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Surgical approaches for nasal dermal sinus cysts*

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SUMMARY

Nasal midline masses of ectodermal origin include nasal dermoids (ND) and nasal dermal sinus cysts (NDSC). NDSC are characterized by an intracranial-extradural extension, while ND are limited to the nasal dorsum, medial canthus, or glabella without intracranial extension. We report our experience in 11 NDSC patients. The goal of this study is to present the management including surgical technique for NDSC and compare it with the literature. Because a transfacial approach for NDSC with vertical incision caused visible scarring in two out of three patients, we applied a new surgical approach in four patients. This approach consisted of a simple excision and mobilisation of the pit while the proximal part is resected using a coronal transfrontal approach. The relation of the nasal fistula to the nasal bone is essential considering osteotomy. Disruption of the bony cartilaginous junction of the nasal dorsum must be prevented to avoid later growth impairment of the nose. There was no recurrence of NDSC in all 7 operated patients after a mean follow-up of 3.9 years (range 0.5 – 7.2 years).

Key words: surgery, nasal cyst, nasal sinus, nasal dermoid, congenital nasal mass

INTRODUCTION

Developmental nasal midline masses occur in one out of 20.000 to 40.000 live births [1-3]. Failure of embryological separation of neuroectodermal and ectodermal tissues during the development of the nose and anterior skull base is believed to result in two morphologic distinctive lesions [3,4]. Congenital nasal midline masses in children and young adults are classified according to their origin into ectodermal and neuroectodermal forms. Neuroectodermal forms include gliomas, meningoceles and meningoencephaloceles, ectodermal forms include nasal dermoids (ND) and nasal dermal sinus cysts (NDSC). ND are cystic lesions, confined to the nasal dorsum, glabella or medial canthus while NDSC may present with cystic components along a tract that from the nasal dorsum to the foramen coecum [5]. In order to rule out an intracranial extension and to exclude neuroectodermal lesions (meningocele, meningoencephalocele), dedicated, high-resolution neuroimaging with CT and MRI is mandatory [6].

There are currently two different theories that may explain the pathogenesis of ND and NDSC: The so-called "*cranial theory*" suggests that during the development of the frontobasis, dura mater that retreats from the prenasal space may adhere to the overlying nasal skin resulting in a sinus tract. According to the "*superficial theory*", ectoderm trapped between the two medial, fusing nasal processes forms a sinus or cyst.

The diagnosis of ND and NDSC can be made on clinical basis, since most of them present with a nasal pit in the midline of the nasal dorsum, columella or upper lip. Recurrent suppurative infections and cosmetic disfigurement are the main symptoms leading to medical consultation. For surgical planning several factors have to be taken into account: its nasal location, the probable involvement of deeper nasal structures, the potential intracranial extension (in up to 45% [8,9]), the patient's age and the frequency of suppurative inflammations [7]. While dermoids in the nasal dorsum can be removed using open rhinoplasty (transcolumellar) approach [10-12] the correct surgical approach for NDSC remains controversial. An optimal exposure of the fistula or sinus tract is crucial to prevent incomplete resection, which may lead to recurrent infections.

Since surgical treatment for ND is well described in literature, the present study aimed to present and discuss our surgical experience in patients treated for NDSC.

PATIENTS AND METHODS

Patients

Between 1998 and 2006, 19 patients with developmental nasal midline masses have been treated at our hospital. As imaging (CT and MRI) of three patients showed neuroectodermal lesions (2 nasal glioma, 1 meningoencephalocele) they were excluded. The remaining 16 patients with ectodermal lesions

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comprised ND (5 patients) and NDSC (11 patients). They were asked when the swelling was discovered for the first time, if a skin discoloration had occurred, if infection had occurred and if a recurrent discharge from the pit has been observed. All patients underwent dedicated high-resolution neuroimaging including computer tomography (CT) and/or magnetic resonance imaging (MRI). To depict the exact location and -intracranial- extension of the lesion, CT was performed in all patients as the primary investigation. Intracranial involvement was considered when CT showed an enlarged foramen coecum and/or a bifid crista galli. If CT could not definitively rule out intracranial extension, MRI was added.
A nasal dermoid was diagnosed, when CT showed a cystic lesion along the midline of the nasal dorsum, glabella or medial canthus in combination with a normal sized and shaped foramen coecum and crista galli. If the foramen coecum appeared enlarged or a bifid crista galli was seen in combination with a cystic midline lesion NDSC was diagnosed.

In five patients a nasal dermoid on the nasal dorsum or medial canthus was surgically removed using open rhinoplasty approach to expose the nasal dorsum. Eleven patients with a NDSC could be included in the study. Indication for surgery for NDSC was based on clinical signs such as recurrent infections or cosmetically disturbing deformities. Four patients have not undergone surgery as they had only few or no symptoms. Seven patients underwent surgery.

Surgical techniques for NDSC
The first three patients with NDSC in our series were operated using a transfacial approach: after mobilising the pit, the fistula was filled with Methylen-Blue. A Lynch incision was extended to the nasal pit. The fistula was exposed and followed up endocranially in all patients. The colouring of the sinus was not considered as a reliable border between the fistula and the more proximally detected fibrous stalk. Hence the whole sinus tissue connected with the pit was excised. Visible scars after the transfacial approach required corrective plastic surgery in two out of these three patients (Figure 1).

Figure 1. Remaining scars after transfacial approach for NDSC and recurrent infections in the right medial canthus at the age of 2 year and without scar revision.

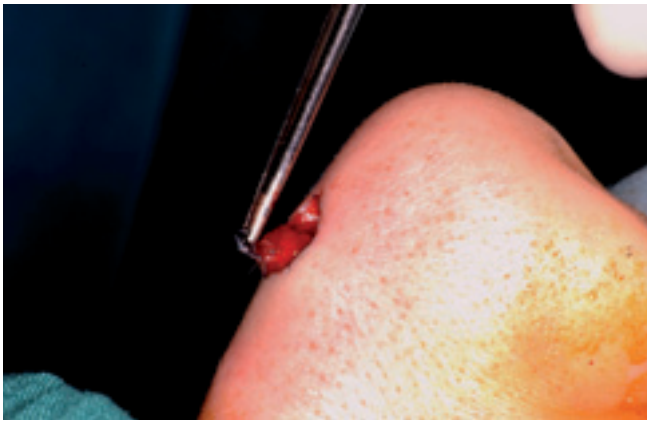

Figure 2. NDSC: Mobilising the nasal pit.

Impressed by the visible scars we changed the technique and operated the remaining four patients using a combined approach: the nasal pit was mobilised by a tiny skin incision and the fistula filled with Methylen-Blue (Figure 2). A coronal approach was used to expose the extension of the fistula up to the bony nasal dorsum. On surgery the sinus could be followed over the nasal bone in two and underneath the nasal bone in two out of three patients as expected on neuroimaging. To expose the sinus properly, osteotomy of the nasal bone was necessary (Figures 3A and 3B) in these two patients.
Imaging of the four not operated patients showed that the sinus could be followed up to the level of or underneath the level of the nasal bone forming a gutter (Figures 4A and 4B). To avoid later growth deficits of the nose, careful attention was paid not to damage the junction between the nasal bone and the upper lateral cartilage. The fistula was explored to its most cranial extension (i.e. foramen coecum) for which osteotomy of the frontal bone became necessary in all four patients. After excision of the sinus the bone was replaced.

RESULTS

All 11 patients with NDSC had contrast enhanced high resolution CT. Seven out of 11 had in addition a contrast enhanced MRI. Diagnosis was confirmed on surgery in all patients. Patient characteristics and clinical signs are listed in Table 1.
Clinical symptoms and findings in patients with a NDSC leading to diagnostic investigation were recurrent infections in 4 (37%), a cosmetically disfiguring nasal mass (Figure 5) in 3 patients and recurrent meningitis in 1 (9%). Four patients have not been operated yet since they presented low morbidity. Two of them were referred by one of the authors (PH) because of a suspicious nasal pit associated with hairs on the nasal dorsum without discharge (Figure 6) while 1 patient presented only recurrent discharge out of the nasal pit on the nasal dorsum.
Surgery was performed in 7 out of 11 patients. Four patients remained asymptomatic and therefore are followed clinically. The initial 3 patients underwent transfacial resection applying a prolonged Lynch incision including the nasal pit. Revision

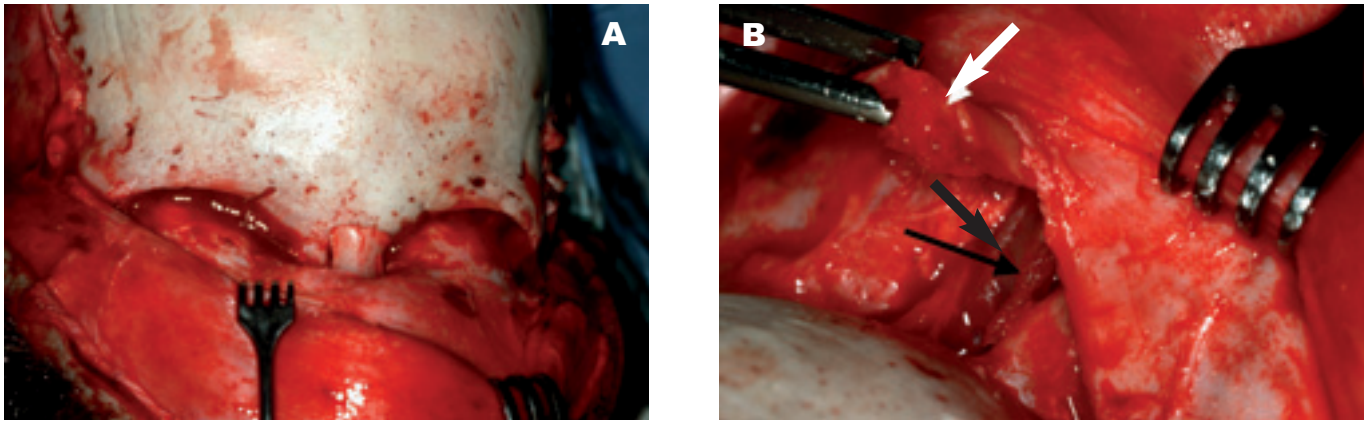

Figure 3. NDSC A: Exposure of the nasal dorsum; B: Osteotomy of the nasal bone (white arrow head) and exposure of the fistula in a gutter (black arrow head).

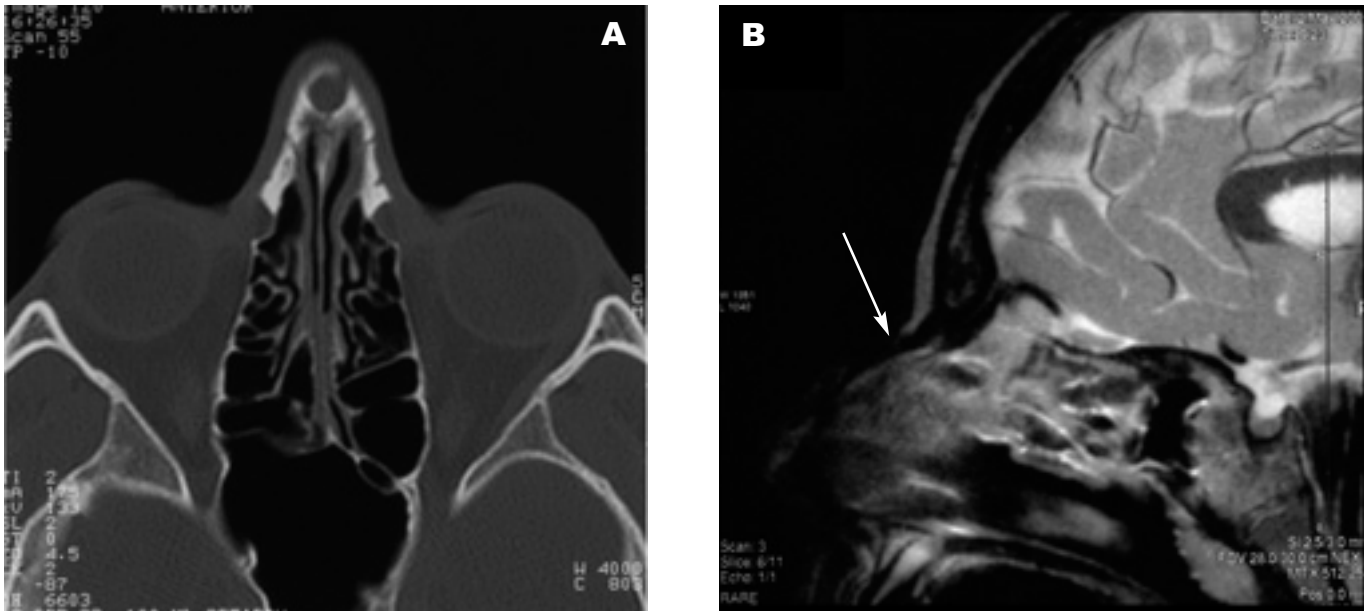

Figure 4. NDSC A: Fistula located within the nasal bone (CT) B: Fistula running underneath nasal bone (white arrow head; MRI).

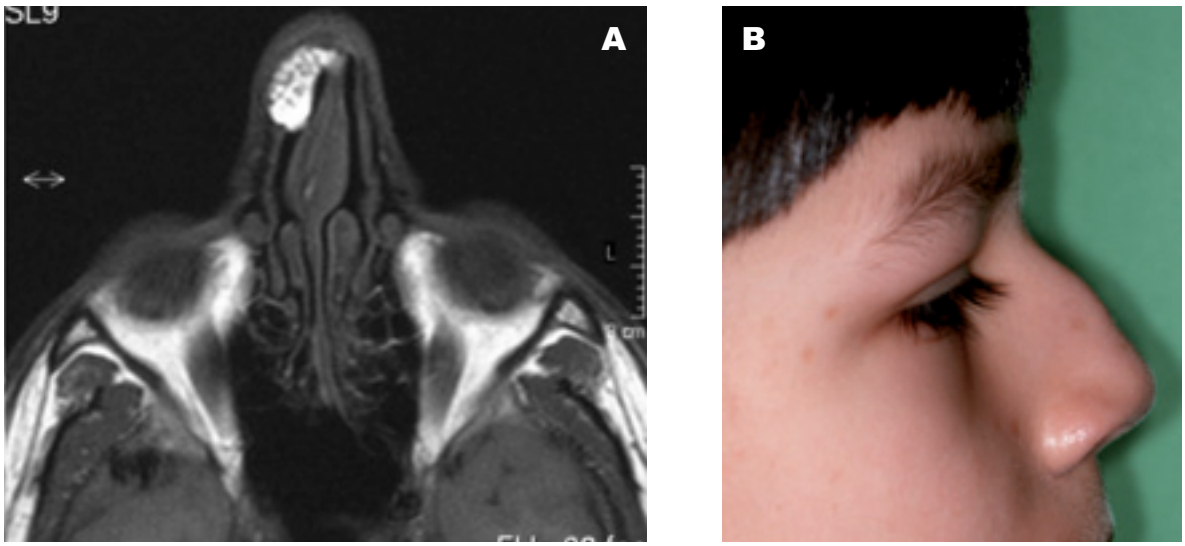

Figure 5. NDSC of the nasal dorsum A: MRI B: clinical appearance with disfiguring hump.

surgery due to visible scar was performed in 2 of them, while the parents of the third child did not consider scar revision necessary. One patient developed a small area of alopecia in the scalp where coronal incision was performed requiring surgical excision, while the other 3 had excellent cosmetic results (Figures 7A and 7B). No blood transfusion became necessary in all operated patients. There was no recurrence of NDSC after a mean follow-up of 3.9 years (range 0.5 - 7.2 years).

DISCUSSION

Although ND's and NDSC's are reported to occur in every 20.000 to 40.000 live births, only few papers discuss the best surgical approach. The most recent reviews and largest series comprised 44, 22 and 42 patients, respectively [9,13,15]. Due to the lack of a proper definition and terminology these studies summarized ND's and NDSC's. While Rahbar et al [15] grouped all ectodermal malformations as one category and named them "nasal dermoids", Bradley [9] named them all NDSC. In contrast to these larger series, ND's were less frequent than NDSC's in our patient population.

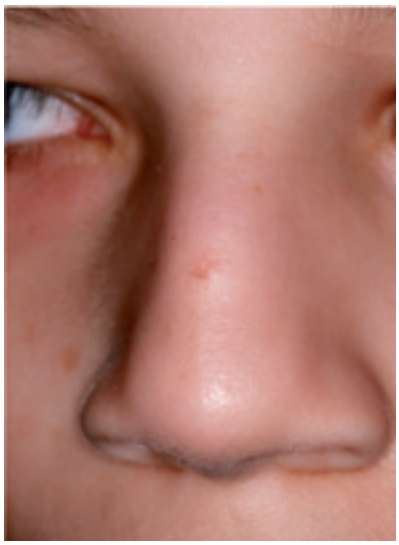

Figure 6. NDSC nasal pit.

Similar to previous case reports [8,15], nasal pit was a pathognomonic sign in all our NDSC patients.

If a nasal midline mass is suggested, high resolution CT and MRI is necessary to define the extent and location of the lesion as well as the possible intracranial-extradural extension. In addition, applying both modalities a classification in neuroectodermal and ectodermal lesion is possible. However, none of the clinical signs can predict the degree of extension. Similar to Pensler et al [16], our series showed that an enlarged foramen coecum and a bifid crista galli indicate intracranial involvement of NDSC. Sessions [5] described in his overview that the most proximal part frequently consists of a fibrous tract or stalk. The histological work-up of the excised fistula in our NDSC patients confirmed this finding. A prospective identification of a fibrous stalk is however impossible on CT or MRI [6]. As a consequence, the surgeon has to be prepared to explore the area of the foramen coecum and crista galli whenever neuroimaging identifies osseous pathology. CT should be the considered to be first choice in imaging, since bony deformities like bifid crista galli and enlarged foramen coecum can be identified easily therein.

Radical resection in NDSC implies an exposure of the proximal end of the fistula. The easiest exposure to access the foramen coecum and crista galli is vertical incision. It still remains a matter of debate whether the transfacial approach leaves significantly visible scaring, which we encountered in our small series. It is speculative whether the growing facial skin might have influenced scar formation despite following the correct skin incisions. As a matter of fact, preoperative recurrent and severe infection of the fistula, like in one of our patient (Figure 1) may contribute to visible scarring. We therefore changed the

Table 1. Patients with NDSC.

			Clinical symptoms		Imaging		Surgery			Follow-up	
sex	**age at DX (years)**	**DX**	**recurrent infections**	**cosmetic disturbance**	**CT/MR**	**at age of (years)**	**approach**	**sequelae**	**revision**	**time (years)**	**recurrence**
M	2.3	NDSC	yes	no	CT/MR	2.5	transfacial	scar	scar revision	7	no
F	8	NDSC	recurrent meningitis	no	CT/MR	8.7	transfacial	scar	scar revision	7.2	no
M	1.2	NDSC	yes	no	CT/MR	1.2	transfacial	scar	no	6	no
M	3.0	NDSC	no	yes	CT/MR	7.1	cornal transfrontal	alopecia	scar revision	2	no
F	13.8	NDSC	no	yes	CT/MR	15.5	coronal transfrontal	no	no	4	no
M	2.1	NDSC	yes	no	CT/MR	2.9	coronal transfrontal	no	no	1.0	no
M	0.8	NDSC	no	no	CT	n.p.					
F	2.8	NDSC	no	no	CT	n.p.					
M	1.9	NDSC	no	no	CT	n.p.					
M	1.1	NDSC	yes	yes	CT/MR	6.7	coronal transfrontlal	no	no	0.5	no
M	8.5	NDSC	no	no	CT	n.p.					

Abbreviations: n.p. = surgery not performed

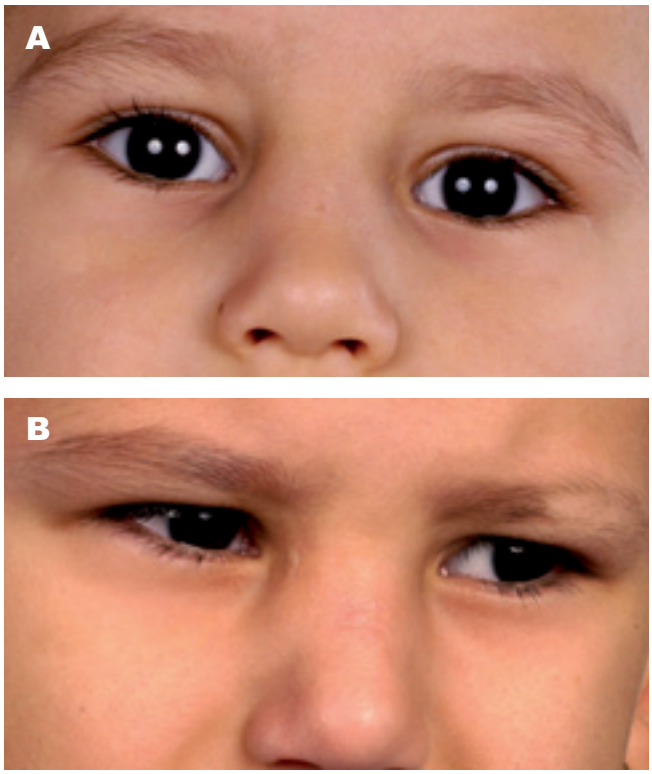

Figure 7. NDSC A: preoperatively B: after coronal subfrontal approach and excision of the pit.

surgical concept exposing the nasal dorsum and foramen coecum using a coronal transfrontal approach similar as proposed by Kellman et al. in a recent study [17]. They recommended a coronal approach with osteotomy of the frontal bone. However, the authors detached the upper lateral cartilage from the nasal bone. In our experience, this is not necessary if the nasal pit is mobilised up to the bony cartilaginous junction. At this point, the fistula either follows at the level of the nasal bone with formation of a gutter or it runs underneath the nasal bone in a bony canal. Performing the osteotomy of the nasal bone cranially and laterally with preservation of the junction to the upper lateral cartilage as shown in Figure 3, the fistula could be pulled through in our experience.

The small number of study patients is one of the shortcomings of our study. In addition, to definitively judge the long-term results the patients in our series should have been followed 10 years at least. We nonetheless would like to emphasise, that the coronal transfrontal approach is feasible even in children, which might contribute to less visible scaring.

CONCLUSIONS

Enlarged foramen coecum and bifid crista galli are important imaging criteria to distinguish ND from NDSC. According to our experience contrast enhanced high resolution CT is sufficient for diagnosis and surgical planning in most of the cases. If a neuroectodermal lesion like meningocele or meningoencephalocele is suspected imaging must be completed by an MRI. Recurrent infections on the nasal dorsum and adjacent skin as well as disfiguring deformities of the nose were the most frequent findings leading to surgery in NDSC. If surgery is attempted in patients with NDSC radical resection of the whole sinus tract is mandatory. Although our series is small we favour the coronal transfrontal approach since patients after transfacial approach had visible scaring postoperatively.

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