International Journal of Health Research and Medico-Legal Practice
Open access full-text article
Available at www.ijhrmlp.org

CASE SERIES

Presentation and management of nasal dermal sinus: a case series

Goswami Rupnayan[1], Goswami JK[2], Sarma Abhijit[3]

[1]Assistant Professor, [2]Professor
(Corresponding author)
Paediatric Surgery, Gauhati Medical College, Guwahati, Assam, India.
***Email**: drjayanta@hotmail.com*
[3]Associate Professor
Department of Surgery

***Received on**: May 19, 2019*
***Editorial approval on**: Jan 25, 2021*
***Plagiarism Checked**: Yes*
***Peer Review**: Double blinded*
***Peer review comments**: Four*
***Editor who approved**:*
Prof. Putul Mahanta

***Background and aims:** Nasal dermal Sinus is a relatively rare congenital condition. We have reviewed our experience in the presentation and management of these cases. **Material and methods**: This is a retrospective study from June 2006 to January 2019. Patients presented with a nasal pit, with or without a hair in it, were selected. They were studied for their various aspects before and after the operation. **Results**: Out of eight patients of our series, one presented with infection, and hence operation was done later. One patient's parents refused this operation at one sitting with his other condition, and therefore seven patients were operated on. Complete excision of dermal sinus by midline vertical incision under general anaesthesia was done. One patient had a recurrence and was reoperated 14 years later. Follow-up was from 1 to 13 years, with a mean of 6 years. **Conclusion**: Nasal dermal Sinus and dermal cyst without fistulae are two different entities. Most of the sinuses of this condition do not bear the intracranial extension. Excision can be done from the nose, and cranial exploration is rarely required.*

***Keywords**: Nasal dermal sinus; sinus with hair; congenital nasal pit; nasal pilonidal sinus.*

***Cite this article**: Goswami Rupnayan, Goswami JK, Sarma Abhijit. Presentation and management of nasal dermal sinus: a case series. Int J Health Res Medico Leg Prae 2021 Jan-June;7(1):89-93. Doi: 10.31741/ijhrmlp.v7.i1.2021.15.*

INTRODUCTION

Nasal dermal sinus in children is relatively a rare disease. A small pit, present since birth in the midline of the nose anywhere from the root of the nasal bridge to the columella base, characterizes it. The opening may have coarse hair coming out from it, and occasionally it discharges white sebum like material. Its incidence is 1:20000 to 1:40000 births.[1] Its Pathogenesis has been explained in various ways. Superficial sequestration or in the development process in the frontonasal region incomplete obliteration of neuroectoderm are the two most accepted theories.[2] Nasal dermal sinus and cyst has been considered as the same spectrum of disease in some papers. Apart from the clinical examination, CT or MRI scan is required to find out size and extension, particularly for the intracranial component, if any.[3] The condition requires early surgical management. Delay in management can cause infection, fistula formation and scarring. These complications cause disfigurement, and then complete excision of the sinus becomes extremely difficult. If there is any intracranial extension of the tract, the infection may cause meningitis or brain abscess.[4]

The cases of nasal dermal sinus encountered by us have been analyzed here.

MATERIAL AND METHODS

This is a retrospective cohort study done in the Paediatric Surgery Department of a tertiary medical Centre. Eight patients with nasal dermal sinus were included in this study. The period was fourteen years, from June 2005 to May 2019. Though dermal cyst and sinus terms were used confusingly by some

authors, pure dermal cyst of this region was not encountered in this study. Few cases of mass in the nose and frontonasal area without sinus were encountered. In clinical examinations and investigations, no sinus was found and hence excluded from the study. In all the cases, the family history of a similar sinus was especially enquired. Anteroposterior and lateral X-ray was taken in some early cases to assess the patients, and since 2008 CT or MRI scans were done.

Two patients had associated anorectal malformation with this condition. In one patient, excision of punctum was done along with the pull-through operation. Other patient was offered a simultaneous operation for dermal sinus and the anorectal malformation operation. Investigations were done. But, the parents wanted to postpone the operation for nasal sinus to a later date. All the eight sittings of operations of the seven patients were done under general anaesthesia. All the excised specimens were sent for histopathological examination. We have obtained the required informed consent from the legal guardian for publishing these works in a scientific journal.

RESULTS

During this fourteen year study period, we encountered only eight cases of the nasal dermal sinus. Out of these, six patients were male, and two were female. Age of diagnosis ranged from day 2 to 6 years and operation from 5 months to 14 years (**Table 1**).

Table 1 Age distributions of the patients at presentation and operation

Cases	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8
Age at presentation	Day 2	1 yr	6 m	2 yr	1½ m	11 m	6 yr	6 m
Age at operation	5 m &14 yr	1 yr	9 m	2 yr 2 m	6 m	1 yr	6 yr	Waiting

Figure 1 14 years old girl with recurrent congenital nasal dermal sinus

In a female patient, the diagnosis of this condition was made at the neonatal period. She had anorectal malformation as well. The punctum region's incomplete excision was only done at five months of age and her anal transposition operation. She had a recurrence and came for treatment at 14 years of age (**Figure 1**). Re-operation was done with complete excision of the tract.

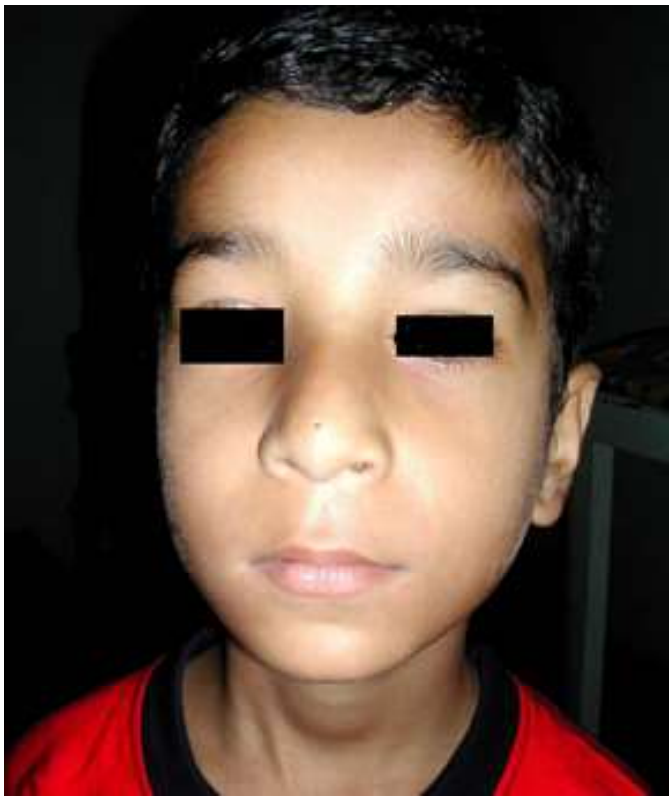

Figure 2 Typical case of nasal dermal sinus without any hair in it

Opening in the dorsum of the nose since birth was the leading complaint (**Figure 2**). One patient (Case No 4) had openig in the middle part of the nasal bridge.

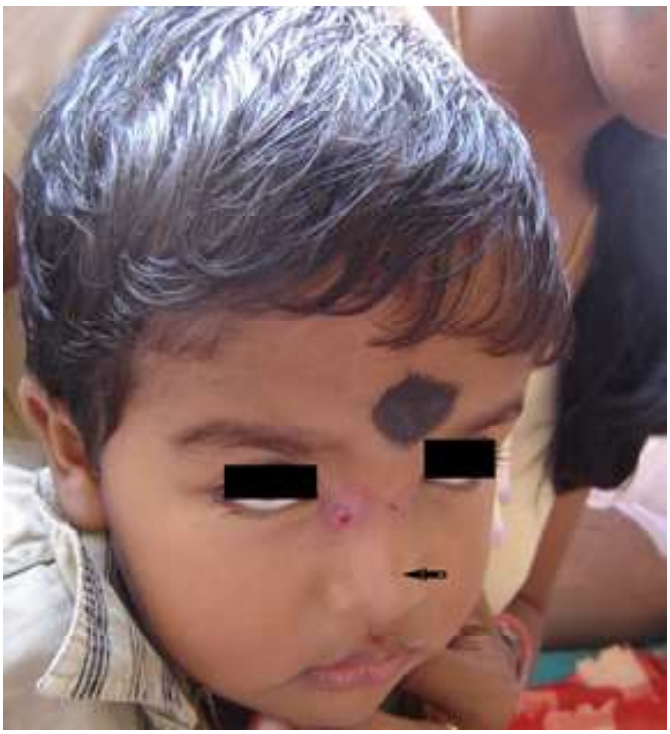

Figure 3 Infected nasal dermal sinus. Punctum can be seen away from the site of infection (Arrow)

He also had slight widening of the nose, inflammatory swelling in the nasal bridge region with a pus discharging sinus on the right side of nasion, close to lacrimal Sinus (**Figure 3**).

The patient was treated with antibiotics and operated on two months later as the infection subsided (**Figure 4**). In another male patient (case number 8), it was an incidental finding. Parents were unaware of the consequences of the condition.

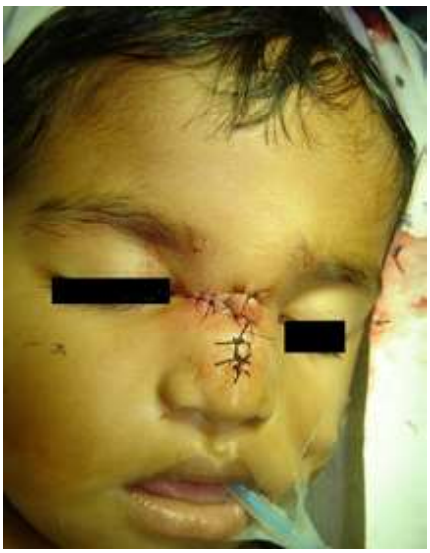

Figure 4 Postoperative view of the patient of figure 3 after the subsidence of infection

This case was associated with anorectal malformation. Though the patient came to the hospital in the neonatal period, the condition was diagnosed only at six months of age while went for a pull-through operation. Parents wanted to wait for the operation of this condition till the treatment for an anorectal malformation is over. In no patients, a family history of a similar condition was present.

Various locations of the openings are as follows. Six cases had their external openings in the nasal bridge, mainly in the lower part. Each was in the base of the columella and the tip of the nose (**Table 2**). Three patients with the opening in the nasal bridge had single coarse hair coming out of the opening (37.5%).

Table 2 Locations of external openings with or without hair

Cases	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8
External opening	Lower nasal bridge	Lower nasal bridge	Tip of nose	Mid nasal bridge	Base of columella	Junction & bridge of nasion	Lower nasal bridge	Junctio of bridge and tip
Presence of hair/ infection	Hair present	None	None	Infected	None	Hair Present	Hair Present	None

In two cases, the disease was associated with anorectal malformations and out of them, each was from either sex.

Computed tomography (CT) scan [n=3 (37.5%)], magnetic resonance imaging (MRI) scan [n=2 (25%) plain X-ray [n=3 (37.5%) was done. Only X-rays were done in three cases and all these were before 2008.

No sinogram was done.

Changes found in the CT and MRI were identical. There was separation of nasal bones; crista galli was bifid and had

wide foramen caecum (**Figure 5**). The tract of various lengths could be seen in the MRI scan. No cases had the defect of the cribriform plate and evidence of associated intracranial mass.

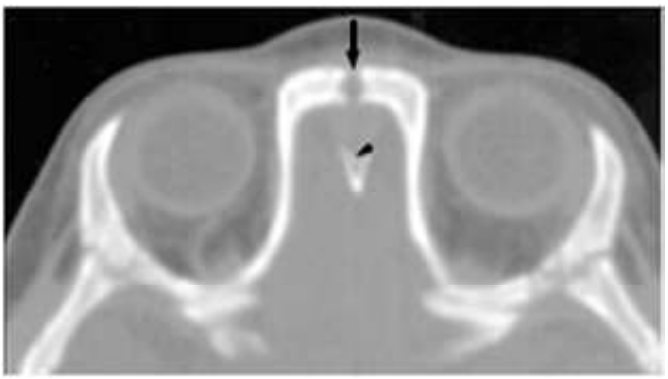

Figure 5 CT scan shows separated nasal bone (Arrow) and wide foramen caecum (Arrowhead)

No case of nasal dermal sinus encountered, which had its intra-cranial extension.

All the patients were operated on under general anaesthesia. A midline vertical incision was made in the nasal crest. The punctum was removed in continuity with an encircling incision. The whole tract was dissected out (**Figure 6**). In all the cases, nasal cartilage and nasal bone failed to fuse in the midline, making the dissection easier. The last part of the track had a thin fibrous cord going towards the foramen caecum divided by sharp dissection. The only complex case was the Case No 4, where skin and deeper structure were adherent due to scarring from a previous infection.

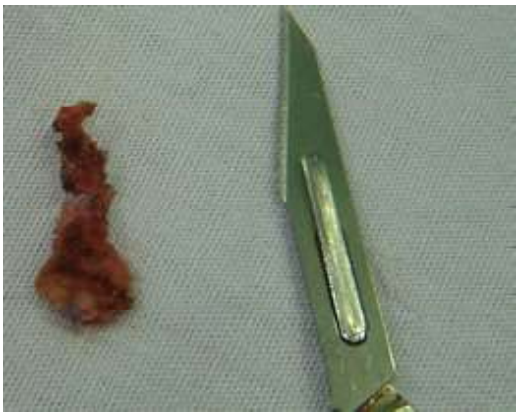

Figure 6 Excised specimen

No cases required any filling or plastic reconstruction procedure. Postoperative management was as usual. No dressing was applied on the wound from day 2 of the operation.

Histopathological examinations of all the excised tissues were done. In light microscopy, the lining of the sinus was squamous epithelium. Other ectodermal elements like hair follicle, sebaceous glands, and sweat glands were also noted. In the sinus with hair, the hair originated from a hair follicle lying inside the sinus. No endodermal derivative was noted.

The systemic antibiotic was given to all the cases.

Stitch removal was done on the 5th day of operation in most of the cases. In case no four, it was done on day 7. Mean hospital stay was six days.

Except in case no four, where there was a history of the previous infection, scars were nominal.

Due to incomplete excision in the first operation (Case No 1), there was one recurrence. It was reoperated at 14 years of age. This case was followed up for six months, and there is no recurrence to date. In other cases, follow-up was for 1year to 13 years and meant follow up was six years. There was no recurrence in these cases.

DISCUSSION

Nomenclature and classification of this condition are confusing.[3] Many authors have used the term dermal sinus and dermal or dermoid cyst of the nose equivocally.[3] In some publications, many unrelated conditions of this region have also been included.

In some studies, a midline nasal mass was the main presenting feature. This was because of the fact mentioned above. As we included only sinus cases, it was the main presenting feature in our series. Hair protruding from the punctum was also a prominent feature. Because of this, some authors have named the dermal sinus of the nose with a hair as Nasal Pilonidal Sinus.[5-7]

Various associated anomalies like ear and craniofacial anomalies were observed in other series.[8] Our series had 2 cases (25%) with anorectal malformation. But no literature with this association was found.

Bifid crista Galli and enlarged foramen caecum in CT and MRI scan is a common sign, and it does not indicate any intracranial extension of the sinus. But the normal-sized foramen confirm the absence of this type of extension.[9] In our series, we found the above bony changes in all the patients, but none had a defect in the cribriform plate. We did not encounter any case with intracranial extension. A narrow fibrous band may be found in the continuity of the tract. It may go beyond the nasal bone up to the nasofrontal suture. Instead of doing extensive dissection, this can be safely divided across at the level of termination of the tubular structure.[10]

Squamous epithelium, sebaceous gland and dermal elements like hair in HPE indicate that this type of sinus develops from ectoderm by sequestration in the fourth to sixth weeks of intrauterine life as forwarded by Bland-Sutton (1893). The presence of a hair follicle differentiates it from the epidermoid cyst (Sebaceous cyst). While the absence of endodermal

element excludes its teratomatous origin.[11]

Many authors employed midline vertical incision. We also employed the same. Many authors advocate other incisions like the transverse, inverted-U, and transnasal approach.[9,11,12] The incisions other than midline vertical type were used for nasal cyst excision and wherever rhinoplasty was required. We did not encounter any difficulty in excising the tracts from the nasal approach. Only case no 4 inverted L shaped incision was employed to excise the scar with the scar of previous infection in continuity.

Recurrence rates in other series were 30% to 100%.[13] Mostly, it was due to incomplete excision and previous infection. In our series, one case out of seven patient operated on had recurrence. There was no recurrence in this case after the second operation. In other cases, there was no recurrence.

CONCLUSION

The present series is a small one as the condition is rarely encountered in our region. We have included only midline nasal sinuses here without any ambiguity. Hence, in our opinion, paper bears some unique characteristics. In our view, nasal dermal sinus and dermal cyst without fistulae are two different entities. Most of these sinuses are limited to the nose outside the foramen caecum, and intracranial extension is rare. The majority of the cases can be managed by excision through the skin of the nasal region. Cranial exploration is very rarely required.

REFERENCES

1. Hanikeri M, Waterhouse N, Kirkpatrick N, Peterson D, Macleod I. The management of midline transcranial nasal dermoid sinus cysts. Br J Plast Surg 2005;58(8):1043-50.
2. Hedlund G. Congenital frontonasal masses: developmental anatomy, malformations, and MR imaging. Pediatr Radiol 2006;36(7):647-62.
3. Rahbar R, Shah P, Mulliken JB, Robson CD, Perez-Atayde AR, Proctor MR, Kenna MA, Scott MR, McGill TJ, Healy GB. The presentation and management of nasal dermoid: a 30-year experience. Arch Otolaryngol Head Neck Surg 2003;129(4):464-71.
4. Pollock RA. Surgical approaches to the nasal dermoid cyst. Ann Plast Surg 1983;10(6):498-501.
5. Sreedharan S, Kamath PM, Hegde MC, Subramaniam V, Lobo FD. Pilonidal sinus of nose–a diagnostic dilemma. Jurnalul de Chirurgie 2010;6:343-7.
6. Jones R. Congenital pilonidal sinus of the nose. Aust N Z J Surg.1992;62:492.
7. Montasir Junaid, Sadaf Qadeer Ahmed, Maliha Kazi, and Naeem Sultan Ali. Pilonidal sinus involving the nasal bridge: a rare manifestation. BMJ Case Rep 2015Jul 6; 2015: PMID: 26150636.
8. Wardinsky TD, Pagon RA, Kropp RJ, Hayden PW, Clarren SK. Nasal dermoid sinus cysts: association with intracranial extension and multiple malformations. Cleft Palate Craniofac 1991;28(1):87-95. PMID:2004099.
9. Denoyelle F, Ducroz V, Roger G, Garabedian EN. Laryngoscope. Nasaldermoid sinuscysts in children 1997;107(6):795-800.PMID:9185736.
10. Bartlett SP, Lin KY, Grossman R, Katowitz J. The surgical management of orbitofacial dermoids in pediatric patient. PlastReconstr Surg.1993;91(7):1208-15. PMID:8497520.
11. Posnick JC, Bortoluzzi P, Armstrong DC, Drake JM. Intracranial nasal dermoid sinus cysts: computed tomographic scan findings and surgical results. Plast Reconstr Surg 1994;93(4):745-54; discussion 755-6. PMID:8134433.
12. Bradley PJ. Results of surgery for nasal dermoids in children. J Laryngol Otol 1982;96(7):627-33. PMID:7086278.
13. Vibe P, Lontoft E. Congenital nasal dermoid cysts and fistulas. Scand J Plast Reconstr Surg1985;19(1):105-7. PMID: 4023638.