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ORIGINAL ARTICLE

Treating nasoethmoidal encephalocele in a low-resource country: a surgical experience from a Philippine multidisciplinary craniofacial team

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Abstract

Purpose It was described that nasoethmoidal encephalocele repair in the Philippines has been limited by insufficient resources, financial constraints, and a lack of surgical expertise. The purpose of this study was to report initial results and complications of Philippine patients with nasoethmoidal encephalocele surgically managed with an approach adapted to an environment with limited financial resources.

Methods All patients ($n = 21$) with nasoethmoidal encephalocele who underwent intracranial and extracranial repairs (frontal wedge osteotomy to access the encephalocele cyst and cranial base defect, dural defect repair, split frontal grafts fixed with polydioxanone sutures to reconstruct the cranial defect and nasal dorsum, and medial canthopexy) from January 2015 to May 2017 were included. The correlations between sizes of masses and cranial defects with the occurrence of complications were tested. The surgical results were classified based on a previously published outcome grading scales I–IV on the need for additional surgery.

Results Nineteen patients (90.5%) had unremarkable post-operative course. Two patients (9.5%) presented with complications (cerebrospinal fluid leak and surgical site infection) which were successfully managed with no additional surgery. The sizes of masses and cranial defects were not correlated ($p > 0.05$) with complications. The overall rate of surgical results ranked according to the need for additional surgery was 2.4 ± 0.5 (between categories II and III).

Conclusions We reported successful surgical repair of nasoethmoidal encephaloceles in Philippine patients by a local multidisciplinary craniofacial team.

Keywords Nasoethmoidal encephaloceles · Surgery · Complications

Introduction

Frontoethmoidal encephaloceles, a congenital disorder characterized by structural changes to the junction of frontal and ethmoid bones internally and the frontonaso-orbital region externally, results in facial disfigurement [1]. Suwanwela and Suwanwela classified frontoethmoidal encephaloceles into nasofrontal, nasoorbital, and nasoethmoidal types [2]. In Asiatic countries, the nasoethmoidal type is the most common [3], with the highest incidence in Southeast Asia (1 in every 3500 births) [4].

Through the years, several surgical approaches, modifications, and refinements have been proposed for frontoethmoidal encephalocele repair [3, 5–20]. In 1991, Mahatumarat et al. described an exclusive extracranial approach, the Chula technique, where a full-frontal craniotomy was avoided and the cranial defect was assessed and repaired

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by a medial orbital composite-unit osteotomy, namely a T-shaped osteotomy over the superomedial orbits bilaterally [19]. Since then, several groups have adopted the Chula technique, and some technical modifications have also been described, including one-stage or two-stage repair for correction of hard and soft tissue defects (i.e., excision of gliomatous encephalocele mass, reconstruction of cranial bone defect, mobilization of the medial orbital walls, repositioning of the medial canthi, and augmentation of the nasal dorsum) and different alternatives of fixing the reconstructed segments (e.g., titanium plates and screws and interosseous wiring with stainless steel wires) [3, 5–18]. The multitude of techniques and approaches is testament to the fact that no treatment consensus and uniformity has been established regarding the ideal protocol including the optimal timing for the procedures and/or type of surgery [3, 5–20].

In this context, a previous report [21] described that repair of frontoethmoidal encephaloceles (including nasoethmoidal type) in the Philippines has been limited by insufficient resources, financial constraints, and a lack of surgical expertise. In the last years, we have managed nasoethmoidal encephaloceles with a combined nasal-coronal approach with some different technical details based on experiential learning of a center that act in an environment with limited financial resources. Instead of adopting the classical T-shaped osteotomy, medialization of orbital bone segments, and fixation with wires and/or titanium plates and screws, we have adopted a wedge frontal craniotomy, with no mobilization of the orbital walls, and fixation of bone grafts with absorbable polydioxanone (PDS) 2-0 suture. Therefore, the purpose of this study was to report initial results and complications of Philippine patients with nasoethmoidal encephalocele surgically managed with this approach.

Methods

An observational prospective study was conducted for all consecutive patients with nasoethmoidal encephalocele operated at the Philippine General Hospital between January of 2015 and May 2017. Only patients with confirmed diagnosis of nasoethmoidal encephalocele [1] by the multidisciplinary team (neurosurgeons, plastic surgeons, and pediatricians), who underwent a combination of a transcranial and a direct anterior approach with some modified technical details by the same surgical team, and with full postoperative follow-up data, were included. Patients who had Chiari malformation, seizure disorders, congenital heart defects, previous reconstructive procedures, incomplete medical records and/or incomplete postoperative follow-up visits (< 12 months) were excluded from the study.

This study was approved by the Institutional Review Board and complied with the declaration of Helsinki. All patients or their parents signed an informed consent.

Surgical approach

Besides pre-operative full clinical evaluation, computed tomography scans were adopted for surgical planning, including assessment and localization of bony defects (Fig. 1) and associated anomalies. Based on previous standardization [12, 22], medial and lateral orbital measurements were performed to define the presence of true hypertelorism of the entire orbital complex. The size of extracranial mass was distributed in small, moderate, or large based on previous classification [23]. The sizes of the bone defects were measured intraoperatively with calipers.

All operations were performed with the patient in supine position with a neutral neck position. For the intracranial step (Figs. 2 and 3), a zigzag coronal incision was adopted to gain subperiosteal exposure from 2 cm above the supraorbital bar and to the superior orbits and frontonasal midline structures. A wedge frontal osteotomy was performed for intracranial exposure, with osteotomy size ranging according to the cranial defect. Both the superior and medial orbital walls were preserved. The encephalocele stalk was disconnected from the rest of the intracranial contents. When the dural defect was not amenable to primary watertight closure, a pericranial graft was used to cover the defect. The bone defect was then sealed with split frontal bone grafts and fixed in position with PDS 2-0 sutures (Ethicon Inc., Somerville, NJ). The nasal dorsum (only the bony pyramid) was also reconstructed with this split graft. The pericranial flap was adopted to cover the grafted split bone, a closed suction drainage was placed, and the coronal incision was closed.

For the extracranial step (Figs. 4, 5, 6; see electronic supplementary material), the disconnected encephalocele cyst

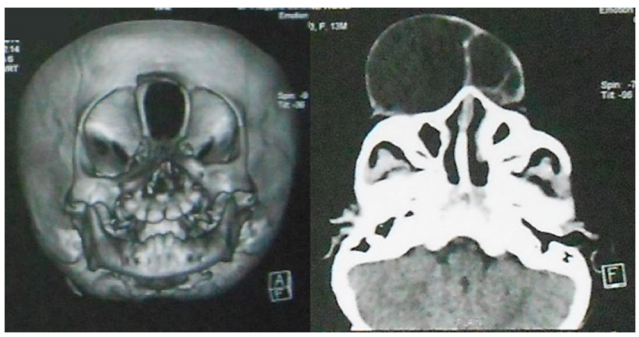

Fig. 1 Craniofacial computed tomography imaging of a patient with nasoethmoidal encephalocele. Note the discrepancy between the sizes of the bone defect and soft tissue mass. This particular encephalocele type presents a long and oblique course from the frontoethmoidal junction to exit at the junction of the nasal bones and the nasal cartilages, with the nasal bones forming the upper border of the external mass

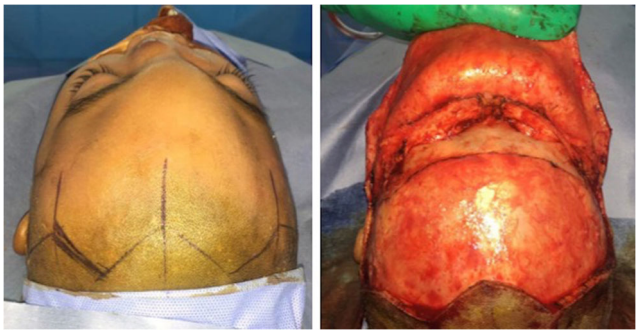

Fig. 2 Coronal approach (wavy-line pattern) in a pediatric patient with nasoethmoidal encephalocele

stalk was addressed by a naso-glabellar cutaneous incision. The extracranial glial mass was carefully dissected and excised as in "cut-as-you-go" principle, preserving tissue that will act as a nasal support as it heals and contracts. The medial canthal tendons were isolated and fixed to the medial suborbital region with prolene 4-0 sutures. If the medial orbital walls were compromised, the medial canthal tendons were fixed by transnasal suturing (prolene 4-0) with no bone support. Excess cutaneous tissue was then carefully excised to reshape the nasal dorsum and to allow for layered tension-free closure.

Postoperative care

Postoperative care was routinely performed in pediatric wards, and admission to the pediatric intensive care unit was considered on an individual basis. Patients were discharged if the post-operative course was unremarkable. The drain was removed when the output was less than 20 mL for 2 consecutive days. Close follow-up was done up to 1 month (weekly visits or according to the needs) post-operatively. The patients were then followed up quarterly and then semiannual clinical appointments.

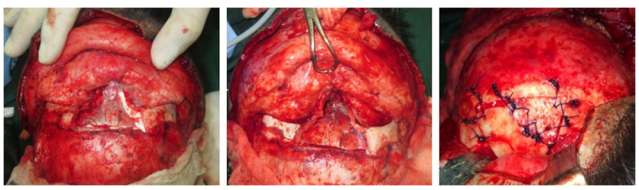

Fig. 3 (Left) Wedge frontal craniotomy with exposure of the encephalocele stalk. Note: the preservation of the orbital bandeau and the medial orbital wall. (Center) Closure of the dural defect after discconection of the encephalocele stalk from the rest of the intracranial contents. (Right) Cranial defect reconstructed with split frontal grafts fixed with polydioxanone 2-0 sutures. The osteotomized frontal bone was split into two pieces with a thin sharp osteotome and contoured with a Tessier bender. Note: the bone graft had a caudal extension adopted to reconstruct the bone nasal dorsum

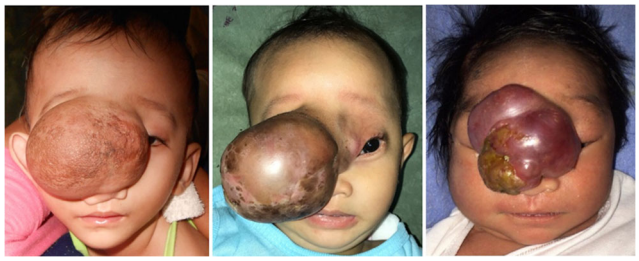

Fig. 4 Patients with nasoethmoidal encephalocele demonstrating the wide spectrum of clinical presentation regarding the cutaneous covering the mass. The planning of the cutaneous incisions to approach the extracranial glial mass and subsequent trimming of the cutaneous excesses was based on the use of the area with the best cutaneous quality within the limits of the nasal subunits (if possible)

Outcomes assessment

Bone correction was measured as residual cranial base defects [8]. The need for additional surgery (the Whittaker classification) [8] was assessed by three plastic surgeons in a blinded fashion (no prior knowledge about the patients and did not participate in taking the photographs or surgical procedures) based on two-dimensional facial photographs: category I, no refinements or surgical revisions considered advisable or necessary; category II, soft tissue or lesser bone contouring revision were advisable; category III, required a considerable

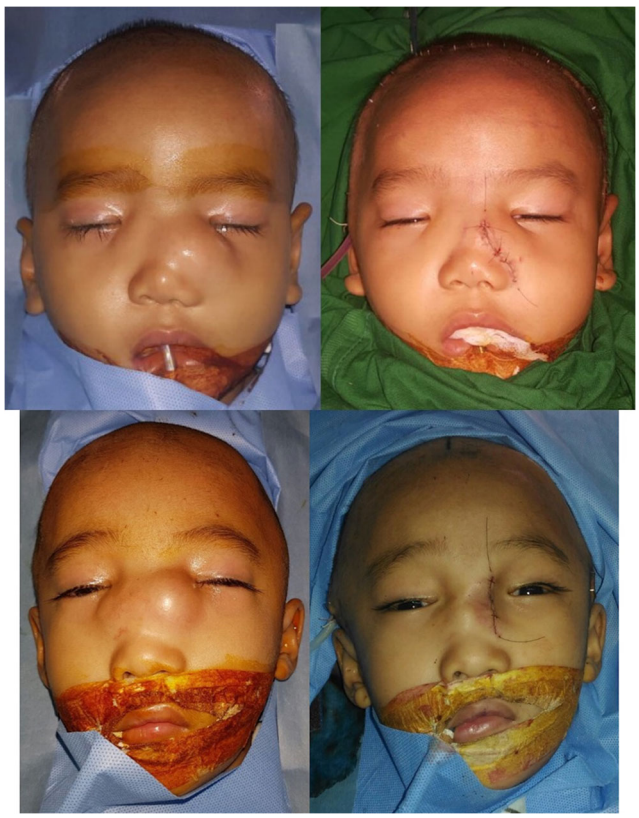

Fig. 5 Patients before and after nasoethmoidal encephalocele repair. The encephalocele cyst stalk was dissected from the skin, but some extracranial glial mass was maintained as a nasal support

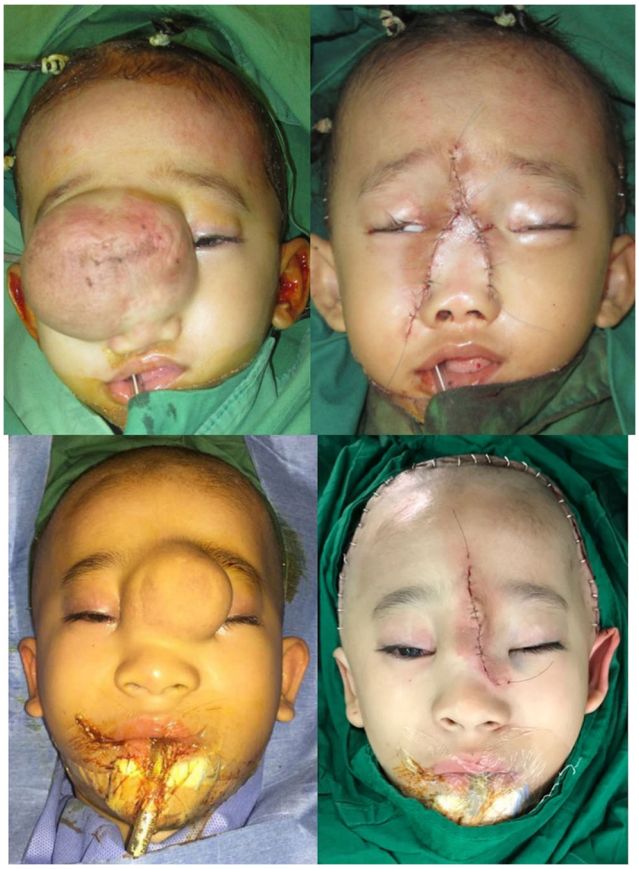

Fig. 6 Patients before and after nasoethmoidal encephalocele repair

amount of additional surgeries (procedures not as extensive as the original procedure); and category IV, major craniofacial procedure advisable, duplicating or exceeding the original procedure. An open final question allowed additional comments.

Surgical-related complications, including excessive bleeding, wound infection, meningitis, cerebrospinal fluid leakage, wound dehiscence, postoperative seizures, and pneumonia, were recorded. Revisionary surgery or subsequent procedures related to complications were also documented.

Statistical analysis

For the descriptive analysis, the mean was used for metric variables and percentages were given for categorical variables. A binomial logistical regression [24, 25] was performed to determine a possible association between one or more continuous independent variables (areas of the mass and bone defect) and a binomial, nominal dependent variable (whether the patient will have a complication or not). Removing outliers (± 2 standard deviation) for both variables was a prudent measure to avoid biases in the generated model. Kolmogorov–Smirnova, Hosmer and Lemeshow, and pseudo-r^2 (for logistic regression model) tests were adopted for analysis [24, 25]. The degree of interobserver agreement was assessed using the Fleiss k values. All analyses were performed with the Statistical Package for Social Science (SPSS version 20.0 for Windows, Chicago, IL, USA). Values were considered significant for a 95% confidence interval ($p < 0.05$).

Results

A total of 21 patients with nasoethmoidal encephalocele met the inclusion criteria. Most patients presented with moderate to large masses (95.3%) and telecanthus (85.7%). They had no true hypertelorism (Table 1). All patients underwent intracranial and extracranial surgical repair of nasoethmoidal encephaloceles, with an average time of surgery of 3 h and average blood loss of 100 ml (Fig. 7; see electronic supplementary material). Average length of hospital stay was 3 days, and no postoperative admission to the pediatric intensive care unit was necessary. Most patients (90.5%) had an unremarkable post-operative course, whereas two patients (9.5%) presented with complications (Table 1). One patient developed cerebrospinal fluid leakage post-operatively which was successfully managed with a lumbar drain. Another patient developed wound infection, which resolved with antibiotics and wound care. There were no reoperations performed

Table 1 Patients with nasoethmoidal encephaloceles surgically managed in our center ($n = 21$)

Characteristics	Patients
Age at surgery, M±SD (years)	3.5 ± 2.3
Gender, *n* (%)	
Female/male	11 (52.4)/10 (47.6)
Pre-operative telecanthus, *n* (%)	
Yes/no	18 (85.7)/3 (14.3)
Pre-operative true hypertelorbitism, *n* (%)	
Yes/no	0 (0)/21 (100)
Pre-operative shunting *, *n* (%)	
Yes/no	4 (19)/17 (81)
Size of mass **, M±SD (cm/cm)	5.2 ± 2.6/4.8 ± 2.3
Small, *n* (%)	1 (4.7)
Moderate, *n* (%)	17 (81)
Large, *n* (%)	3 (14.3)
Size of bone defect **, M±SD (cm/cm)	1.9 ± 0.6/1.8 ± 0.8
Complications, *n* (%)	
Yes***/no	2 (9.5)/19 (90.5)
Cerebrospinal fluid leakage/wound infection	1 (50)/1 (50)
Need for additional surgery****, M±SD	2.4 ± 0.5
Category II/III, *n* (%)	12 (57.1)/9 (42.9)

N, number of patients; M, mean; SD, standard deviation; cm, centimeters; *, this was performed weeks or months before the encephalocele repair; **, size was defined as the longest mass/defect diameter and its longest perpendicular diameter; ***, 2 patients with size of mass classified as moderate; ****, no category I or IV in this sample

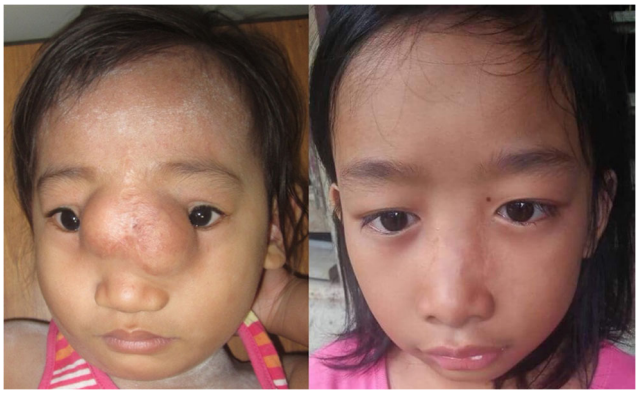

Fig. 7 Patients before and after nasoethmoidal encephalocele repair

for complications. No residual bone defect was detected during follow-up. No further surgical-related complications were noted with an average follow-up of 22 months.

For correlation between size of the mass and bone defect and post-operative complications, the Hosmer-Lemeshow statistics ($H = 6.394$; $p = 0.603$) indicates that the model fits the data provided. The model explained 17.3% (Nagelkerke R^2) the variance in post-operative complications and accurately classified 90.5% of the included patients. However, none of the individual variables significantly determine the tested outcome ($p = 0.940$ and $p = 0.162$ for sizes of mass and bone defect, respectively). Prognosis of complications of nasoethmoidal encephalocele patients were modeled through the binomial logistic regression equation: $y = -0.123x_1 + 0.130x_2 - 0.899$, where x_1 and x_2 represent areas of mass and bone defect, respectively. A y value greater than or equal to 1 means a patient is most likely to sustain a complication post-surgery.

Surgical results ranked according to the need for additional surgery was 2.4 ± 0.5 (between categories II and III; Table 1), with variations according to the spectrum of soft tissue involvement (e.g., the most common complaint between the raters was related to residual soft tissue on the lateral nasal areas; and the increased inner intercanthal distance was also commented by some raters). There was no category IV. Interobserver agreement was almost perfect ($k = 0.879$). No revisionary operations have been performed to date. All patients continue to undergo follow-up at our center.

Discussion

For nasoethmoidal encephalocele reconstruction, there are overall key principles: all nonfunctioning extracranial brain tissue herniating through the skull base should be debrided and/or reduced intracranially; the dural defect should be repaired to provide a watertight closure around the viable cerebral contents; the skull base defect should be repaired to prevent future herniation and encephalocele recurrence; and craniofacial, nasal, and medial canthal reconstruction should be performed to restore the appropriate craniofacial anatomy [3, 5–20]. Interestingly, there is no doubt in the fact that the first surgery has to include the correction of the encephaloceles and several approaches have been described to achieve these described principles [3, 5–20], but the timing and extent of surgical repair of facial bony deformities remain as a point of debate among craniofacial teams, with no evidence-based results to aid in decision-making [3, 5–20, 22]. Our patients have been managed by a therapeutic approach that encompasses all these key principles, although we did not have to perform an aggressive orbital-nasal reconstruction.

For our osteotomy planning, the frontal craniotomy size has been variable according to the identified bone defect size. We are in favor of performing a vertically broader frontal craniotomy than to include the superior-medial orbital bandeau. This planning has allowed reconstruction of the entire skull base and nasal bone defects by means of split frontal bone grafts, with no increase in surgical time. While the neurosurgical team is managing the encephalocele stalk and dural defect, the plastic surgery team splits and molds the frontal bone grafts for later skull base and nasal reconstructions. Other groups [26, 27] have also described their experience with splitting cranial bone in children with craniofacial deformities as young as 2 months.

Instead of adopting the classic bone fixation with stainless steel wires and/or absorbable or nonabsorbable microfixation plates [3, 5–20, 28–30], all split frontal bone grafts were fixed with PDS sutures. As an absorbable material, PDS has been used successfully in orbital, nose, skull base, and cranial vault reconstructions, including a long-term follow-up of pediatric patients with craniosynostosis [28, 31, 32]. No local irritation or additional morbidity related to PSD sutures were reported in our and previous pediatric craniofacial surgical reconstructions [28, 32]. We also had no problems with postoperative bone migration into undesirable positions. As showed in craniosynostosis reconstruction reports [28, 32], a series of properly distributed holes allows for the stable fixation of bone grafts with PDS sutures. Although this is not a comparative or cost-effectiveness study, the adoption of PDS sutures has reduced our overall costs [32] and reduced patient waiting time for surgery. The PDS material is available in our health system and patients can be operated after diagnosis, while absorbable plaques or meshes are not covered by our national health system and its use depends on a long acquisition process based on a system of price quotation and bidding. Other groups [8, 21, 33] have treated Philippine patients with classical surgical materials, but they used external resources (i.e., medical team and surgical materials from other countries). We demonstrated the feasibility of our current approach by only adopting resources from our national health system. Additionally, our current surgical management reduced the

necessity of unnecessary surgical intervention to remove wires or titanium plates during follow-up, as it was a concern (palpable or extrusion in the nasofrontal area) in our center previously. Future comparative studies should test our hypothesis and also compare the adopted fixation method with other potential materials such as nonabsorbable nylon suture adopted for bone fixation in craniosynostosis repair [34].

Regarding the nose reconstruction, other groups have adopted a long bone graft to reconstruct the entire nasal dorsum (the bone pyramid and the cartilaginous vault), from the radix to the nasal tip [3, 5–20]. We only reconstructed the bone pyramid by using split bone grafting connected to the cranial bone grafting. We also resected part of the encephalocele cyst stalk as the scarred and fibrous soft tissue can provide further nose support during the healing process. As this approach has been subjectively based on the "cut as you go" principle, soft tissue excess may be evident after edema resolution and scar maturation. However, we prefer a future nose bulging than a nasal depression which is more difficult to reconstruct. We can perform a secondary nasal surgery when patients reach skeletal maturity by reshaping the soft tissues associated (or not) with further dorsal nose augmentation by rib or dermal fat grafting.

For orbital bone reconstruction, those advocating concomitant orbital and encephalocele repair believe that it allows the expanding brain and face to model the developing skeleton and diminish the defect [3, 5–21, 35–37]. However, there are some arguments against this concomitant correction, including the extent of blood loss and surgical time and the potential facial growth disturbance and the relapse of orbital deformity [9, 13, 22]. Furthermore, as Philippine patients with frontoethmoidal encephalocele had lateral bony and soft tissue widening within normative averages [22], it was previously questioned the necessity of orbital bone osteotomies as an essential step for their surgical management [22]. Therefore, as other groups [22], instead of the orbital bone osteotomy and mobilization, we only performed a medial canthopexy. It logically reduced intraoperative bleeding, time on the operating table, and as a consequence the length of hospital stay. In developing countries, intraoperative blood loss (10–300 mL), surgical time (2–7 h), and length of hospital stay (4–20 days) data have been variables according to types of encephalocele and surgical approach [14, 21, 35–38]. Our patients have been closely monitored in pediatric wards, with no need for intensive therapy. Even with the availability of pediatric intensive care unit, this approach has increased the turnover of patients in our hospital and also reduced the overall costs of hospitalization. The successful use of intensive care unit according to the individual needs has been demonstrated in other craniofacial surgical experiences [39].

As there is no standardization for the assessment of outcomes in nasoethmoidal encephalocele repair, different studies [3, 5–21] have diversely stratified the results. Similar to the UCLA group [8], we adopted the Whittaker score. The UCLA group [8] presented satisfactory outcomes (categories I and II) treating patients by a complete surgical approach, including lower supraorbital bar repositioning, medialization of medial orbital walls, medial canthopexy, and nasal dorsum augmentation. Not surprisingly, our patients were classified as categories II and III, reveling that further surgeries may be necessary during follow-up. These patients are being regularly monitored and complementary surgical interventions will be performed according to individual needs after craniofacial skeletal growth finalization. As our sample did not have patient with true hypertelorism, a less invasive correction (medial canthoplasty and/or medial orbital walls mobilization by an extracranial approach based on the clinical findings of each particular patient) can be performed than a more invasive hypertelorism correction. This rationale is not new, as several groups have treated their patients in multiple stages [3, 5–22, 35–38]. The impact of our approach at skeletal maturity should be the target of future investigations, including developmental tests for neurologic outcomes.

In this study, we carefully evaluated the complications and detected two surgical-related complications, namely cerebrospinal fluid leakage and wound infection. The complication rate has been variable according to the included samples [3, 5–22, 35–38], and our overall rate is in accordance with the lowest published rates [3, 5–22, 35–38]. In developing countries, variable rates of complications were previously reported, including wound dehiscence (0–1.75%), cerebrospinal fluid leak (0–18%), skin wound infections (0–9%), osteomyelitis (0–2%), meningitis (0–3%), signs of increased intracranial pressure (0–7.3%), and operative mortality (0–3.3%) [5, 14, 15, 21, 35–38]. We demonstrated no correlation of mass size and bone defect size with the presence of complications, as our two complications occurred in masses with moderate size, whereas outliers for mass size (i.e., > 30 cm^2) and the largest bony defect (12.25 cm^2) did not present with any complication post-surgery. Based on our data, the prognostic model had a sensitivity of 90%. However, as we have only two complications, a larger sample size is necessary to further strengthen the predictive value of this equation.

Besides the limitations detailed above, we reported successful surgical repair (primarily encephalocele and cranial defect corrections) of nasoethmoidal encephaloceles in Philippine patients by a local multidisciplinary craniofacial team. We believe that this approach can be applied and adopted in environments with similar resources [21, 33, 35–38, 40–42] and further research may confront or ratify our data and also try to solve our limitations.

Compliance with ethical standards

Conflict of interest None

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