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ORIGINAL PAPER

Frontoethmoidal encephalocele: clinical presentation, diagnosis, treatment, and complications in 400 cases

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Abstract

Purpose The purpose of this study is to review a large series of frontoethmoidal encephalocele (FEE) regarding their clinical presentation, the progressiveness of the mass volume, the skin stigmata as well as its surgical approach and post-surgical complications.

Method Records of all FEE patients treated in Soetomo General Hospital, Surabaya, and Charity Foundation Program from 2008 to 2015 were reviewed. Detailed patient's demography, clinical findings, radiology results, operative procedures, and complications were documented. Follow-up was organized in weekly basis for the first 1 month after surgery or more often when situation or complication occurred. Wound healing, neurological assessment for new or progressive deficit, pseudomeningocele, skin breakdown, cerebrospinal fluid (CSF) leakage, exposed implant, recurrent mass, and cosmetic results were documented. Since most of the patients had no direct phone line at their hometown, we relied on social worker to contact them.

Results One-stage surgery was performed for 400 patients with FEE (212 were male and 188 were female). Of 400 patients, 388 (97%) were younger than 18 years old. Most FEEs were nasoethmoidal, either isolated or combined with nasoorbital type (347 cases [86.75%]); nasofrontal subtypes were seen in 34 cases (8.5%) and nasoorbital in 14 cases (1.5%). The mean operative time was 2 h (range 30 min–3 h). There were only two patients (0.5%) needed postoperative blood transfusions. Mean hospitalization time was 5 days (range 4–7 days). Overall, complication rate in our series was 12.5%, mostly was CSF leakage and wound dehiscence.

Conclusion The current socioeconomic conditions and local facility should be considered to treat these specific disease processes. The refined and meticulous technique, especially in choosing the approach and handling the dural closure, is essential in lowering the complication rate.

Keywords Neural tube defect · Pediatric neurosurgery · Developing country · Surgical technique

Introduction

Encephaloceles are considered as a family of neural tube defect characterized by a herniation of the brain and meninges through anatomical bony defect of the skull [1–5]. Suwanwela and Suwanwela classified encephalocele based on the location and type of skull defect as occipital encephalocele, encephalocele of the cranial vault, frontoethmoidal encephalocele, and basal encephaloceles. In frontoethmoidal encephalocele (FEE), the internal skull defect is located in the midline, but the external skull defect may vary in the facial bony structure. It is further divided into three subtypes: nasofrontal (NF), nasoethmoidal (NE), and nasoorbital (NO) [6]. Mahatumarat added the fourth subtype: combined nasoethmoidal and nasoorbital [7]. Abundant cases of frontoethmoidal encephaloceles have been found in Southeast Asian countries such as Burma, Cambodia, Thailand, Malaysia, Indonesia, and India, with very rare cases reported in Europe, North America, and the Middle East [1, 4, 5, 8–14]. The reasonable explanation for this particular geographical distribution is unknown.

Several literatures pertinent to frontoethmoidal encephaloceles had been published. There have been few reports from a large series of FEE regarding their clinical

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presentation, the progressiveness of the mass volume as well as the skin stigmata. The various aspects of frontoethmoidal encephaloceles clinical presentations were reviewed as well as its choice of surgical approach based on the clinical findings.

Method

Records of all FEE patients treated surgically in Soetomo General Hospital, Surabaya, and Charity Foundation Program from 2008 to 2015 were reviewed. Detailed patient's demography, clinical findings, radiological results, operative procedures, and complications were documented.

Computed tomography (CT) scanning, pre-operative blood test and chest x-ray were performed routinely as part of pre-surgical preparation in all patients. Magnetic resonance imaging (MRI) was not performed routinely for a financial reason and it gives no additional critical information to plan a surgical approach. All patients underwent CT scan as a routine pre-operative imaging study. No MRI was performed during study period.

Surgical technique

Three surgical techniques were employed in our institution: Chula technique [7], modified Chula technique with no facial incision [15], and extracranial technique. Facial incision (Chula technique) was indicated on (1) a large mass that needed skin reduction, (2) a lesion with facial scar from previous surgery (in recurrence case), and (3) presentation of an ectopic mass or skin stigmata. The extracranial technique involved direct nasal incision or bicoronal incision based on its skin appearance and no nasal repair or bony grafting to close the external defect. Extracranial technique was indicated to a small mass volume, very small external bone defect (less than 7.5 mm), and no requirement for nasal repair. All surgery was performed under general anesthesia. Hair shaving only preserved for the modified Chula technique without facial incision. Operative field were prepared with povidone-iodine and draped. Lidocaine with adrenaline in concentration of 1:100.000 was infiltrated along the incision line.

The incision line in Chula technique may vary from lazy S, inverted Y, to T incision (Fig. 1). The plane between the normal skin and the encephalocele was identified. The mass was then dissected free of the covering skin and the bony rim of the defect was exposed carefully without injuring the nasolacrimal duct. Periosteal incision approximately 1 cm from the rim of the external bony defect was done to develop a periosteal flap. The subfrontal osteotomy using a chisel was performed to remove the deformed nasal bone. The osteotomy lines started from few millimeters medial of supraorbital foramen and extended 1 cm upward from superior orbital rim. Bilateral supraorbital cut was then connected to each other through horizontal osteotomy. The cut was continued along the medial part of the orbital wall on both sides until it reached the external bony defect. The final result of the bone cut was a T-shaped bone (Fig. 2a). Formal frontal craniotomy is not necessary. The subfrontal T-shaped bone was then detached from its dural attachment to expose the neck and the herniated sac. The encephaloceles usually had a thinner and more irregular overlying dura than the normal dura. With smaller encephaloceles, the sac and abnormal dura can be invaginated and sutured. With larger encephaloceles, excision of the dural sac along with removal of its non-functional neuroglial tissues followed by a watertight closure was recommended. Double breasting the dura or using a graft of temporal fascia or periosteum can often achieve a good seal.

To address telecanthus, we cut the T-shaped bone into three parts (Fig. 2b). We took out the central portion (part 3) of the T-shaped bone so that the medial orbital walls (part 1 and 2)

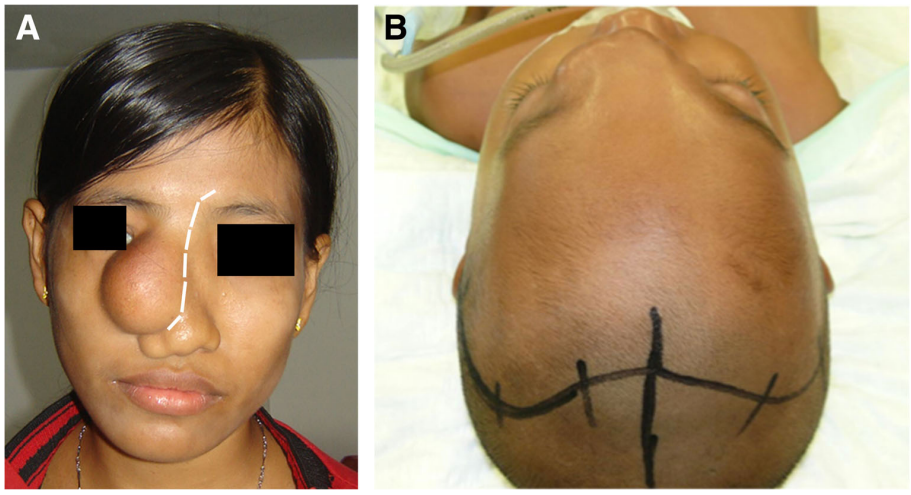

Fig. 1 **a** Lazy S skin incision planning for transfacial approach. **b** Bicoronal incision planning for modified Chula technique without facial skin incision

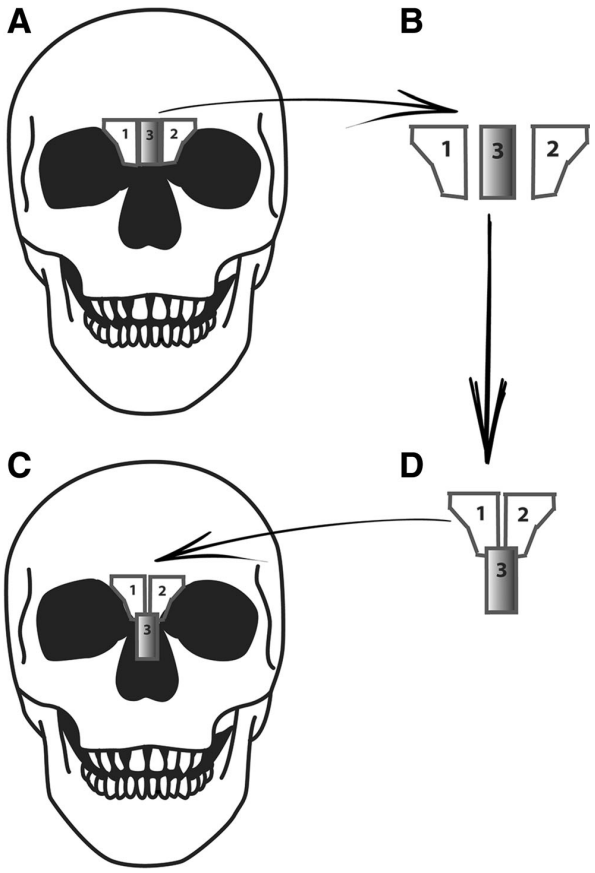

Fig. 2 Subfrontal osteotomy and bony reconstruction applied to the medial portion of the superior orbital rim, upper medial orbital walls, and nasal bones. **a**–**b** The bone was divided into three part (right medial orbit: *1*; left medial orbit: *2*; central part: *3*). **c**–**d** The cut bones were reconstructed. The central part was removed and is used for nasal augmentation

could be moved medially to recreate a new appropriate medial interorbital distance (see Fig. 2c). The bone defect is usually eliminated with this technique. To close a large external bone defect and to reconstruct the nasal bridge, we used the cut bone (part 3) from previous T-bone or a costochondral bone graft (Fig. 2c).

After completion of the bony reconstruction, the medial canthal ligaments are isolated and looped. Standard medial canthopexy was performed. Frequently, the ligaments have to be detached from its attachments before being fixed to the reconstructed T-shaped bone or the new nasal bone with monofilament 2–0 sutures. Care should be taken while releasing the periorbita from the orbit and upper anterior cheek from the inferior orbital rim as well as protecting the lacrimal apparatus. This is essential to achieve successful canthopexy with minimal tension.

In the past, our institution used small wire to fix the bones. However, once miniplate fixation widely available with lower price, we used it with good results. On skin closure, the degenerated skin should be reduced as completely as possible without jeopardizing the result. If the final nasal covering was considered to be too thin and the underlying bones and wires would be exposed, the deepithelized redundant skin was used as a dermal flap.

Modified Chula technique used bicoronal incision. The coronal incision stopped at the subcutaneous plane. The dissection continued subcutaneously under the posterior edge of the incision and left the temporal muscle fascia intact. The dissection continued anteriorly in the subgaleal plane as in the coronal dissection. The skin was flapped toward the superior orbital rim and left the periosteum intact. Once the dissection arrived at the nasofrontal junction, the plane between encephalocele dura and skin covering should be demarcated. The first dissection was to detach the mass from its skin attachment using a blunt-tip scissors. The later detachment included the attachment of the mass to its underlying bone and nasal cartilage. The periosteal incision was limited to the width of subfrontal osteotomy performed later. Subfrontal osteotomy was then carried out with the same technique as above. The rest of the step was similar to the previous technique except for the skin closure that did not involve skin reduction or modification. We never made facial incision to access the lesion when using this technique.

Follow-up

Follow-up was organized in weekly basis for the first 1 month after surgery or more often when situation or complication occurred. Wound healing, neurological assessment for new or progressive deficit, pseudomeningocele, skin breakdown, cerebrospinal fluid (CSF) leakage, exposed implant, recurrent mass, and cosmetic results were documented. Since most of the patients had no direct phone line at their hometown, we relied on social worker to contact them. Another limitation for follow-up was the financial problem.

Results

Surgical treatment was performed for 400 patients with FEE in our institution between 2008 and 2015. Of the 400 patients that underwent surgery, 212 were male and 188 were female (male/female ratio 1.13:1); 388 (97%) were younger than 18 years old (312 were 0–6 years old, 38, 7–12 years old, and 38, 13–18 years old) and 12 were older than 18 years. The mean duration of follow-up was 3 months (range 1–6 months).

Socioeconomic status

All patients in our series came from poor family, with average daily income of less than Indonesian Rp. 30.000

(approximately US $2). The parents' occupation was rice farmers in 90% of cases. Others were industrial or construction workers and traders. Parent's educational degree was elementary school in 60% of cases, high school in 20%, and the rest has no degree.

Classification of FEE

Most FEEs were nasoethmoidal, either isolated or combined with nasoorbital type (347 cases [86.75%]); nasofrontal subtypes were seen in 34 cases (8.5%) and nasoorbital in 14 cases (1.5%). In five cases, we identified no external bone defect at the base of the FEE that was considered as sequestered FEE or nasal glioma. Although most cases were simple, combined cases were also seen as detailed in Table 1.

FEE-associated conditions

We categorized the associated conditions into intracranial- and extracranial-associated conditions. Intracranial abnormalities was found in 64 cases included ventricular malformation (50%), ventricular enlargement (8%), porencephalic cyst (20%), arachnoid cyst (12%), and others (10%). Ophthalmological problems, found in 10% cases, were the main extracranial associated issues. Detail of associated conditions is presented in Table 2. Four patients had a recurrence mass and scars from previous surgery via a transfacial approach in another facility.

Surgical procedures

The authors (M.A and W.S) performed all surgeries either together or alone. The mean operative time was 2 h (range 30 min–3 h). Accurate blood losses were not documented; nevertheless, only two patients (0.5%) needed postoperative blood transfusions. Mean hospitalization time was 5 days (range 4–7 days). Examples of pre- and postoperative photographs are shown in Fig. 3.

Table 1 Type of surgically treated FEE

Type of FEE	Number of cases
NE	347
Isolated NE	306
Bilateral NE	216
Unilateral NE	71
NE with maxillary involvement	19
NE-NO combined	41
Bilateral NE + unilateral NO	24
Bilateral NE + bilateral NO	7
Unilateral NE + unilateral NO	10
NF isolated	34
NO isolated	14
Nasal glioma (sequestered FEE)	5

Table 2 Associated conditions observed in our 400 surgically treated patients

Condition	Number of cases
Size	
Small	104
Medium	230
Large	66
Enlargement/growing mass	
Constant	19
Proportional to the facial growth	372
Rapid enlargement	9
Skin stigmata	
Hyperpigmentation	23
Irregular skin	24
Hypertrichosis	3
Ectopic mass	8
Content on palpation	
Cystic/mostly CSF	27
Brain/compressible	127
Firm gliotic mass/uncompressible	78
Mixed of cystic and brain	168
Pulsating mass	12
Translucency on trans illumination	21
Eye-related conditions	41
Eye displacement	21
Microphthalmia	6
Anophthalmia	5
Strabismus	7
Pulsating orbit	2
Intracranial-associated conditions	
Ventricular enlargement	5
Ventricular malformation/asymmetry	32
Porencephalic cyst	12
Arachnoid cyst	10
Corpus callosum agenesis	4
Cavum vergae	3

Postoperative complications

The main postoperative complications were CSF leaks and wound dehiscence. Summary of postoperative complications is presented on Table 3. Postoperative CSF leak under the skin (pseudomeningocele) was observed in nine (2.25%) patients; CSF leak through the facial scar in four (1%) patients; CSF leak through canthal corner mimicking epiphora in three (0.75%) patients. The leak through canthal corner was resolved spontaneously within 2 weeks. The

Fig. 3 Pre- and postoperative photographs of representative FEE patients. **a**–**d** Example of facial incision used in the Chula technique. **e**–**f** The bicoronal approach (the modified Chula technique without facial incision) allowed removal of FEE in most cases with fairly good skin unless the herniated mass was large or a facial scar already existed

pseudomeningoceles were treated by multiple aspirations as the first line treatment. Four cases were resolved within 1-month follow-up. One case needed a shunt placement and two cases needed reopening and reclosure of the dura using fibrin glue. Wound dehiscence came with the consequences of exposed implant or exposed bone graft in seven (1.75%) cases. Implant removal or wound repair was performed and the skin issue was addressed.

Benign intracranial hypertension was encountered in five patients, and progressive hydrocephalus in three cases. Lumbar drain successfully reversed the benign intracranial hypertension. All progressive hydrocephalus underwent ventriculo-peritoneal shunt placement 2 weeks after the FEE surgery. Eye-related problems were found in a form of diplopia in two cases, postoperative epiphora in two cases, and obstructive nasolacrimal duct (OND) in four cases. Epiphora was improved 2 months after surgery. Diplopia was resolved 4 weeks after the onset. Unacceptable cosmetic results were found in 22 patients (discoloration, hypertrophic scar, and persistent canthal dystopia). Twelve out of 22 patients with cosmetic problem had a distorted maxillary bone. Total removal of the mass would leave the thin skin in direct contact

Table 3 Complications, follow-up treatment, and results

Type of complication	*n* (%)	Treatment	Results
CSF-related problem			
Skin CSF leak	4 (1%)	Re-opened, dural reinforcement suture, and fibrin glue	Resolved
Canthal CSF leak	3 (0.75%)	Observation	Resolved spontaneously
Pseudomeningocele	9 (2.25%)	Multiple aspiration (first line); VP shunt (second line); re-opened and dural reinforcement suture plus fibrin glue	4 resolved on first line treatment; 1 resolved with shunt; 2 resolved after reopening
Implant-related wound dehiscence	7 (1.75%)	Implant removal and reclosure	Resolved
Eye-related problem			
Diplopia	2 (0.5%)	Observation	Resolved
Epiphora	2 (0.5%)	Observation, ophthalmological follow-up	Improved
Obstructive nasolacrimal duct	4 (1%)	Nasolacrimal duct recanalization done by the ophthalmologist	Resolved
Intracranial complication			
Benign intracranial hypertension	5 (1.25%)	Lumbar drain placement for 5–7 days	Resolved
Progressive hydrocephalus	3 (0.75%)	VP shunt	Resolved
Cosmetic problem (bad scar, distorted bone, and skin discoloration)	22 (5.5%)	No cosmetic treatment	Patients had no complaint with the result

with the sunken maxilla, which resulted in a skin discoloration or distorted looks. To avoid this issue, we left a small chunk of gliotic mass at place to help shaping the maxillary bone level.

Discussion

The debate whether FEE is a neural tube defect continues as the pathogenesis itself is hardly understood. Many have defined it as a combination of genetic and environmental factors leading to the pathogenesis. The majority of our cases came from rural area with low socioeconomic status, suggesting the possible role of malnutrition during pregnancy. Similar to our study, reports of large studies suggest that this disease is linked to poverty [9, 16]. It could also explain the higher prevalence of FEE in developing countries than their counterpart developed Western countries. The socioeconomic impact of the disease might not be able to measure directly as several aspects played role in the resultant. Poor neighborhood and cosmetic appearance were two major aspects that influenced the patient's education level, household income level, and a chance to get a better job. School age patients were dropped out from school due to psychological impact of cosmetic issue despite the poverty itself.

All frontoethmoidal encephaloceles are present at birth but not all of cases will be considered as pathology by the family to seek help. In many cases, the pathology is asymptomatic even at adult age group. Suwanwela and Suwanwela proposed the widely used classification for encephaloceles based on external bony defect, in 1972 [6]. Later in 1998, Boonvisut and colleagues published their more detailed classification also based on external bone defect [17]. In 2003, Rojvachiranonda and colleagues proposed a more detailed classification including the facial deformity, external bone defect, exit pathway, and brain malformation [18]. This most recent classification provides us with a more detailed structural pathology but the ophthalmological problem is not addressed. Although an ophthalmology-related problem may not need urgent treatment, it may cause some problem. In our series, we found two kids who experienced diplopia after surgery that needed a further adaptation before they could gain their normal single vision. On a medium to large volume mass, we also found involvement or deformed maxilla. This involvement was not explicitly stated in Rojvachiranonda's classification but we suspected that the distortion of facial structure criteria covered this finding.

Specific finding from our series that has not been reported by other author is the pulsating mass and the pulsating orbit. Suryaningtyas et al. explained this finding as the nature of CSF pulsatile circulation and different displacement magnitude from different intracranial entities [19]. The connectivity of extracranial sac, large arachnoid cyst, and the intracranial subarachnoid space (SAS) made the theory of the pulsatile nature of CSF applicable.

The natural history of this malformation is not completely understood in terms of the enlargement progressivity and intracranial abnormalities. Several authors noted that the FEE grew larger with time [5, 20]. Based on parents' observation, it was said that at birth, the herniated sac was generally smaller and that it grew with time. In our series, we found only 5% of

cases that grew faster than its age. Ninety percent of cases grew according to age and "ceased" at certain age. The parents in this group confirmed that the mass was stable after their eighth birthday. Factors affecting the growing mass were the size of the external bone defect, the content of the sac, the length of exit pathway, and the existence of CSF-related intracranial abnormalities. A larger bony defect allows more brain or CSF to pass through as the intracranial pressure increased by age. A sac containing more fluid than solid mass tends to enlarge faster. A short exit pathway, especially the blowout type, plays role in progressive mass.

CT scan is routinely performed as pre-operative radiologic examination as, nowadays, it is widely available and the price is affordable at the range of $40–70. The use of MRI and digital angiography, as advised by some authors, was not applicable in our setting. For the price of $200–300, MRI as well as diagnostic angiography gives no additional critical information pertinent to operative procedure. We experienced zero complication related to vascular accident.

Early surgical correction is indicated to avoid deleterious effects on facial growth, especially in medium to large encephalocele. Undernourished, younger infant, low hygiene, and low socioeconomic conditions should be taken into account when adopting this principle. In developing country like Indonesia, delaying surgical treatment to certain age and weight should be considered to minimize complications. Controversy of the lethal prognosis of unoperated cases, as some authors claiming that only few patients will attain mature age, has no robust evidence. We have similar series of patients that survived to their adulthood age as other reports [10, 11, 21].

Our institution employs the Chula and the modified Chula techniques, as it does not need a formal frontal craniotomy. Modified Chula technique has additional benefit of no facial scar and reduces the CSF leakage through skin incision. Both techniques save more time, preserves frontal bone contour, less implant use, and reducing the risk of dural tear. We have no issues with visibility of the pathology during surgery as other authors reported and it was safe with low complication rate. We use chisel to perform the subfrontal osteotomy for it is superior to power craniotome in terms of bone loss and cheaper. The bone gap made with the chisel is almost negligible. The use of wire for bone reconstruction had been long gone. Before miniplate widely available, a small wire was the only measure to fix the bone. Some events occurred with the use of wire such as implant-related wound dehiscence, infection, and loose fixation. Miniplate might not eliminate those events but it reduced the number.

Most authors agree that resecting the herniated brain, which is considered as gliotic, will not cause any neurological consequences [22, 23]. We identified that the herniated brain was not all gliotic. Some part was normal brain and need to be cut to close the dura. Cutting this normal part of gyrus rectus caused no neurological issue in our series. However, we have no data regarding long-term consequences on the neuropsychological aspect.

Main complication in FEE surgery was CSF leakage. Two factors associated with CSF leaks were the transfacial approach and inadequate watertight dura closure. Mahapatra reported that 20% patients had CSF leak and 15.4% suffered from CSF rhinorrhea underlining the importance of a watertight dura closure [13, 24]. Overall, complication rate in our series was 12.5%, which is lower than the previous reports. The CSF-related complication rate was as low as 4% in our series. It is essential to be careful while reconstructing of the nasal skeleton not to perforate the nasal or ethmoid mucosa so that, even with dura leakage, CSF rhinorrhea can be avoided and the risk of infection decreased. We found no issue regarding the CSF rhinorrhea in our patients.

Endoscopic procedure was never proposed as the first line treatment for this type of encephalocele. It is conserved for basal encephalocele or to treat CSF leakage after skull base procedure. All of the reported pathology was intranasal and basal encephalocele that needed no intervention for the skin and bony part [25–28]. Endoscopy comes with several limitations including inabilities to reconstruct the nasal bone, to remodel the redundant skin, to close the external bony defect, and to do watertight dural suture. Malformed ventricle shape and abnormal anterior skull base anatomy make a surgeon difficult to perform endoscopy in FEE patient with CSF-related intracranial abnormalities.

Conclusion

The prevalence of FEE is still high in our country. The current socioeconomic conditions and local facility should be considered to treat these specific disease processes. The refined and meticulous technique, especially in choosing the approach and handling the dural closure, is essential in lowering the complication rate.

Compliance with ethical standards

Conflict of interest There is no conflict of interest.

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