Management of cerebrospinal-fluid-related intracranial abnormalities in frontoethmoidal encephalocele using “Shunt algorithm for frontoethmoidal encephalocele” (SAFE)

Muhammad Arifin Parenrengi (corresponding author), Wihasto Suryaningtyas

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Bukti korespondensi

Proses korespondensi	Tanggal	Keterangan
Submitted the manuscript	21 November 2023	page 1
Manuscript received	21 November 2023	page 2
Revised paper (1)	6 Januari 2024	page 3
Major revision from revised paper (1)	10 February 2024	page 5
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Article proof for online publish	5 Maret 2024	page 12
Published online	9 Maret 2024	page 17

Neurosurgical Review - Receipt of Manuscript 'Management ofCerebrospinal-Fluid-Related Intracranial...'

1 message

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Ref: Submission ID df98add2-ad8b-41fd-8093-e44466823c08

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REVIEWER REPORTS

Reviewer Comments:

Reviewer 1
Parenrengi et al. report a retrospective review of patients of children treated with frontoentmoidal encephaloceles and assessed the shunt insertion protocols of two different eras. 129 patients were included. The authors reported that early shunting resulted in a higher rate of complications. However, the policy with the other group was to shunt after complications in the moderate risk group, but these were not considered to be shunt complications since they occurred before shunting but after the principle surgery. The authors report a high tendency to shunt all CSF anomalies as recently as 2011. This was not the standard of care at my institution at the time and so it is difficult to judge the validity of this. It is our center’s preference to place a lumbar drain if there is concern that elevated intracranial pressure may increase the risk of CSF leak post-op and only then is a shunt considered unless a separate diagnosis of hydrocephalus has been made.

Some attempt at statistical analysis should be made if the authors wish to draw conclusions of the validity of their protocol. In particular, the 3 complications in group 2 should be compared to the 7 of group 1.

Reviewer 2

In this paper, the authors review 129 patients with fronto ethmoidal encephaloceles and develop an algorithm (termed SAFE) to determine which patients need a VP shunt. They looked at the period from 2007-2011 (before SAFE) and 2012-2019 (after SAFE), and found that after SAFE was instituted, the VP shunt rate went down. This was a very interesting paper, and they see significantly more patients with this pathology than neurosurgeons in the US/Europe. It is obvious that they have a lot of experience in treating these patients and I value their expertise in this field.

My comments:

1. They picked multiple factors to include in their algorithm, such as pulsatile mass, size of the bony defect, length of the exit pathway, etc. I would like more of a discussion regarding how and why they picked these factors. I assume it was based on their experience, but were there other reasons they picked these factors.
2. They should include a short paragraph regarding why FEE is more prevalent in southeast Asia compared to the rest of the world.
3. There are a number of grammatical errors that should be corrected in the text.
4. The text should be shortened and can be made more succinct, especially the discussion.

Overall, I applaud the authors with sharing their experience with this condition.

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Sat, Feb 10, 2024 at 04:48

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REVIEWER REPORTS

Reviewer Comments:

Reviewer 2
n/a

Reviewer 3
The authors present an interesting series of patients with frontoethemoidal encephaloceles (FEE) and their approach to managing “CSF-related abnormalities”. They introduce an algorithm used to guide decision making regarding timing of CSF-diversion surgery. Although retrospective, this is an interesting real-world quality improvement initiative that has shown to be useful and meaningful for their institution and patient population. I find this manuscript very interesting, mainly to share their experience given the higher incidence of this pathology in their region. It also illustrates

how rare conditions are managed differently in various institutions based on experience and resources.
The authors addressed the reviewers' comments with a first revision. After assessing their response and revision, I have additional comments.
1) A thorough review of English grammar/syntax is needed in the abstract and main manuscript.
2) Abstract:
a. Abstract background: the authors say "CSF diversion is the preferable measures (…)". This sentence is confusing. One can assume that the authors mean that the preferred measure to treat CSF-related anomalies seen in association with the encephalocele is CSF diversion. But the way it is written, it is unclear if they are saying that the preferred way to treat encephaloceles is with a CSF diversion. If they truly mean to say that they treat encephaloceles with CSF diversion, then I would specify that this is unique to their institution, as that will not be the standard approach around the world.
b. Abstract results: the authors use the abbreviation "FEEM". It has not been defined elsewhere.
3) Introduction:
a. Paragraph 3: The authors say "We categorize the abnormalities in two groups (…)" I would recommend specifying who represents the "we". For example, saying something like "At our institution, the surgeons categorize (…)" or "For the purpose of this study, the authors categorize (…)"
b. Paragraph 3: "Before 2012, almost all CSF-related abnormalities were shunted (…)" I would suggest specifying that you are providing local context to explain why this project came about. For example, "Before 2012, almost all CSF-related abnormalities were shunted at our institution, leaving only (…)"
c. Paragraph 4: same comment as above. Suggestion "Starting in January 2012, SAFE has been used by all surgeons at our institution treating children with FEE harboring CSF-related intracranial abnormality."
4) Method:
a. Paragraph 1: "(…) 3) the rate or ratio of shunted non-shunted cases; (…)" please rephrase, it is not clear. It should be only one of the two choices (rate or ratio), as both define different things (calculated differently).
b. Shunt algorithm for frontoethmoidal encephalocele (SAFE) section – paragraph 1: The authors mention table 1. But table 1 should be mentioned in the results section, and doesn't seem to make sense with the description of the SAFE score. Do they mean figure 1?
c. Shunt algorithm for frontoethmoidal encephalocele (SAFE) section – paragraph 1: "The shorter one determines the score in an FEE patient with multiple exit pathways." Flow is difficult to follow from the previous. Suggest "In a patient with FEE characterized by multiple exit pathways, this variable is scored based on the shortest measured pathway."
d. Shunt algorithm for frontoethmoidal encephalocele (SAFE) section – paragraph 1: "The CSF canal in the exit pathway is a cyst that extends beyond (…)". It is not clear to me which variable in the table (figure 1) they are referring to.
e. Shunt algorithm for frontoethmoidal encephalocele (SAFE) section – paragraph 1: What formula are they using to calculate the volume of the FEE?
5) Figure 1: the title of the first column has a typo "variabel"
6) Shunt algorithm for frontoethmoidal encephalocele (SAFE) section – paragraph 3: The authors refer to figure 2 in the first sentence. But this is an illustration and no a decision tree. Did they mean figure 3? If that's the case, seems like they have not made a reference to figure 2 anywhere else?
7) Figure 3: Although I understand that this algorithm focuses on deciding when to shunt or not. But they should clarify the timing of the FEE repair also.
a. Score /= 8 or any sign of neurological deficit related to the CSF-abnormality consider shunt insertion as first-line treatment observation vs repair of FEE?
8) Results:
a. Paragraph 1: the authors use the abbreviation "FEEM" here again (see comment #2b).
b. Paragraph 1: the patient sample meeting inclusion criteria totals 129 (FEE patients with CSF-related anomalies). Can the authors provide the total number of patients with FEE (without and with CSF-related anomalies)?
c. The authors could consider adding % to the results in places where it could be helpful to understand the proportion. For example, arachnoid cysts were seen in 46 cases (36%) …
d. Paragraph 4: the authors mention that they would shunt patients who had a persistent leak of more than 2 weeks. Is this standard practice at their institution to wait this long with a CSF leak? Do they give iv antibiotics while it leaks?
9) Discussion:
a. I think it would be interesting if the authors comment somewhere in the discussion that when applying the SAFE algorithm to the cohort prior to 2012, 19 patients could have possibly avoided getting a shunt. This is quite a significant impact as 7 of those 19 patients had pretty severe shunt-related complications.
b. The authors should add "et al." when citing authors in the manuscript.
c. Throughout the discussion, the authors alternate between citing published literature and commenting on their own institutional findings. It is sometimes unclear when they are referring to their findings versus published literature. One example, paragraph 2 of the discussion: "Intracranial structural anomalies accompany sincipital encephalocele in 15 to 40% of cases [1, 4, 6, 15, 16]. Sibayan reported in their series that the intracranial abnormalities rate was as high as 55% of sincipital encephalocele cases [32]. The most frequent findings were CSF-related intracranial abnormalities such as ventriculomegaly, arachnoid cyst, porencephalic cyst, and ventricular malformation. (…)" Is the last sentence regarding Sibayan et al.'s work, or regarding this study? Suggestion would be "Sibayan et al. reported intracranial abnormalities in 55% of sincipital encephaloceles with the most frequent being CSF-related intracranial abnormalities such as ventriculomegaly, arachnoid cyst, porencephalic cyst, and ventricular malformation [32]." Or "Sibayan et al. reported intracranial abnormalities in 55% of sincipital encephaloceles [32]. In our patient sample, the most common CSF-related intracranial abnormalities were ventriculomegaly, arachnoid cyst, porencephalic cyst, and ventricular malformation."
d. Paragraphs 3-6: The flow of the discussion is a bit hard to follow. I suspect the authors wanted to provide historical context and explain why they did what they did. I think they need to rework the discussion to improve clarity, having a careful review of the English language of this text might help to summarize and clearly articulate the point the authors are trying to make.
e. Either in the discussion or the methods, the authors should specify how many surgeons are involved in the care of patients with FEE. Is it a single-surgeon series, or multiple surgeons? Did all the surgeons accept and follow the SAFE?
f. Paragraph 11: "There is no apparent agreement between the results of the surgery (morphological changes) and the alleviation of symptoms [23, 35]." Do the two cited publications refer to surgery for arachnoid cysts only? Or surgery on the arachnoid cyst in patients with FEE? Or surgery to repair FEE in patients that have arachnoid cysts?
g. Paragraph 11: What do the authors mean "(…) developed well (…)"?
h. It was very interesting to read the authors explanation for their local circumstances that lead to high shunt complications. Where do they insert the proximal catheter in relationship to the FEE repair site?
i. Paragraph 15: The authors say "Several risk factors for shunt infection in our series included being undernourished, female, having a neural tube defect, and less than one year old." I don't think they can say "in our series" since they did not study these variables like nutrition status. They can maybe say something like… "Patients with FEE treated at our institution generally come from low-income families and a high-right of

malnutrition is seen in our pediatric patient population. Thus, we hypothesize that these local risk factors contributed to the shunt infections observed in this series."

10) Mention of ethics or IRB approval is missing?

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Journal: Neurosurgical Review.
DOI : 10.1007/s10143-024-02342-y.
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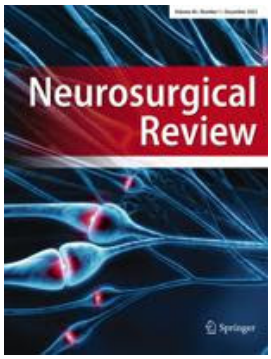

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Management of cerebrospinal-fluid-related intracranial abnormalities in frontoethmoidal encephalocele using "Shunt algorithm for frontoethmoidal encephalocele" (SAFE)

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Neurosurgical Review

Muhammad Arifin Parenrengi · Wihasto Suryaningtyas

A cerebrospinal-fluid-related (CSF-related) problem occurred in 25-30% of frontoethmoidal encephalocele (FEE) cases. Since there was no algorithm or guideline, the judgment to treat the CSF-related problem often relies upon the surgeon's experience. In our institution, the early shunt was preferable to treat the problem, but it added risks to the children. We developed an algorithm, "Shunt Algorithm for Frontoethmoidal Encephalocele" (SAFE), to guide the surgeon in making the most reasonable decision. To evaluate the SAFE's efficacy in reducing unnecessary early shunting for FEE with CSF-related intracranial abnormality. Medical records of FEE patients with CSF-related abnormalities treated from January 2007 to December 2019 were reviewed. The patients were divided into two groups: before the SAFE group as group 1 (2007 – 2011) and after the SAFE group as group 2 (2012 – 2019). We excluded FEE patients without CSF-related abnormalities. We compared the number of shunts and the complications between the two groups. One hundred and twenty-nine patient's medical records were reviewed. The males were predominating (79 versus 50 patients) with an average age of 58.2±7.1 months old (6 to 276 months old). Ventriculomegaly was found in 18 cases, arachnoid cysts in 46 cases, porencephalic cysts in 19 cases, and ventricular malformation in 46 cases. Group 1, with a score of 4 to 7 (19 cases), received an early shunt along with the FEE repair. Complications occurred in 7 patients of this group. Group 2, with a score of 4-7, received shunts only after the complication occurred in 3 cases (pseudomeningocele unresponsive with conservative treatment and re-operation in 2 cases; a sign of intracranial hypertension in 1 case). No complication occurred in this group. Groups 1 and 2, with scores of 8 or higher (6 and 8 cases, respectively), underwent direct shunt, with one complication (exposed shunt) in each group. The SAFE decision algorithm for FEE with CSF-related intracranial abnormalities has proven effective in reducing unnecessary shunting and the rate of shunt complications.

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