Analysis of Various Factors and Techniques to Improve Outcome of Surgical Management of 243 Cases of CSF Rhinorrhea

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Abstract Cerebrospinal fluid (CSF) rhinorrhea occurs when there is a communication between subarachnoid space and sinonasal mucosa due to meningeal, osseous and mucosal defects in the cranial base leading to discharge of CSF from the nose. The risk of developing meningitis after CSF rhinorrhea may vary from 5.6 (Leech and Paterson in *Lancet* 1:1013–1016, 1973) to 60% (Eljarnel and Foy in *Br J Neurosurg* 5:275–279, 1991). Hence surgical management of CSF rhinorrhea is highly recommended. Transnasal endoscopic approach first described by Wigand in 1981, has been proven to be the approach of choice in comparison to intracranial and external nasal approach (Jones and Becker in *Br Med J* 322:122–123, 2001) in most cases. The next defining milestone was the pedicled naso septal vascularized flap described by Hadad et al. (*Laryngoscope* 116(10):1882–1886, 2006), which could be used to manage large defects. In the present study we assessed 243 cases of CSF rhinorrhea managed by transnasal endoscopic approach. We compared the various factors associated with CSF rhinorrhea and the correlation with the outcome of the surgical treatment. We also analyzed the different sites and techniques of surgical repair and have certain recommendations to improve the surgical outcome. The commonest cause of CSF leak was spontaneous (54.32%) and the commonest site was cribriform plate (43.24%). Patients presented most commonly with watery nasal discharge (82.3%). CT scan with cisternography or MR cisternography is the gold standard to identify the suspected site of leak. Out of 243 patients, 77.77% were operated using free

grafts and 22.22% by flap repair. Results are comparable. Hence we would advise simple conservative technique with free grafts to reduce morbidity and shorten the post-operative recovery.

Keywords CSF rhinorrhea · CT cisternography · MR cisternography · Transnasal endoscopic repair · Free flap · Hadad flap

Objective

A retrospective detailed analysis of etiological factors, clinical presentation and treatment modalities was done and correlated with the outcome of surgical treatment. Findings of the case series was compared to findings of other studies.

Materials and Methods

A comprehensive retrospective analysis of 243 cases of CSF rhinorrhea from 1996 to 2018 was done. These patients were managed at Bombay Hospital, Mumbai (a tertiary care hospital). The diagnosis of CSF rhinorrhea was established by history, clinical assessment and radiological diagnosis. The gold standard for diagnosis is CT with cisternography. In some cases, if lumbar puncture was not possible or patient did not give consent, then plain CT scan with MR cisternography was done. In all cases of encephalocele or meningoencephalocele an MRI is essential. Whenever there was doubt about the nature of the nasal fluid, a beta 2-transferrin test was used to confirm the diagnosis. Patients with traumatic CSF leakage were given conservative treatment of head end elevation, lumbar drain,

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antibiotic coverage and diuretic therapy. Coughing, sneezing, lifting of heavy weight was to be avoided. The risk of developing meningitis after CSF rhinorrhea may vary from 5.6 [1] to 60% [2]. Hence traumatic CSF rhinorrhea which does not stop or reduce after 1 week of conservative management, spontaneous CSF leaks and CSF leaks of congenital etiology were repaired by transnasal endoscopic technique, as applicable. Cases of pituitary surgery operated outside and referred with CSF leaks were included. Our own cases of pituitary surgery with intraoperative CSF leaks were excluded. All cases were operated by single author Dr. Nishit Shah.

The surgical techniques vary according to the site of the defect. For defects involving the medial cribriform plate the defect was approached without sacrificing the middle turbinate and defects of lateral cribriform plate required the removal of middle turbinate and anterior and posterior ethmoidectomy and frontal sinus surgery. The lateral recess of sphenoid sinus was approached via posterior wall of maxillary sinus leading through the pterygopalatine fossa and pterygoid plates into the lateral recess of sphenoid. Small defects were repaired with free grafts and fat and tissue glue. Moderate size defects were repaired with fat plug, cartilage, free grafts and tissue glue. Large and multiple defects were repaired with fat plug, cartilage, vascularized nasoseptal flap (Hadad flap [3]) and tissue glue. In all cases merocel (Medtronic) pack was kept in situ for 1 week.

Data collected included etiology, location of defect, type of repair, graft material used, complications and need for revision surgery. Success rate of first and subsequently performed repair surgeries was analyzed. Only patients who approved of the review of their data were included in the study.

Results

The mean age of our 243 patients was 35.9 years, ranging from newborn to 72 years. 58.02% (141/243) were females and 41.97% (102/243) were males.

The commonest cause of CSF leak was spontaneous 54.32% (132/243) followed by trauma 25.10% (61/243), iatrogenic in 16.04% (39/243) and least common is congenital cause of CSF rhinorrhea 4.52% (10/243). We did not consider in this study, over 400 cases of intra-operative CSF leaks during endoscopic skull base surgery (about 1500 operated cases), only those that occurred during sinus surgery were considered.

The commonest site of CSF leak was cribriform plate in 43.24% (105/243), followed by sphenoid sinus in 27.98% (68/243), multiple sites in 11.93% (29/243) cases and ethmoid sinus in 11.11% (27/243) cases. Frontal sinus was the least common site of CSF leak in 2.057% (5/243).

The commonest presentation, is watery nasal discharge in 82.3% (200/243) patients, followed by meningitis in 9.46% (23/243), headache in 4.93% (12/243), fever in 1.23% (3/243), altered sensorium in 1.23% (3/243) and giddiness and pneumocephalus in 0.41% (1/243).

96.29% (234/243) cases presented with active leak, were treated surgically 0.3.7% (9/243) were traumatic cases without active leak but had history of meningitis (6/9 cases) or recurrent leaks (3/9 cases). Out of 6 cases who presented with meningitis, 3 showed suspect area of leak on radiographic examination and were treated with selective grafts and 3 cases showed no suspect areas, hence were treated with carpet grafts. 3 cases presented with recurrent leaks but no active leak demonstrated by radiographic examination, therefore were treated surgically with carpet grafts. Out of the 243 patients treated surgically, 77.77% (189/243) were operated using free grafts and 22.22% (54/243) by flap repair. Free grafts had 99.47% (188/189) success rate and flap grafts had 90.74% (49/54) success rate.

Success of surgery was assessed in terms of closure of defect and stoppage of CSF rhinorrhea including early and late postoperative period. The success rate of primary surgery was 97.53% (237/243). 6 cases needed 2nd transnasal endoscopic surgery, which had 83% (5/6) success.

The complications seen in our case series were bleeding 1.23% (3/243) and recurrence of CSF leak in 2.46% (6/243). 0.41% (1/243) patient died due to sepsis as a result of meningitis. The 3 cases of bleeding occurred from anterior ethmoidal artery, posterior ethmoidal artery and descending palatine branch of maxillary artery were successfully controlled intraoperatively.

Discussion

Case series of 243 cases of CSF rhinorrhea at Bombay Hospital in over 2 decades were retrospectively analyzed. The cases were operated by transnasal endoscopic procedure because this technique has been proven to have excellent success rate and lower morbidity [4–11] compared to intracranial and external nasal approach [12]. The complications seen in intracranial approach include intracranial haemorrhage, blindness, cerebral edema, meningitis and in transfacial approach are facial numbness, septal perforation and orbital complications, including diplopia and epiphora [13].

Ommaya classified etiology of CSF Rhinorrhea into 1. Traumatic due to accidental and iatrogenic cause 2. Non-traumatic due to intracranial tumours, hydrocephalus, congenital anomalies and idiopathic cause. In our study CSF leak occurred most commonly due to spontaneous cause (54.32%), followed by accidental trauma (25.10%),

iatrogenic (16.04%) and least common cause was congenital (4.52%). Some studies report head injuries as the commonest cause whereas some report iatrogenic trauma as the commonest cause. However in our case series, the commonest cause was spontaneous. This can be attributed to the fact that, in most traumatic cases (> 50%) rhinorrhea stops within 1 week and in most cases within 6 months. Ours is a tertiary care hospital and included mostly those traumatic cases in which CSF rhinorrhea did not stop with conservative management.

Commonest site of CSF leak was cribriform plate (43.24%). This can be attributed to the fact that the lamella of the cribriform plate is the thinnest part of the bone in the anterior cranial fossa. The cribriform area is particularly vulnerable to development of spontaneous leak because of the presence of mal-developments with extension of the sub-arachnoid space through the foramina of the cribriform plate [14].

Traumatic Cases with No Active Leak

Plain CT scan with MR Cisternography is the gold standard in diagnosis of suspect area of CSF leak. In traumatic cases with no active leak, but having history of meningitis or recurrent leaks CT scan with MR Cisternography is done in hope of finding suspected area of leak. In few patients no active leak is demonstrated. Ideally sodium fluorescein to be used to demonstrate the leak but due to lack of access to sterile medical grade sodium fluorescein, it is not routinely used. In the present study we have a high incidence of 9 cases of traumatic injury with no active leak but with history of meningitis or recurrent leaks, as these are referred cases (tertiary care centre) as it was undecided about their treatment. In these cases CT scan with MR cisternography was done. If it showed a suspected area of leak, then repair with select graft was done. In cases with no suspected area of CSF leak frontoethmoidectomy was done, followed by removal of middle and superior turbinate. Rule out frontal and sphenoid leak. Layered graft (3 grafts) was placed from below the frontal ostium to anterior wall of sphenoid (each layer overlapping the other layer from anterior to posterior).

The preferred modality of treatment of CSF rhinorrhea is transnasal endoscopic repair which provides better field of vision with enhanced illumination, angled visualization, accurate placement of graft, shortened operating time, minimizes intranasal trauma and preserves the bony framework supporting the frontal recess and other critical areas [15–18]. However, intracranial repair is indicated in complex anterior skull base fractures, nerve involvement and leaks involving posterior table of well pneumatized frontal sinus. Anosmia and intracranial infection are the most common complications of intracranial repair.

Medial Cribriform Leaks

Spontaneous CSF leaks are commonly found in medial cribriform area usually at the junction of middle and superior turbinates. These leaks are small and relatively easy to repair. We prefer to lateralise the middle and superior turbinates, expose the cribriform area, identify the leak, take off the surrounding mucosa and cauterize the small meningocele. If there is no active CSF leak we are happy to place a small fascia lata graft, buttress it with a gelfoam or fat and use tissue glue to secure it in place. A nasal pack would then be kept for 3–5 days. If there is an active leak we would use a plug of fat as a bath plug to close the dura defect prior to using the overlay fascia. We believe that amputating the middle and the superior turbinate is not required in most cases unless the defect extends to the lateral cribriform. If you do take the middle turbinate then one needs to do a good ethmoidectomy and frontal sinus surgery to prevent an inadvertent iatrogenic frontal blocking.

Lateral Cribriform Leaks

Lateral cribriform defects could be spontaneous or traumatic and may be small or large involving fovea ethmoidalis. To tackle these defects one must do a complete frontal and ethmoid surgery, clearly delineate the defect and then repair it depending on the size. The surrounding mucosa must be denuded off the bone for repair material to adhere and successfully close the leak. Small leaks are addressed similar to medial cribriform defects. Larger defects will require either cartilage or fat plug closure prior to an overlay fascia. For very large defects we use the vascularized pedicled nasoseptal flap. When using this Hadad flap in this area, it is usually required to take off the superior and the middle turbinates. These are secured in place with tissue glue and nasal packs are kept in place for 4–5 days.

Frontal Sinus Leaks

Frontal sinus leaks are often traumatic in origin and may involve the posterior table. With a well pneumatized frontal sinus, these sites are extremely difficult to access even with a DRAF 3 procedure. In these situations, we prefer a bicoronal route for repair, unless the patient insists only on an endoscopic approach. However, in some cases when the defect is closer to the frontal recess or if there is a hypoplastic frontal sinus, one may do the endoscopic repair more comfortably.

If the frontal sinus is hypoplastic one can even clear all the mucosa and obliterate the sinus along with the repair. If the sinus is well pneumatized then one must do the repair

and yet ensure adequate drainage for the frontal sinus. For a larger defect one may use a cartilage or fat as a plug with an overlay fascia, whereas a smaller defect may need a small fat plug or only overlay fascia.

Sphenoid Sinus leaks

Sphenoid sinus leaks are often traumatic in the planum and the lateral walls. We do see spontaneous leaks from meningoencephalocele in the lateral recess or Sternberg's canal. Rarely we may see spontaneous leaks from clivus itself.

Sphenoid sinus leaks are usually not difficult to repair and one can use a uni or binostril flap repair with a Hadad flap. As the openings are wide and the site of defect precisely repaired, one never needs to obliterate the sinus. Defects in the lateral recess of the sphenoid wall require a transpterygoid approach through the posterior wall of maxilla. As the internal maxillary artery and the sphenopalatine artery need to be cauterized for a transpterygoid approach one may have to use a flap from the opposite side for the repair. When the pneumatization of the lateral recess is significant these flaps are usually not long enough and a free repair is done using fat or cartilage with a fascia or mucoperiosteum from the septum.

With the recent developments in the surgical techniques, the middle and superior turbinates are preserved during the surgical repair of fractures involving medial cribriform and small defects of the lateral cribriform. The middle and superior turbinates are removed if fracture is extending from medial to lateral cribriform or if Hadad flap is used to repair large defects involving lateral cribriform.

Lumbar Drain

The use and need of lumbar drain has reduced over the years. Most CSF repairs nowadays don't need lumbar drain unless intracranial CSF pressures are high, in obese patients or there is reconstruction of large to very large defects. In these cases we like to keep the lumbar drain for 2–4 days.

The success rate of transnasal endoscopic repair in the present meta-analysis was 97.53%. This is comparable to a meta-analysis conducted by Hegazy et al. which had success rate of 90% in the first surgical repair attempt and another case series of 267 cases by Kirtane et al. [19] had success rate of 96.63%.

The various types of graft materials available for repair of the defect include:

1. Free grafts: autologous muscle, fat, nasal mucosa, fascia, lyophilized dura

2. Composite grafts: autologous chondro-mucoperichondrial and osteo-mucoperiosteal flap
3. Flaps: nasal mucosa advancement flaps
4. Reinforced nasal mucosa flaps: free cartilage, bone with osteo-mucoperiosteal or chondromucoperichondrial flaps

Autologous muscle was not used in repairs because in our experience, muscle would become fibrinous reticulum over time, and make the repair prone to late failure.

Out of the 243 patients treated surgically 77.77% were operated using free grafts and 22.22% by using flap repair. Free grafts had 99.47% (188/189) success rate and flap grafts 90.74% (49/54) success rate. The higher failure rate of flap repair could possibly be attributed to flaps being used in larger, more difficult to repair cases. Also, flap grafts being thicker than free grafts, there is difficulty to adjust the precise width of the flap grafts to the defect, flap grafts do not adhere to the contour of the defect easily and pedicled flap grafts may undergo contraction and eventual failure of flap grafts. Hence flap grafts are advised only in cases of large defects and multiple site fractures.

Correlation of the failure of the primary repair with the site of the leak, intraoperative procedure and the type of the repair. Primary transnasal endoscopic repair failure was most commonly associated with defects in the sella post transnasal pituitary surgery. This result is supported by the study conducted by Mayo clinic in which closures in sphenoid leaks failed most commonly, that is in 28.6% (12/42) of cases [20]. Flap grafts were used in both cases and it was further noted that 45° endoscope was not used during the procedure.

Bleeding was seen in 1.28% of the cases. Bleeding occurred from anterior ethmoidal artery, posterior ethmoidal artery and pharyngeal branch of 3rd part of maxillary artery. This can be avoided with careful scrutiny of preoperative CT scan. The anterior ethmoidal artery is commonly injured at the roof of the ethmoid and posterior ethmoidal artery in the posterior ethmoidal air cells and the upper part of the posterior nasal septum.

Conclusion

CSF rhinorrhea is a potentially life threatening condition and warrants an early diagnosis and intervention. The CT scan with or without cisternography, MRI and beta-2 transferrin are used to diagnose CSF rhinorrhea. The cases of active CSF leak need to be treated surgically preferably by transnasal endoscopic approach.

The commonest site of CSF leak was lamella of cribriform plate followed by roof of sphenoid sinus.

Careful pre-operative assessment of the CT-scan is necessary to know the bony anatomy, to locate the site and size of the defect, and to reduce the risk of intraoperative bleeding.

It is vital to demonstrate the site of bony defect and do a definite closure of the defect area after debridement of the surrounding mucosa. If this is done, irrespective of the type of repair material used one is likely to have a high success rate. In view of this it is our opinion that in the majority of CSF leaks it is preferable to use a conservative approach with preservation of turbinates and avoiding additional sinus surgery, unless required for that patient. The repair may be easily done using fascia or local mucoperiosteum with or without a fat plug. We would reserve the use of a vascularized pedicled nasoseptal flap only in those patients who have large defects, multiple defects or have failed a primary repair. This would help in decreasing nasal morbidity thus shortening the postoperative recovery and reducing the patient follow-up visits.

The use of various types of endoscopes (0°, 30°, 45° and 70°) improves the intraoperative visualization especially the frontal sinus and lateral recess of sphenoid sinus and hence the outcome of the surgeries.

The factors contributing to failure of the primary repair are the large size of defect, failure of localization of the defect, multiple sites of leak which were not recognized during the primary repair, the area of the bony defect on the skull base with dural dehiscence which opened upon repair of the area of initial CSF leak and the presence of comorbid conditions such as chronic cough and constipation.

The free grafts are preferred over flap repair. For moderate size defects free grafts with underlying cartilage or fat plug is used. For large and multiple defects, pedicled flap grafts with underlay cartilage or fat is recommended.

The use of lumbar drain is restricted to cases of high intracranial pressure, obese patients and in reconstruction of large defects.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interests.

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