International Surgery Journal
Singh S et al. Int Surg J. 2019 Sep;6(9):3251-3257
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: http://dx.doi.org/10.18203/2349-2902.isj20194061

Original Research Article

Autologous bone reconstruction as sandwich technique in pediatric posterior fossa surgery: finding a noble eightfold path for developing countries

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Received: 20 June 2019
Revised: 07 August 2019
Accepted: 09 August 2019

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ABSTRACT

Background: Sub-occipital craniotomy in pediatric population is difficult owing to uneven surface of growing calvaria and thin dura. Our novel technique using autologous bone chips and gelfoam bridges the two standard techniques. In this study, we intend to compare the surgical outcome in pediatric posterior fossa tumours.
Methods: We included patients, operated via midline sub-occipital approach, from January 2013 to October 2018 and grouped them, on basis of whether or not sandwich reconstruction was done. We compared pseudomeningocele, post-operative headache, CSF leakage and postoperative hydrocephalus requiring CSF diversion. The aesthetic outcome was assessed using Stony Brook scar evaluation scale (SBSES).
Results: 124 patients, divided into group A (n=53), group B (n=58) and group C (n=13) based on technique of surgical closure. The sandwich closure is significantly better in terms of both aesthesis and post-operative pain ($p<0.05$). There was a trend showing that sandwich closure decreases risk for pseudomeningocoele, wound infection, CSF leak and post-operative hydrocephalus. Median SBSES Score in group B was 4 compared to 2 in group A and patients were significantly more satisfied.
Conclusions: The uniform bone coverage with sandwich closure provides nearly similar reconstruction to craniotomy. All risks of using drill over pediatric calvaria are eliminated and advantages are carried. The bony barrier prevents adhesion and decreases both immediate and delayed headache. The technique is not only technically easier and aesthetically better, but also has better long term satisfactory results with possibility of neo-bone formation.

Keywords: Stony Brook scar evaluation scale, Delayed dragging headache, Satisfaction score, Neo bone

INTRODUCTION

Sub-occipital craniotomy in pediatric population is difficult owing to uneven surface of growing calvaria and thin dura. The sandwich technique is the plausible answer to the unsolved debate between "craniectomy" and "craniotomy", especially for paediatric posterior fossa (PPF) surgery.[1] The bony reconstruction, either with autologous chips or metallic implants, is credited with lower post-operative complications.[1,2] The technical familiarity ad ease in doing craniectomy creates a disfavour for micro drills and craniotomies.

Objective

In our article, we have attempted to restate the craniectomy for PPF tumors using the novel sandwich closure technique. The technique embraces autologous

bony chips sandwiched between two layers of gelfoam, thereby providing a scaffold for possible osteogenesis.

METHODS

The article is written according to the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement guidelines. Individual consent from the patient to use clinical and radiological details for publication was taken as per our department policy. Institutional ethical clearance for retrospective analyses was obtained.

Patient spectrum

In this study design, 167 paediatric patients (age less than 15 years), operated via midline sub occipital approach, from January 2013 to October 2018 were included (Figure 1). Majority of cases were operated by three senior authors of this study. We adopted sandwich technique in later three years of study, majority of patients operated in former time line were grouped as craniectomy without sandwich closure. The cases were retrospectively studied with all available data including age, sex, indication of surgery, histopathology, immediate and long term outcomes were recorded from hospital case records, outpatient files and the hospital information system.

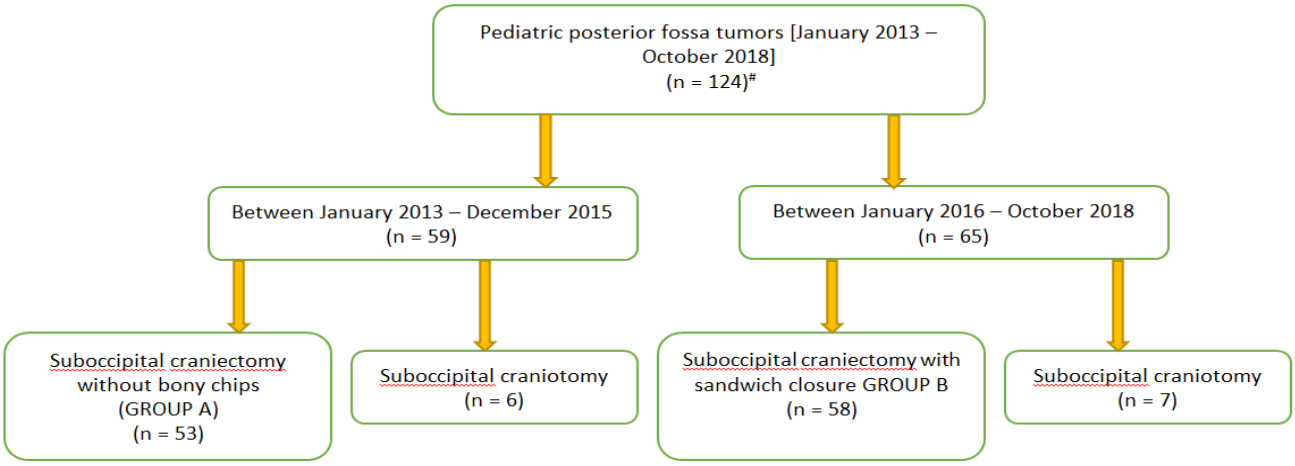

Figure 1: Study design.

Study parameters

We discharged all patients of posterior fossa tumor at seventh day of surgery. Any increase in stay more than seven days was labelled as long duration hospital stay. Clinically evident fluctuant swelling either symptomatic or not was labelled as pseudomeningocele (Figure 2A). Skin discharge or dehiscence associated to a positive microbiology culture was labelled as a wound infection. The infection was treated according to culture and sensitivity as targeted approach, and in all cases of CSF leak, a wound revision was performed and extra stitches were put on the dehiscence site.

We analysed following parameters (a) whether graft duroplasty was done or not, (b) pseudomeningocele formation, (c) wound infection, (d) osteomyelitis, (e) post-operative headache, (f) CSF leakage, (g) post op hematoma within 7 days, (h) postoperative hydrocephalus requiring hydrocephalus (i) histopathology. Wound aesthetic outcome, at last follow up, was assessed using Stony Brook Scar Evaluation Scale (SBSES) which comprises 5 point score (width less or more than 2mm; height with reference to surrounding skin; colour; suture marks; and overall appearance).

All caregivers were questioned for delayed post-operative pain (at 6th to 12th month post-surgery).[3] The caregivers were asked about dragging/stretching pain at surgical site. Their response was assessed using "five-point scale" (which included the following categories: 1= very satisfied; 2= somewhat satisfied; 3= neither satisfied nor dissatisfied; 4= somewhat dissatisfied; and 5= very dissatisfied). We also assessed whether bony chips fused or not. In all the available follow up computed tomography (CT) scans (n=28), bone formation at surgical site was analysed in terms of complete, partial or incomplete.

Exclusion criteria

We included patients up to 15 years only. Patients operated by lateral sub-occipital and retro-mastoid surgical approaches were excluded from our study. We excluded 9 cases, wherein dural closure was not acceptable and fibrin glue was used. Patients operated for Chiari malformation (n=32) were also excluded from study. 2 patients had immediate post-operative hematoma (in post-operative scan) and bony chips were not replaced back in second look surgery. These 2 patients were also excluded in view of crossover bias (Figure 2B).

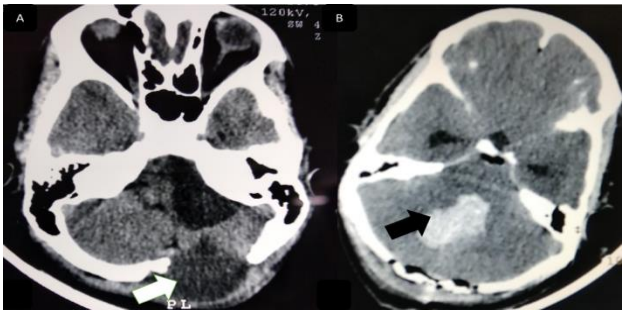

Figure 2: (A) Axial CT scans showing pseudomeningocele in patient where bony chips were not replaced (group A) as white arrow; and (B) black arrow denotes post-operative hematoma in a patient who was excluded from study as bony chips were not kept back in second look surgery.

Comparison between two groups

The patients were further grouped on basis of whether autologous sandwich closure was done or not and the study parameters were compared between two groups.

Surgical technique

The craniectomy was performed with single or two burr holes using perforator and rongeurs. The bone dust or chips preserved during craniectomy and was conserved in normal saline and crushed into similar sized pieces. Using sterile scale, gelfoam was fashioned to match the size of craniectomy defect (Figure 3). The autologous bone dust and small bony chips were sandwiched between two gelfoam pieces and placed into surgical defect (Figure 4). We allowed small amount of bleeding for chemical reactions on gelfoam to occur before layered closure of wound was done. Craniotomies were performed with control-depth-attachment high speed pneumatic drill.

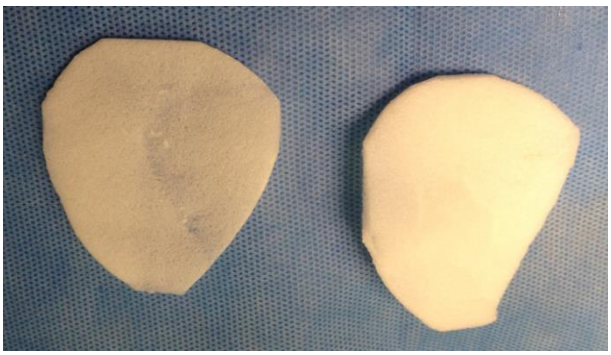

Figure 3: Sandwich shaped to match surgical site.

Radiological outcome assessment

The radiological assessment, using CT scan, was done at the time of discharge as well as at six months' and last available (for status of bony fusion) follow-up.

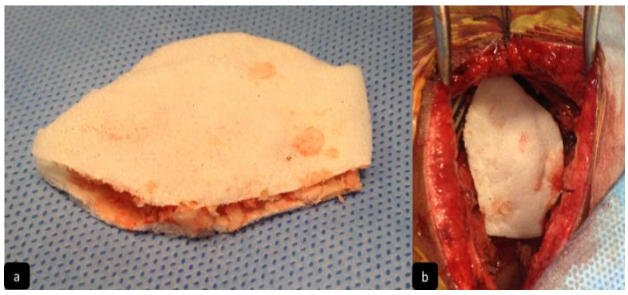

Figure 4: (a) Two gelfoam pieces and bone chips were conjoined to make sandwich (b) and kept over dura at surgical site.

Statistical analysis

The Statistical Package for the Social Sciences (SPSS) version 22.00 (IBM, New York) was used for statistical analysis. The confounding effect of age and gender distribution was compared using Fisher exact test. Fisher's test was used to compare study parameters between groups.

Follow up

The neurological condition at the last follow up was noted from the records and when needed the patient's relatives were called in outpatient clinic follow up and condition was ascertained.

RESULTS

167 PPF tumors were operated via sub-occipital approach between January 2013 to October 2018 at our institute. 124 patients met our stringent inclusion criteria, further divided into group A (craniectomy without bony chips) (n=53), group B (craniectomy with sandwich) (n=58) and group C (craniotomy) (n=13). The age and gender distribution between patients in group A and group B was similar.

Table 1 shows clinical and histopathological distribution in patients of both groups. Majority of patients were younger than 10 years with mean age in group A being 9.38±4.04 years and in group B being 7.06±3.78 years. Overall the male gender was dominant. The most common histopathology was medulloblastoma (52.8% in group A and 58.6% in group B) followed by ependymoma (18.9% in group A and 18.9% in group B). More than 96% of patients were admitted with features of raised intracranial pressure (of whom 37.7% in group A and 32.7% in group B required pre-operative CSF diversion). Other symptoms include ataxia, vision loss, cranial nerve involvement, and hemiparesis. Nearly 85% of patients in both group A and B underwent adequate (total and near total) excision. As the overall number of medulloblastoma patients was more in group B, 75.8% were subjected to adjuvant radiotherapy, in comparison to

60.3% in group A. The follow up ranges from 1 to 52 months.

The technical modification in closure of wound (sandwich technique), seems to nullify disadvantages of craniectomy while retaining all advantages of craniotomy. The sandwich closure is significantly better in terms of both aesthesis and postoperative pain ($p<0.05$) (Table 2). The additional advantage was that the bony chips were distributed uniformly throughout the surgical wound. We also found a trend showing that sandwich closure decreases risk for pseudomeningocoele (7 patients in group B as compared to 12 patients in group A), wound infection, CSF leak and post-operative hydrocephalus.

Patients in group B were significantly more satisfied in terms of delayed pain or dragging/stretching sensation on 5-point questionnaire. Median SBSES score in group B was 4 compared to 2 in group A, suggesting better aesthetic scar with sandwich closure in follow up also (Figure 5). 28 patients (with more than 1 year follow up) were assessed radiologically. We found that 22/28 (78.5%) patients had complete bone formation and 6 patients had partial bone formation (Figure 6). Three patients died of primary pathology (10 and 16 months' post-surgery). One patient was lost to follow up after radiotherapy.

Table 1: Demographical and clinical characteristics of patients analyzed in our study.

Characteristics	Group A (n=53)	Group B (n=58)
Mean age in years	9.38±4.04	7.06±3.78
Gender	M:F=37:16 (69.8% males)	M:F=45:13 (77.1% males)
Histopathology, N (%)		
Medulloblastoma	28 (52.8)	34 (58.6)
Ependymoma	10 (18.9)	11 (18.9)
Pilocytic astrocytoma	8 (15.1)	9 (15.5)
Diffuse astrocytoma	3 (5.7)	0
Others	4	4
Clinical features, N (%)		
Headache	52 (98.1)	56 (96.5)
Vomiting	52 (98.1)	56 (96.5)
Ataxia	41 (77.4)	41 (70.6)
Vision loss	14 (26.4)	13 (22.4)
Diplopia	11 (20.8)	05 (8.6)
Facial palsy	06 (11.3)	02 (3.4)
Hemiperesis	05 (9.4)	06 (10.3)
Hearing loss	02 (3.8)	0
CSF diversion in pre-operative period, N (%)		
EVD	3 (5.7)	5 (8.6)
ETV	2 (3.8)	4 (6.8)
VP shunt	15 (28.3)	10 (17.2)
Surgical excision, N (%)		
Total	18 (34.0)	17 (29.3)
Near total	29 (54.7)	33 (56.9)
Sub total	06 (11.3)	08 (13.8)
Adjuvant therapy, N (%)		
Radiotherapy	32 (60.3)	44 (75.8)
Chemotherapy	07 (13.2)	09 (15.5)
Follow up range	1-52 months	

Table 2: Study parameters compared in two groups.

Factors	Group A (n=53)	Group B (n=58)
Patient's relative perspective		
Bad immediate post-operative aesthesis at surgical site[#]	13	4
Local site itching or dragging[#]	31	12
Post-operative headache[#]	36	11
Objective criteria's		
Pseudomeningocoele[*]	12	7
Minor surgical site infection[*]	7	4

Continued.

Factors	Group A (n=53)	Group B (n=58)
Major surgical site infection	3	1
Osteomyelitis[@]	None	None
Post-operative CSF leak	8	10
Graft duroplasty	12	14
Post-operative hydrocephalus	2	4
Long term outcomes	2	4
Median SBSES Score	2	1
Delayed pain at surgical site (5-point scale)		
Radiological recurrence after 6 months	9	8

[#]Using Fisher's exact test, the values found to be statistically significant.
[*]The values significantly differ when compared to group C using nonparametric tests.
[@]Only 1 patient in study (group C) had osteomyelitis.

Figure 5: Representative photographs of patients with their SBSES of 2, 3 and 0 in (a), (b) and (c) subsequently.

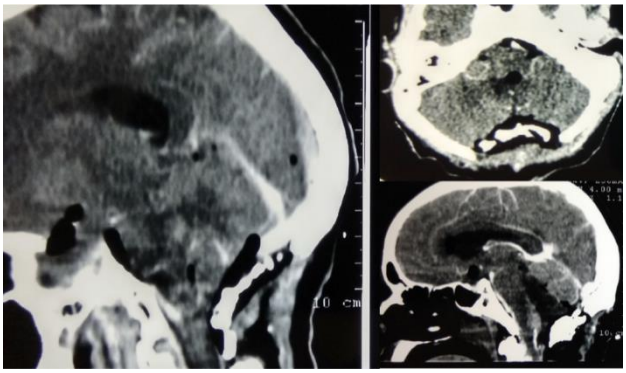

Figure 6: Representative sagittal and axial CT scans of patients showing variable degree of neo-bone formation at follow up.

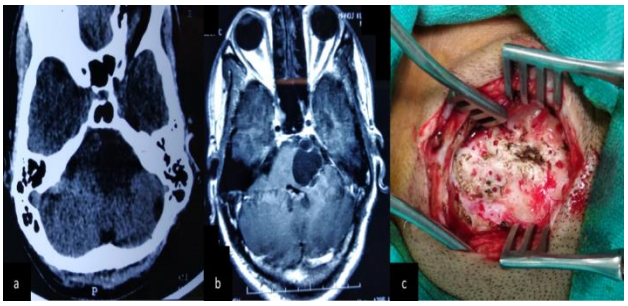

Figure 7: (a) CT scan (b) MRI axial section and (c) intra operative photograph of a patient being operated for recurrence with evidence of neo-bone formation.

DISCUSSION

While Yasargil and Fox were pioneers in describing reconstruction for posterior fossa; it was Missori who proposed the technique of posterior fossa reconstruction using autologous bone chips.[4,5] Since decades authors have tried to compare outcomes between craniotomy and craniectomy. There is persistent resistance in using a craniotome drill, considering the procedure dangerous; both because of the irregular contour of the inner bone surface and the tight adhesion of the dura to the skull.[6]

The thin and growing calvaria in pediatric population demands surgical expertise in dural and wound reconstruction after surgery.[7] To perform craniotomy on pediatric calvaria is difficult because of irregular shape and thin dura. The autologous bone reconstruction technique has been found easy, safe, inexpensive and less time consuming.[8] Although, there was no statistical difference between the two groups, a clinical trend suggesting better outcome in terms of pseudo-meningocoele and delayed pain. Previous studies do suggest significant advantage of craniotomy over craniectomy in terms of pseudomeningocele, CSF leak and wound infection.[9] We also found a similar trend in our study when craniectomy patients (n=111) were compared with craniotomy patients (n=13). The satisfactory bone coverage with uniform distribution of bone chips provides nearly similar reconstruction to craniotomy. Thereby, with sandwich closure, all risks of using drill over pediatric calvaria are eliminated and advantages are carried.

Status of surgical site eventually becomes an important parameter in malignant PPF surgery. There is indirect effect of surgical site infection on survival of patients also, as any such complication delays adjuvant radiotherapy for the patient. There was no significant difference in terms of surgical site infection and hospital stay between group A and B. In a study by Missori et al, no patient had wound related complication in immediate post-operative period.[5] 13 patients in group A and 4 patients in group B had bad wound due to muscular

atrophy or scar tissue. We applied objective Stony Brook scar evaluation scale to analyze delayed wound related outcomes.[10] Although the reliability of criteria is poor, because results are confounded by effect of radiotherapy and wound closure could not be generalized especially in a teaching institute, we got an overview that wound closed with autologous bone chips, look aesthetically better both in long term follow up also.

The post-operative CSF leak depends on integrity of dural closure rather than type of bony reconstruction. The exact mechanism of preventing CSF leak, by the bony shield is not known. Gnanalingham et al proposed that the bony shield prevents dural sutures from tearing out by postoperative subarachnoid CSF refill or raised pressure inside the posterior fossa.[11] In our study, the incidence of post-operative CSF leak is confounded by the fact that majority of patients (nearly one third in both the groups) had CSF diversion prior to definitive surgery. One of the important factors for high CSF leak rates may be poor nutritional status of patients in developing country.

None of the patient in sandwich closure group had osteomyelitis even in late post-operative period or even post radiotherapy. Contrary to our belief, one patient in craniotomy group had osteomyelitis and required re-exploration with removal of infected bone. We found 4 patients of hydrocephalus in Group B which might be because CSF is prevented from leaking or pressure effect of bone itself. Since all patients had ependymoma biopsy, hydrocephalus may be because of disease per se.

There was significant difference in terms of post-operative headache between groups. Objective analysis of delayed pain in a pediatric population is difficult, so we asked question to caregivers and their response was assessed on 5-point satisfaction score. We found that patients in group B were more satisfied even after long term follow up. The exact pathophysiology of delayed operative site headache is unknown. It is proposed that adhesion of cervical muscles with exposed dura at surgical site, result in traction during neck motion and thereby produce dural stretching headache. The "bony" barrier prevents adhesion and decreases pain thereby.[12]

We noted an interesting finding in our patients with long term follow up. There was radiological evidence of fusion at surgical site (Figure 7). We could evaluate 28 CT Scans and found complete bony fusion. We could not found histological details or "matrix strength" of this neo-bone, but believe that certain blood products promote osteogenesis and gelfoam sandwich might be acting as a scaffold. Additionally, 6 patients in group B and 5 patients in group A were operated for tumour recurrence. The time to expose dura in these recurrent cases was significantly less ($p<0.05$) in patients where sandwich closure was done. It is statistically not correct to justify our statement (n=6), but at least we can infer that uniform bony layer shields and prevents underlying vital structure. It is also easier to lift off "sandwich" as whole rather than multiple small bone chips.[6] Missori et al also experienced similar advantage where bony coverage makes re-exploration safer, with a diminished risk of dural tear and cerebellar damage.[5,8]

We intentionally allowed some bleeding over gelfoam and observe finally for any CSF leak, before closing subcutaneous tissue. There is indirect advantage of using gelfoam. We did not evaluate any statistical objective advantage of allowing to bleed but literature evidence shows that fibrin and Factor VIII stimulate fibroblast growth and facilitate wound healing in experiments.[13]

None of our patient had intra operative dural tear or technique related complication. During craniectomy, the rounger depresses underlying dura and thereby prevents dural tear. In a study by Chowdhury et al the complication rate in craniotomy group was 7% compared to 32.6% in craniectomy group.[6] We believe the technical complication is biased by surgeon's expertise and familiarity with surgical instrument.

Limitations of our study

Our study included a spectrum of histopathology from benign to malignant. Although the distribution was similar, a histology wise comparison or subgrouping may provide a better conclusion. The surgical outcome is confounded by surgeons expertize and overall experience. Majority of patients were operated by three senior authors of article but still technical bias is always there. The number of patient in group C were less and not comparable to group A and B. We analyzed the groups using nonparametric statistics but still a randomized control study with proper matching and blinding will provide better evidences justifying superiority of a novel technique over conventional.

CONCLUSION

Craniotomy requires specific instruments and a theoretical risk of dural tear and sinus injury; especially in paediatric patients, we propose that our sandwich closure technique is a comprehensive solution. Our study highlights the fact that, a little modification in technique is not only technically easier or aesthetically better, but also has better long term satisfactory results with possibility of neo-bone formation. The major problems like headache (both immediate and long term) and wound related complications are less in comparison to craniectomy.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: The study was approved by the Institutional Ethics Committee

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Cite this article as: Singh S, Bhaisora KS, Srivastava AK, Behari S. Autologous bone reconstruction as sandwich technique in pediatric posterior fossa surgery: finding a noble eightfold path for developing countries. Int Surg J 2019;6:3251-7.