canal in a living adult”	Since the author does not provide any details of this case, it cannot be included in our review
Grohs [20]	Allard et al. [2]	Grohs [20] writes “In one case a continuous ductus naso-palatinus was observed, connecting nasal and oral cavities”. No information is provided on how NPD patency was ascertained	The text is a short abstract, probably from a histologic perspective; the author provides no details on how patency was evaluated
Rapp and Winter [40]	Signy and Rule [47]	Original presentation is a single photo in a textbook with the following caption “Palatal Pit: This 4-year-old girl shows an unusual small midline defect in the mucosa of the hard palate immediately posterior to the incisive papilla”	There is no conclusive information that the “palatal pit” represents the mucosal opening of a patent NPD

Table 3 Analysis of data of cases with patent NPDs ($n=57$)

	<i>N</i>	%	Comments
Gender ($n=54$)			
Male	36	66.7	
Female	18	33.3	
NA	3		
Age ($n=57$)			
Specific age not provided	7	–	2 × newborn, 5 × adult
Specific age provided	50	–	See Fig. 7
Location ($n=55$)			
Bilateral	33	60.0	33 bilateral cases = 66 NPDs
Unilateral	11	20.0	4 × right side 6 × left side 1 × unspecified side
Central	11	20.0	
NA	2		
Patency ($n=88$)			
Complete	65	73.9	
Partial	23	26.1	
NA	2		
Symptoms ($n=54$)			
None	14	25.9	
Symptoms	40	74.1	See Table 4
NA	3		
Treatment ($n=42$)			
None	25	59.5	
Surgery	14	33.3	
Instilling chemicals	3	7.1	2 × chlorhexidine 0.1% and Ledermix® 1 × alcoholic amino-acid prolamin solution
NA	15		

NA data missing in case report(s)

Table 4 Analysis of reported clinical signs and symptoms in 40 cases ($n=63$)

Signs/symptoms	<i>N</i>	%
Whistling/chirping sound, squeaky noise	15	23.8
Passage of air from nose to mouth	9	14.3
Discharge/secretion/drainage	9	14.3
Pain, discomfort	8	12.7
Bad/foul taste	6	9.5
Passage of liquids from mouth to nose	6	9.5
Inflammation	3	4.8
Swelling	3	4.8
Debris/food collection	2	3.2
Salty taste	1	1.6
Suppuration	1	1.6

Multiple signs and symptoms were possible per case

Fig. 7 Age distribution (per age decades) of cases with patent NPDs as reported in the literature ($N=50$)

patients presented with signs and symptoms. The ability of the patient to produce a squeaky or whistling sound was the most frequent finding, but a multitude of signs and symptoms were associated with patent NPDs.

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Compliance with ethical standards

Conflict of interest The authors report that they do not have any conflict of interest.

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