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FOCUS SESSION

The reconstruction of skull base defects in infants using pedicled nasoseptal flap—a review of four cases

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Abstract

Introduction Benign lesions of the skull base are common in the paediatric population, and are usually congenital in aetiology. Majority of these lesions are treated transcranially exposing the patients to a number of risks. Although endoscopic endonasal surgery (EES) helps avoid many of these potential morbidities, CSF leak with its attendant complication remains a big concern. This study reports the use of the Hadad flap in the reconstruction of skull base defects in infants to prevent this problem. The study was conducted on four infants with a mean age of 7 months, who underwent repairs for CSF leaks associated with congenital lesions like meningocele or meningoencephalocele, using the Hadad flap. Of the four patients, three cases were revision cases and one was primary where the patients presented with complaints such as nasal obstruction and watery nasal discharge. Post surgery, the infants were monitored for a mean period of 23 months and no major complications or recurrent CSF drainage were observed. Minor complications that were observed include vestibulitis and crusting in the nose. The utility of the Hadad flap in the reconstruction of skull base defects in the paediatric age group has been controversial while its utility in infants has not been studied in literature so far. We report here in our series four infant patients in whom we believe that the nasal septum and the skull base will develop proportionally to each other, hence lowering the chances of a short flap and eliminating the occurrence of future complications.

Conclusion The nasoseptal flap is an effective and safe technique for reconstructing skull base defects in infancy. It can be concluded that this technique does not have any potential effect on septal or craniofacial growth as the flap is harvested only on one side with normal mucosal cover on the other side. There is no posterior septectomy or any form of bony or cartilaginous resection that is performed, hence avoiding any effects on bony growth. No studies have been published in literature so far and to the best of our knowledge, this is the first report describing the efficacy of the nasoseptal flap in infants.

Keywords Nasoseptal flap · Hadad flap · Infancy · Congenital skull base lesions

Introduction

Benign lesions of the skull base are common in the paediatric population, and are usually congenital in aetiology. Majority of skull base lesions were treated traditionally by transcranial approach. A bifrontal craniotomy exposes the patient to the risks of blood loss, brain retraction, disruption of growth centres, and injury to the supraorbital and supratrochlear neurovascular complexes during the surgical exposure of the anterior skull base from above. Endoscopic endonasal surgery (EES) avoids many of these potential morbidities [1].

Adequacy on endonasal approach for treating a CSF leak endoscopically is a major worry, apart from recurrence and nasal morbidity in the form of crusting and the potential effect it could have on the craniofacial growth. However, with the advent of vascularised nasoseptal flap [Hadad flap], the incidence of CSF leaks has decreased drastically [2]. The utility of the Hadad flap in reconstructing skull base defects in the paediatric age group has been questioned in literature and there is

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no study describing its utility in infants so far. The risk of the septal flap adversely affecting septal and craniofacial growth is a factor to bear in mind and we were encouraged by our experience of using this flap in the paediatric age group [above 5 years] in whom no adverse outcomes have been noted even after many years of follow-up [3–8 years]. Hence, we reviewed four cases treated so far and the utility of this flap in infants and report the use of the nasoseptal flap in reconstructing skull base defects in infants.

Materials and methods

The aim of this study was to assess the utility of the Hadad flap in infants with congenital skull base lesions.

This is a retrospective review, performed on all patients who had CSF leaks associated with congenital lesions like meningoencephalocele. This study was conducted at a tertiary referral centre with a team consisting of a neurosurgeon, an ENT surgeon, a paediatric anaesthetist, a paediatrician, and an intensivist.

Four infants underwent repair of CSF leaks using the Hadad flap. Data collected included the patient's age, gender and other details like clinical presentation, duration of the illness, site of involvement, methods of repair, complications, recurrence, and follow-up**.** CSF leaks were confirmed by clinical history, CT/MRI cisternography, and intraoperative visualisation of a CSF fistula with the presence of meningocele and meningoencephalocele.

Clinical presentation

Of the 4 patients, 3 cases were recurrent cases while 1 was primary. The complaints presented were mainly nasal obstruction and watery nasal discharge. One child developed meningitis after the primary transcranial surgery. Out of the three recurrent cases, two cases presented a failure from transcranial surgery while one from a transpalatal approach. The mean duration of recurrence after the primary surgery was 20 days.

Diagnosis

All the infants underwent neuroradiological evaluation with computerised tomography [CT] and magnetic resonance imaging [MRI] of the nose, paranasal sinuses, and brain. Additionally, a CT Angiography was done to identify the presence of a blood vessel in the herniated tissue. Collection of fluid for Beta 2 transferrin assay was not possible in any of our cases. Intraoperatively, prior to beginning the surgery, a diagnostic nasal endoscopy was performed in all cases.

Instrumentation

In all cases, a 2.7-mm Hopkins rod endoscope was used. The instruments essentially consisted of micro ear instruments. Coagulation and haemostasis were achieved by endoscopic bipolar forceps.

Surgery

The principle of the surgery was essentially the same in all our cases with slight variation from case to case. The surgery was done under general anaesthesia without a lumbar drain. After thorough decongestion of the nose, a diagnostic nasal endoscopy was performed. The meningocele/meningoencephalocele was identified. In all our infants, no posterior septectomy or reverse Hadad flap was performed in order to minimise the possible effects of bony or cartilaginous resection on septal and craniofacial growth. In addition, as the blood supply and periosteum of the opposite side remain intact, the impact on the septum is also reduced**.** The defect was identified, surrounding mucosa denuded and covered with the Hadad flap. This was reinforced with tissue glue, surgicel, and gel foam. The bare area on the septum was covered with a silastic splint sutured to the septum. The nose was packed with a small nasal pack.

Case 1 This infant had a defect in the anterior medial cribriform plate on the right side with meningoencephalocoele herniating. After identifying the meningoencephalocoele, bipolar-assisted reduction was done. The gliotic segment was coagulated and excised. Care was taken to prevent any inadvertent entry into the cranial cavity. The site of the defect was traced and careful coagulation was done with endoscopic bipolar forceps. The mucosa around the defect was denuded bare to the bone. Hadad flap was harvested from the side of the lesion. The defect was reconstructed with an underlay fascia and an overlay Hadad flap. This was reinforced with tissue glue and gel foam. The bare area on the septum was covered with a silastic splint. A right-sided nasal pack was placed postoperatively for 5 days and the splint was removed 2 months later.

Case 2 An 8-month-old infant had earlier undergone a transcranial procedure. In this infant, there was a suspicious leak with a meningoencephalocoele. The cranial bone and pericranium had been used in the primary procedure to repair a wide posterior cribriform defect. However, the patient developed meningitis 2 days later. At this point, a transnasal procedure was planned. On transnasal endoscopic examination, the posterior septum was found deficient due to the compressive effects of encephalocoele. A unilateral flap with a thin inferior pedicle was raised from the posterior septum. This flap was used as overlay after repositioning the intracranial bone which

had gotten tilted. This repositioning was done transnasally using an elevator until it felt straight with palpation. Once again, the bare septum was covered with a silastic splint and the nose packed with a mini Merocel nasal pack.

Case 3 In this case, a transcranial repair had been performed, 3 months after which the child developed a leak. The leak was from the anterior part of the cribriform plate. The meningocele was coagulated and the defect was denuded with bare bone all around. A nasoseptal flap was used as an overlay. The flap fell marginally short and hence a small portion of tensor fascia lata was used to cover the anterior part of the defect over which the Hadad was placed. The flap now covered the fascia and the bare bone. This was reinforced with tissue glue and gel foam. The silastic splint was kept for 6 weeks and the nasal pack was removed on the 5th postoperative day.

Case 4 This patient had a large defect from the fovea ethmoidalis to the clivus and had presented with inability to feed and nasal blocking. A transpalatal repair had been attempted elsewhere by a different team who were not successful in completing the repair and the child subsequently developed an active leak**.** Ten days after the primary procedure, a transcranial surgery was performed. The transcranial repair was done on the anterior part of the defect up to the planum. Two days later, the child underwent an endoscopic repair. Endoscopic examination revealed a large meningoencephalocoele in the nasopharynx. The mass was excised and the margins of the defect identified and freshened. The lower level of the defect was at the level of Eustachian tube. The flap was rotated downwards to cover the defect. Post-surgery, a no. 3 endotracheal tube was cut and placed in the nasal cavity to allow for nasal breathing. In the other nasal cavity, a trimmed nasal pack was placed. Imaging and intraoperative pictures of this patient are represented in Figs. 1 and 2.

Postoperatively, on the fifth day, pack removal was done and a leak from the inferior edge was suspected. The flap was then reinforced with a small piece of fascia lata which was then kept between the lower portion of the flap and the nasopharyngeal mucosa. This was again reinforced with glue and gel foam. Nasal packing was done again for 3 days. Post procedure, after removal of the pack, the infant is doing fine. The silastic splint is to be removed after 2 months.

Postoperative care

All the infants were monitored by an intensivist in a neonatal intensive care unit. Intravenous antibiotics, namely a 3rd-generation cephalosporin, was administered. Nasal saline drops were used. The pain was monitored so as to reduce the crying of the infant and to avoid a subsequent raise in the intracranial pressure. Careful monitoring of the vitals was done**.** The anterior nasal packs were removed on the 5th postoperative day and the silastic sheet was removed 2 months later. Intermittent nasal debridement was done to clear the crusting. All these procedures were done under short general anaesthesia.

Results

Four infants who had congenital defects in the skull base were managed with the Hadad flap in our series. The mean age of the infants was 7 months. Of the four infants, two infants underwent a transcranial repair and one a transpalatal repair. Only in one infant, a primary endonasal surgery was performed.

The details of each case are summarised in Table 1.

There were two males and two females in our study, with a mean age of 7 months. The youngest child was 3 months old. The defect was located in the cribriform plate in two infants, in the sphenoethmoid junction in 1 infant, and in the sphenoclival junction in the fourth.

No major complications have been observed in our study. Minor complications included vestibulitis and crusting in the nose.

No evidence of recurrent CSF drainage has been detected, to date, after a mean follow-up of 23 months.

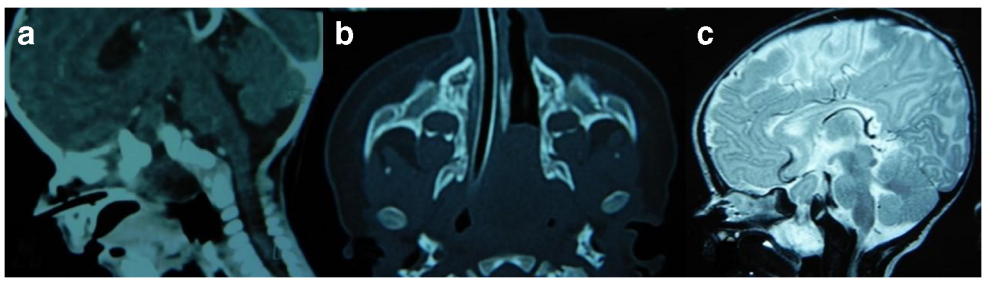

Fig. 1 **a** CT brain sagittal cut showing defect in the sellar floor with herniation of meninges and gliotic neural tissue with CSF extending inferiorly up to the nasopharynx. **b** CT PNS showing the visualised soft tissue obscuring the nasopharynx completely. **c** Sagittal T2–weighted MRI sequence with CSF seen extending inferiorly up to the nasopharynx. Pituitary gland cannot be visualised separately from the mass. Anterior cerebral artery is seen at the level of planum sphenoidale with no vessels herniating into the herniated tissue

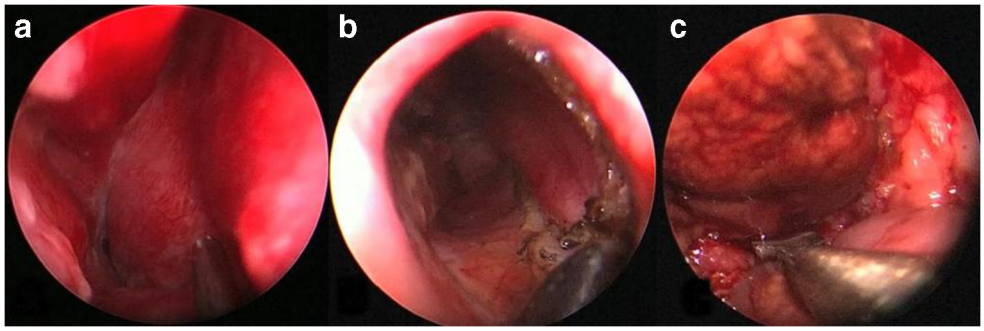

Fig. 2 Intraoperative images. **a** Intraoperative endoscopic image showing a bulge of the meningoencephalocoele in the choana and nasopharynx. **b** Markings of the Hadad flap. **c** Showing repositioning of the Hadad flap to reconstruct the defect after excising the meningoencephalocoele

Discussion

With the advent of endoscopy, there has been a paradigm shift in the skull base surgeries from the transcranial route to an endonasal route. A skull base lesion in adults is preferably approached through a transnasal route. However, the same in children is usually approached through a transcranial route**.** The reasons for a transcranial route being preferred are as follows:

1. Narrow corridor-small nasal cavity
2. Difficulty in manoeuvring instruments
3. Need for small-sized instruments
4. Difficulty in reconstructing a large defect which could potentially occur
5. Difficulty in obtaining sufficient length and width of the flap
6. Effect of this surgery on craniofacial growth.

The Hadad flap based on nasoseptal artery (Fig. 3) is a revolutionary technique that has drastically reduced the incidence of recurrent CSF leaks. The utility of this flap in the reconstruction of skull base defects in the paediatric age group has been controversial while its utility in infants has not been studied in literature so far.

A study, *Endoscopic pedicled nasoseptal flap reconstruction for paediatric skull base defects*, was conducted by RN Shah et al. in 2009, where they studied 6 paediatric patients (less than 14 years of age). They concluded that the pedicled nasoseptal flap may not be a viable option for EEA reconstruction in children < 10 years of age. This flap is a reliable option for patients > 14 years of age, as their septums are comparable to adults. Patients 10 years to 13 years of age require careful consideration of facial analysis and preoperative radioanatomic evaluation on an individual basis [3].

Another study by Patricia L. Purcell in 2015 concluded that the length of the nasoseptal flap is not a limiting factor in the reconstruction of paediatric sellar defects. When compared to older patients, younger patients tend to have greater nasoseptal flap length relative to sellar defect length. The mean age in their study was 15 years [4].

Our understanding of this controversy in the literature is that although the cranio-orbitozygomatic skeleton reaches 85% of adult size by 5 years of age, the septum attains its adult size only by puberty. Hence, there is a mismatch between the required lengths of the flap to the defect in sella. Recent literature has suggested that the nasoseptal flap is only a viable option in subjects older than 6 to 7 years [5].

Another study conducted by Ghosh et al. in 2015 in which the clinical outcome of 16 patients was analysed according to the age groups (10 years and under, over 10 years), revealed only one postoperative CSF leak in a patient greater than 10 years. The leak was caused due to an intraoperative error rather than inadequate flap coverage; the flap was noted to have complete coverage. Their results demonstrated that septal lengths in children less than 10 years of age were adequate to cover the defects created by the suprasellar resections [6].

They further stratified patients into two groups: under 6 years and over 6 years. Results clearly depicted sufficient potential nasoseptal flap length and width for the potential suprasellar defect. When compared, the total potential length was reliably significantly longer than the potential defect length, with P values < .05. Thus, there was consistently more than adequate coverage.

A recent study in 2018 by Ben Ari in 2018 concluded in their study that endoscopic endonasal NSF was both an effective and a safe technique for anterior skull base defect reconstruction in 12 children for both benign and malignant neoplasms. It had a high success rate and a low complication rate. No apparent negative influence on craniofacial growth was observed in their series [7].

We report our series of 4 patients and believe that the nasal septum and the skull base develop proportionally to each other, hence lowering the chances of a short flap. Nevertheless, a detailed preoperative radiological evaluation, measuring the size of the defect and the length of the flap, would be useful in identifying rare cases where there could be a possibility of failure. A pedicled nasoseptal flap on a secure repair poses a greater advantage than a free tissue repair**.** The flap is taken only from one side of septum without any posterior septectomy. We believe maintaining a normal mucosal cover on one side will prevent any growth issues. Although the

Table 1 Details of cases 1 to 4

Sr. no.	Age	Gender	Clinical presentation	Site of involvement	Prior surgery	Method of repair	Complications	Recurrence	Follow-up
Case 1	4 months	Male	Nasal obstruction	Right anterior medial cribriform	No	Transnasal endoscopic Coagulation of the meningocele Underlay fascia and an overlay Hadad flap	None	None	4 years
Case 2	8 months	Male	Nasal obstruction and watery nasal discharge	Sphenoethmoidal defect. Posterior ethmoid and sphenoid	Yes-Transcranial	Transnasal endoscopic Transnasal manoeuvring of the bone with elevator was done to reposition the displaced bone over which the Hadad flap was layered	None	None	3 years
Case 3	6 months	Female	Watery nasal discharge	Right medial cribriform plate	Yes-Transcranial	Transnasal endoscopic Coagulation of the meningocele anterior half of the defect was covered with fascia lata and overlay Hadad flap	None	None	6 months
Case 4	3 months	Female	Inability to feed and nasal obstruction	Fovea ethmoidalis up to the clivus	Yes-transpalatal	Transcranial following a craniotomy pericranial flap was layered up to the planum Ten days later, transnasal endoscopic repair of the sphenoclival defect was done. It was layered with the Hadad flap	5th postoperative day after the transnasal approach, On removing the pack, there was a probable leak from the inferior edge of the defect which was layered again with small fascia and glue	None	1 month

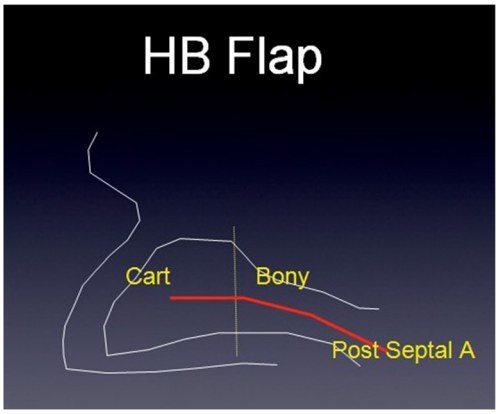

Fig. 3 Line diagram showing the markings of the nasoseptal flap [Hadad flap]

septum will most likely reepithelialise in 4 weeks, the splint, which is quite inert, was retained for 6–8 weeks in order to avoid crusting or frequent cleaning under GA.

Limitation of our study

Preoperative radiological analysis on the length of the nasoseptal flap versus the size of defect was not done.

Conclusion

The nasoseptal flap is an effective and safe technique for reconstructing skull base defects in infancy. It can be concluded that this technique does not have any potential effect on septal or craniofacial growth as the flap is harvested only on one side with normal mucosal cover on the other side. There is no posterior septectomy or any form of bony or cartilaginous resection performed, hence avoiding any effects on bony growth. No studies have been published in literature so far and to the best of our knowledge, this is the first report describing the efficacy of the nasoseptal flap in infants.

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Compliance with ethical standards

Conflict of interest The authors declare that there is no conflict of interest.

References

1. Gump WC (2015) Endoscopic endonasal repair of congenital defects of the anterior skull base: developmental considerations and surgical outcomes. J Neurol Surg B Skull Base 76(4):291–295. https://doi.org/10.1055/s-0034-1544120
2. Wheless SA, McKinney KA, Carrau RL, Snyderman C, Kassam A, Germanwala A, Zanation A (2011) Nasoseptal flap closure of traumatic cerebrospinal fluid leaks. Skull Base 21(2):93–98. https://doi.org/10.1055/s-0030-1266763
3. Shah RN, Surowitz JB, Patel MR, Huang BY, Snyderman CH, Carrau RL, Kassam AB, Germanwala AV, Zanation AM (2009) Endoscopic pedicled nasoseptal flap reconstruction for pediatric skull base defects. Laryngoscope. 119(6):1067–1075. https://doi.org/10.1002/lary.20216
4. Purcell PL, Shinn JR, Otto RK, Davis GE, Parikh SR (2015) Nasoseptal flap reconstruction of pediatric Sellar defects. Otolaryngol Neck Surg 152(4):746–751. https://doi.org/10.1177/0194599815571284
5. Ghosh A, Hatten K, Learned KO, Rizzi MD, Lee JY, Storm PB, Palmer JN, Adappa ND (2015) Pediatric nasoseptal flap reconstruction for suprasellar approaches. Laryngoscope. 125:2451–2456. https://doi.org/10.1002/lary.25395
6. Ghosh A, Hatten K, Learned KO et al (2015) Pediatric nasoseptal flap reconstruction for suprasellar approaches. 125:2451–2456. https://doi.org/10.1002/lary.25395
7. Ben-Ari O, Wengier A, Ringel B, Carmel Neiderman N, Ram Z, Margalit N, Fliss D, Abergel A (2018) Nasoseptal flap for skull base reconstruction in children. J Neurol Surg Part B Skull Base 79(01):037–041. https://doi.org/10.1055/s-0037-1617435