Surgical management of cerebral venous sinus thrombosis: Case series and literature review

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ABSTRACT

Background: Cerebral venous sinus thrombosis (CVST) is not a common type of stroke (5%) but still hazardous to be misdiagnosed or mistreated. Aggressive medical treatment is usually failed to hinder increase intracranial tension. Therefore, decompressive craniectomy (DC) is the final measure to mitigate the deleterious effect of supratentorial herniation. The purpose of the study is to illustrate our experience with the surgical treatment of CVST and reviewing the previous works of literature.

Methods: Forty-two patients were admitted to Kasr Al-Ainy University Hospital from June 2019 to March 2020. The admission was either to the neurology department or intensive care unit or neurosurgery department. Every patient who was diagnosed with CVST received an emergency neurosurgery consultation. Seven patients were operated on with DC according to the criteria mentioned above. Therapeutic heparin was given in addition to intracranial pressure lowering measures.

Results: The mean and standard deviation of the age was (25.14 ± 10.1) years. There were five females (71.45%) in our series. The mean and standard deviation of clinical manifestations are (8.5 ± 7.77) weeks with range (3–14 weeks). Most of the cases were presented by a decreased level of consciousness (6/7) and anisocoria (6/7), followed by fits (3/7). Four cases out of seven had the previous history of oral contraceptive administration.

Conclusion: DC provides an urgent last arm for intractable increased intracranial tension. Patients with CVST need urgent consultation for neurosurgical intervention.

Keywords: Cerebral venous sinus thrombosis, Decompressive craniectomy, Hemorrhagic infarction

INTRODUCTION

The cerebral venous sinus thrombosis (CVST) is considered an ischemic episode due to blockage of main cerebral drainage of brain tissue with a resultant marvelous acute increment of intracranial pressure (ICP). The clinical manifestations of CVST are headache (75–90%), papilloedema, seizures, altered consciousness, and focal deficit. It is not a common type of stroke (5%) but still hazardous to be misdiagnosed or mistreated.

The pathophysiology of CVST can involve any territory of the venous system but commonly found at sinus-vein junction. Treatment is by far based on anticoagulants therapy, aiming to reanalyzed thrombosed veins, prevent propagation and progression of thrombus, and finally reverse coagulation process. Besides to anticoagulants, increased ICP is managed...
medically.[13] Supratentorial parenchymal herniation and increased cerebral edema are found to be present in 4% of cases.[4,18,24] This edema is resistant to all kinds of treatment and hence it is called (malignant edema). Aggressive medical treatment is usually failed to hinder increase intracranial tension. Therefore, decompressive craniectomy (DC) is the final measure to mitigate the deleterious effect of supratentorial herniation.[4,30,40]

The clinical and radiological signs for which DC is indicated included: third cranial nerve palsy, deterioration of conscious level, uncal herniation, midline shift ≥5 mm, ICP over 20 cm H₂O, and hypodensity of posterior cerebral artery territory.[1,16]

In this article, we are illustrating our experience with the surgical treatment of CVST and reviewing the previous works of literature.

PATIENTS AND METHODS

Study sample

It is retrospective study including 42 patients were admitted to Kasr Al-Ainy University Hospital from June 2019 to March 2020. The admission was either to the neurology department or intensive care unit (ICU) or neurosurgery department. Every patient who was diagnosed with CVST received an emergency neurosurgery consultation. Seven patients were operated upon with DC according to the criteria mentioned above after taking the consent from relatives. Therapeutic heparin was given in addition to ICP lowering measures. An ethical approval was retrieved from IRB of our college to start gathering information.

Perioperative treatment

For surgical patients, therapeutic heparin was discontinued 12 h before surgery and re-admitted again postsurgery with a prophylactic dose every 24 h. Prophylactic anticoagulant was given for more than a weak (11.48 ± 5.5 days).

DC

DC is simply bone excision and duraplasty. It acts by permitting space for edematous tissue to expand beyond intracranial anatomy with the creation of a closed space or room. It counteracts three models of herniation. The bone flap should include frontal, parietal, and temporal bones. The dural opening should guarantee maximal decompression. The dural edges are left with no approximated edges with the closure of overlying skin flaps only. A routine computed tomography (CT) scan should be obtained within 24 h of surgery. Re-implantation of the bone flap should not be discussed before 6 months.

Outcome analysis

Assessment of outcome included Glasgow coma scale (GCS), motor deficit, and modified Rankin scale [Table 1].

Statistical analysis

Data retrieved were processed using the Statistical Package for the Social Sciences (SPSS®) program version 25. Data of numerical values were compared using Chi-square t-tests while the categorical comparisons were tested by Fisher exact test. P = 0.05 was regarded as significant.

RESULTS

All patients’ criteria are illustrated in [Table 2]. The mean and standard deviation of the age was 25.14 ± 10.1 years. There were five females (71.45%) in our series. The mean and standard deviation (SD) of clinical manifestations are 8.5 ± 7.77 weeks with range (3–14 weeks). Most of the cases presented by a decreased level of consciousness (6/7) and anisocoria (6/7), followed by fits (3/7). Four cases out of seven had the previous history of oral contraceptive administration, while case #7 had a strong history of Antiphospholipid syndrome. Conscious level was recorded before surgery immediately. Four cases underwent intubation for securing airways. Most cases involved superior sagittal sinus at either segment with or without cortical vein thrombosis. Frontal infarction was seen 6/7 of cases (85.71%). Midline shifting was mostly over 0.5 cm with a mean and SD of 1.01 ± 0.34 cm. An apart from the dyed case, all cases showed improvement in GCS (2–5) points postoperatively with less than 0.5 cm midline shift. Hospital stay is measured in days. The mean and SD are 11.57 ± 5.5 days (21–5 days). The scale according to the modified Rankin scale showed a prevalence of three and four grades. Two cases died later after surgery (28.5%) in our series.

Table 1: Modified Rankin scale.

| Grade | Description |
|-------|-------------|
| 0     | No symptoms |
| 1     | No significant disability. Able to carry out all usual activities, despite some symptoms |
| 2     | Slight disability. Able to look after own affairs without assistance, but unable to carry out all previous activities |
| 3     | Moderate disability. Requires some help, but able to walk unassisted |
| 4     | Moderately severe disability. Unable to attend to own bodily needs without assistance, and unable to walk unassisted |
| 5     | Severe disability. Requires constant nursing care and attention, bedridden, incontinent |
| 6     | Death       |
## Table 2: Patients' criteria.

| No | Age | Sex | Onset | Clinical presentation | Risk factor | GCS pre | Thrombosed sinuses | Hemorrhagic infarctions | Volume (L×W × H) in cm³ | Midline shifting | Location of v infarction | Midline shifting | GCS post | Hospital stay | Modified Rankin scale |
|----|-----|-----|-------|-----------------------|-------------|---------|-------------------|------------------------|--------------------------|------------------|----------------------|------------------|----------|---------------|----------------------|
| 1  | 30  | F   | 14    | Headache, left sided weakness, fits anisocoria | OCP         | 13      | Bilateral frontal  | Ant 1/3 SSS left transverse and sigmoid hypoplasia cortical veins | 4.5×5.2×4, 2×0.7×2     | 1.2              | f                     | <0.5            | 15       | 7             | 1                     |
| 2  | 35  | F   | 8     | DCL, anisocoria | OCP         | 6T      | Right frontal     | Ant v3 SSS left transverse and sigmoid hypoplasia cortical veins | 5.5×4.5×4               | 0.8              | f                     | <0.5            | 10T      | 11            | 4                     |
| 3  | 40  | F   | 10    | DCL, bilateral unreactive | OCP         | 5T      | Right frontal     | Anterior 1/3 of SSS and frontal cortical vv Bilateral cortical frontal and parietal occlusion | 7.5×5.2×6               | 1.4              | f                     | <0.5            | 2T       | 5             | 6                     |
| 4  | 11  | M   | 7     | Fits, anisocoria | CKD         | 7T      | Left frontal      | Bilateral cortical frontal and parietal occlusion              | 4×5 × 4.5               | 1.2              | f                     | <0.5            | 15       | 15            | 5                     |
| 5  | 17  | M   | 7     | DCL, anisocoria | Trauma      | 11      | Left frontal      | Anterior 2/3 SSS cortical veins                                | 4.2×6.1×5               | 1.3              | f                     | <0.5            | 15       | 14            | 3                     |
| 6  | 23  | F   | 12    | DCL, anisocoria | OCP         | 13      | Left frontal      | Anterior 1/3 left frontal cortical veins                       | 6×6 × 4                 | 1.2              | f                     | <0.5            | 6T       | 8             | 3                     |
| 7  | 20  | F   | 3     | Fits, DCL, anisocoria | Antiphospholipid syndrome | 6T      | Right transverse and sigmoid    | Right occipito-parietal                                       | 5×6 × 4.5               | 0.4              | o.p                   | <0.5            | 9T       | 21            | 6                     |
CASE PRESENTATION

Case #1

A 30-year-old female with a history of oral contraceptives administration for long time, experienced an episode of headache 2 weeks before ictus, presented to ER with a slight confusion and left-sided weakness grade 4/5. She underwent CT brain, magnetic resonance imaging (MRI), and magnetic resonance venography (MRV). CT findings were of right frontal intracerebral hemorrhage with midline shift [Figure 1a]. MRI and MRV showed an evidence of superior sagittal sinus occlusion with hemorrhagic transformation of venous infarction [Figures 2-4]. One day later, her conscious level deteriorated to localizing to pain, no eye opening with sounds, and left side weakness became 3/5 and anisocoria. Surgery was done by DC and lobectomy. Grossly, cortical veins were thrombosed render hemostasis difficult. CT scan was done immediately postoperatively and shows no collection of hematoma with decrease shift. Conscious level improved immediately, while her weakness improved to 4/5 3 days thereafter.

Patient was discharged after 1 week with GCS 15/15, with gradual improvement in motor power, she was maintained for four days on clexane, in addition to Apixaban and antiepileptic. Three months after surgery patient presented with an attack of tonic clonic convulsions, as she was not compliant to medical treatment. At follow-up visit, she was fully conscious with full motor power, no hypertonia, or exaggerated reflexes. MRI brain with/without contrast were done and revealed resolution of cortical and subcortical damage with small area of encephalomalacia, MRV showed obliteration of cortical veins and anterior half of superior sagittal sinus [Figures 5 and 6].

Case #7

A 20-year-old female patient presented with known history of antiphospholipid syndrome. Three days ago, she started to have severe headache. One day later, she developed fits and decrease conscious level (E3V3M4). CT scan was done and a large occipitoparietal hematoma was seen on right side [Figure 7]. MRI obtained early before surgery showed signal void in the right transverse sinus [Figures 8 and 9]. She was operated on by decompressive craniotomy and evacuation of venous hemorrhagic infarction. Cortical veins were extensively thrombosed intraoperatively [Figure 10a]. Blood clots were retained from within the infarcted brain

Figure 1: Case 1, noncontrast computed tomography (CT) brain. (a) preoperative CT showed right frontal intracerebral hemorrhage with compression of ipsilateral frontal horn and midline shift. (b) postoperative CT brain. Decompression of the mass effect exerted by the hematoma with pneumocephalus in the bed of surgical site.
Figure 2: Case 1, (a) T1 WI with contrast showed right frontal intracerebral hemorrhage with surrounding edema, obliteration of ipsilateral frontal horn and displacement of contralateral horn with midline shift. (b) FLAIR image. The marked hypointensity of the ICH is most likely caused by the T2 shortening effects of intracellular methaemoglobin present abundantly in the clot at this stage.

Figure 3: Case 1 preoperative coronal T1WI with contrast (a) and T2 WI (b).
Figure 4: Case 1 preoperative magnetic resonance venography. Note occlusion of anterior half of SSS as well as left transverse sinus and sigmoid sinus.

[Figure 10b]. The patient was admitted to ICU for 21 days with no change in her consciousness. She developed severe acute respiratory distress syndrome and died due to severe respiratory failure.

**DISCUSSION**

In our study, failure of usual medical treatment of elevated ICP in the management of hemorrhagic infarction of CVST is an indication for DC. Sinus thrombosis is not a common cause of stroke. Certain textbooks referred to it as a rare category. In [Table 3], many records were in the form of case reports or case series. It is unethical to perform a randomized trial for DC to establish meaningful data. There is lacking data on surgical interventions worldwide.

Noncontrast CT scan was the first modality of choice to investigate patient with acute deficit or seizure. Thrombosed veins may manifest as elongated, cordlike hyperdense lesions (cord sign). The pathophysiology of this sign is clotted vessel. It is found in 20% of cases only. Empty delta sign is a triangular defect visualized on postcontrast imaging when SSS is filled with thrombus. This sign was seen in our series in postoperative MRIs for survived cases.

A DC is a good option for an enormous increment of intracranial tension, in comparison with data published before on implementation of DC in malignant edema due to MCA infarction, it has been found the survival rate was increased in the entity of “failed medical treatment” who underwent surgery versus sustained medical treatment. DC at either type of stroke (i.e., arterial or venous) provides a good “room” for an edematous brain, broken the vicious circle of increased ICP, and compression of neural tissues. Besides, DC prevents tentorial herniation and compression of respiratory receptors effectively. Hemorrhagic infarction after CVST is a major predicting factor for the unfavorable outcome or mortality either.

Various studies noticed the ominous sign of right-sided hemorrhagic infarction. In our study, right and left frontal hemorrhages were equally distributed (3:3). Girot et al. found that among old age, motor deficit, and deep venous thrombosis as predictors for bad outcome in mRS, right lateral (transverse
and sigmoid) occlusion is an important predictor for mortality or morbidity.\[^{13}\] Interestingly, the second case of mortality (#7) had had lateral system occlusion and large intracranial hemorrhage (ICH). Those with vein of Labbe occlusion may develop large hematoma with considerable midline shift if compared with the second 1/3 of SSS.\[^{1}\]
In [Table 3], our literature review was done from 1983 till now and covers many countries in the West and East world. Eleven articles registered mortality in their series. Our article represents the first case series came from Middle-East in the past 30 years. Other articles were devoid of clear data about the percentage of a favored and unfavorable outcome.

In our series, we followed the indications of DC strictly.[6] However, certain papers discussed performing of DC before pupillary reactivity and linked this with good outcome thereafter. The presence of nonreactive pupillary reflex means that ICP and edema are likely to persist even after surgery.[30] This point is needed to be clarified furtherly in the future. Are there special indications for DC in CVST other than known?

Endovascular treatment has no role in the management of hemorrhagic transformation with impending herniation.[1,7,16,20,40] Before 2006, the University of Amsterdam adopted a policy to perform DC, most of CVST patients with emergent transtentorial herniation died despite maximal conservative treatment and endovascular thrombolysis.[40] Aaron et al. found high mortality in the endovascular treatment of CVST.[1]

Stam et al.[12] tried thrombolysis and thrombectomy for severe CVST. Those with high midline shift, fulminant symptoms, and large venous territory affection had higher mortality.

Figure 6: Case 1 postoperative magnetic resonance venography. Note occlusion of anterior ½ of the SSS as well as cortical veins on right side.

Figure 7: Case 2 noncontrast computed tomography scan showed right occipito-temporal hematoma with midline shift and slight extension to ventricle. Note hypodensity of the ipsilateral hemisphere.
Giving anticoagulants in CVST are a crucial step in treatment. Anticoagulation prevents thrombus propagation, fasten spontaneous resolution, and helps in the prevention of deep vein thrombosis or embolism, without adversely promoting ICH.\textsuperscript{[20,38]} On the other side, ICH is not considered an anticoagulation drawback.\textsuperscript{[23]} Unfractionated heparin has to be given intravenously and it requires a dose adjustment based on activated partial thromboplastin time. Low-molecular-weight heparin is advantageous in that it can be administered as a subcutaneous injection based on body weight, and it has a more predictable pharmacokinetic profile. However, its effects are injection based on body weight, and it has a more predictable pharmacokinetic profile.\textsuperscript{[13]}

The time to restart therapeutic anticoagulation after DC is questionable.\textsuperscript{[12,20,40]} Previous studies suggested that anticoagulation can be restarted after 24–48 h and authors preferred to restart with half the dosage for a period of 72 h. Permanent anticoagulation is needed in those with prothrombotic states or with recurrent venous thrombosis.\textsuperscript{[22]} Other patients can be treated with oral Vitamin K antagonists for a period of 3–12 months. There is limited safety data for oral anticoagulants such as Apixaban.\textsuperscript{[14]}

Figure 8: Preoperative T2 WI. Note signal void of the right transverse sinus.

Figure 9: Case 2 magnetic resonance venography. Note stenosis of the right transverse-sigmoid sinus.

Figure 10: Case 2 (a) dural incision showed congested and marvellous edema with engorged cortical veins. (b) Blood clots retrieved from hematoma.
Table 3: Literature review.

| S. No. | Name               | Year   | Country     | n  | Age (range) | GCS | Favorable outcome | Unfavorable outcome | Follow up (months) |
|--------|--------------------|--------|-------------|----|-------------|-----|-------------------|---------------------|-------------------|
| 1      | Nagbal [26]        | 1983   | India       | 34 | -           |    | 19/34             | 15/34               | 120               |
| 2      | Kuroki et al. [17] | 1999   | Japan       | 3  | 23–46       | 4   | 0/1               | 1/1                 | NA                |
| 3      | Stefini et al. [33] | 1999   | Italy       | 3  | 40–54       | 4.7 | 2/3               | Nill                | 3–22              |
| 4      | Barbati et al. [3] | 2003   | Italy       | 1  | 15          | 5   | 1/1               | Nill                | 24                |
| 5      | Weber and Spring [36] | 2004 | Germany     | 1  | 62          | NA  | 1/1               | Nill                | 12                |
| 6      | Keller et al. [34] | 2005   | Switzerland | 4  | 37–66       | 6–13 | 4/4              | Nill                | 3–6               |
| 7      | Armonda et al. [2] | 2006   | USA         | 1  | 37          | 96  | 1/1               | –                   | 15                |
| 8      | Zeng et al. [19]   | 2007   | France      | 1  | 48          | 7   | 1/1               | Nill                | 6                 |
| 9      | Coutinho et al. [6] | 2008  | Netherland  | 3  | 36–55       | 5–13 | 2/3              | 1/3                 | 12                |
| 10     | Lin et al. [23]    | 2008   | Taiwan      | 1  | 29          | 11  | 1/1               | Nill                | 4                 |
| 11     | Lanterna [39]      | 2009   | Italy       | 3  | 32–51       | 3–8  | 2/3              | Nill                | 1–6               |
| 12     | Lath et al. [20]   | 2009   | India       | 11 | 18–46       | 3–14 | 8/11             | 3/11                | 6–10              |
| 13     | Galarza [41]       | 2009   | Spain, Italy | 1  | 34          | NA  | 1/1               | Nill                | NA                |
| 14     | Pfeilschifter et al. [20] | 2009 | Germany     | 1  | 49          | NA  | NA                | NA                   | NA                |
| 15     | Dohmen [8]         | 2010   | Germany     | 46 | NA          | NA  | NA                | NA                   | 3                 |
| 16     | Ebke [5]          | 2010   | Germany     | 29 | NA          | –   | 1/1               | Nill                | 2                 |
| 17     | Theaudin [34]      | 2010   | France      | 8  | 16–68       | 5–10 | 6/8              | 1/8                 | 19.7–45.6         |
| 18     | Mohindra [35]      | 2011   | India       | 13 | 14–45       | 5–10 | 11/13            | 2/13                | 26–60             |
| 19     | Zuurberg [40]      | 2011   | Netherland  | 10 | 26–52       | 2T–13 | 6/10            | 3/10                | 12                |
| 20     | Ferro [40]        | 2011   | Portugal    | 31 | 20–66       | NA  | 25/31             | 8/31                | 14.5 median       |
| 21     | Vivakaran [39]     | 2012   | India       | 34 | 18–65       | 4–13 | 26/34            | 6/34                | 4–22              |
| 22     | Raza et al. [30]  | 2012   | Pakistan    | 7  | 15–66       | 3–15 | 4/7              | 3/7                 | 2–77              |
| 23     | Aaron et al. [1]  | 2013   | India       | 44 | 19–60       | <9 in 30% | 27/44        | 17/44              | NA                |
| 24     | Graf [44]         | 2015   | Germany     | 1  | 20          | 6T  | 1/1               | Nill                | 12                |
| 25     | Lecchinoene et al. [31] | 2018 | France      | 1  | 45          | 6T  | 1/1               | Nill                | 60                |
| 26     | Gioti et al. [32] | 2019   | Cyprus      | 1  | 20          | 9   | 1/1               | Nill                | 4                 |
| 27     | Selisly           | 2020   | Egypt       | 7  | 11–40       | 6T–13 | 5/7              | 2/7                 | 1–3               |

NA, not assessed by the original author

CONCLUSION

DC provides an urgent last arm for intractable increased intracranial tension. Patients with CVST need urgent consultation for neurosurgical intervention.

Acknowledgment

A great thanks to neurosurgery department of our institute for their help and support.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Alselisly AM, Al-Shami H, Salah AM. Surgical management of cerebral venous sinus thrombosis: Case series and literature review. Surg Neurol Int 2021;12:133.