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Effect of Cranioplasty on Cerebrospinal Fluid Hydrodynamics in Patients with the Syndrome of the Trephined*

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Summary

Forty patients with cranial bone defects after craniectomy underwent extensive cerebrospinal fluid (CSF) hydrodynamic investigations by means of a CSF infusion test before and after cranioplasty. The results of these investigations were related to the clinical signs of the patients before and after cranioplasty and to the size and location of the skull bone defect. Twenty-two patients were considered to have "the syndrome of the trephined" (ST). The remaining patients were either free of symptoms or had symptoms not related to ST.

CSF hydrodynamic variables that were changed before and normalized after cranioplasty include the following: Resting pressure, sagittal sinus pressure, buffer volume, elastance at resting pressure and pulse variations at resting pressure. The changes were statistically significant mainly in ST patients who were also relieved of their symptoms after cranioplasty.

Keywords: Cerebrospinal fluid hydrodynamics; craniectomy; cranioplasty; "syndrome of the trephined".

Introduction

Patients subjected to craniectomy may experience a variety of clinical symptoms, some of which have been grouped as "the syndrome of the trephined" (ST)[3, 8–10, 15, 17, 20, 22, 24–26], "the

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postconcussive or posttraumatic syndrome"[10, 24] or "the syndrome of the sinking skin flap"[29]. The most commonly described symptoms are headache, vertigo, tinnitus, fatigue, discomfort, lack of concentration, insomnia, memory disturbance, irritability, and mental depression with or without dysphasia, dyspraxia, extremity paresis, and epilepsy. Some of these symptoms are aggravated by changes in body position or during the Valsalva manoeuvre.

A variety of theories have been developed regarding the role of the skull bone defect in the development of the neurological symptoms. The size and location of the cranial defect may be important[20, 21]. Derangement of cerebrospinal fluid (CSF) hydrodynamics have been described[4, 11, 13, 14], as well as regionally impaired cerebral blood flow[8, 18]. It has also been suggested that the atmospheric pressure acts directly on the cerebral cortex in cases with a concave deformity[11, 16, 19, 21, 29]. Since the prognosis for the patients with ST is favourable following cranioplasty, it was considered important to separate the ST patients from those having similar symptoms but a different aetiology. The comparison of CSF hydrodynamics between patients with true ST, partial symptoms of ST, and symptoms not associated with ST forms the basis of this report.

Material and Methods

Clinical and Radiological Assessment

The patient material consists of 40 consecutive craniectomized patients, 23 men and 17 women aged 22–74 years, mean 47 years. The disease leading to craniectomy was trauma in 17 cases, meningioma in 13 cases, angioma in 2 cases, cerebral aneurysm in 4 cases, arteriovenous malformation in 2 cases, intracerebral haemorrhage in one case, and encephalitis in one case. Six of these patients also had a postoperative intracerebral abscess. The time interval between craniectomy and cranioplasty was 3–51 months, mean 15 months. The operative technique used was a modified Woringer method using directly applied mesh reinforced methylmethacrylate[5, 27, 28]. All patients were examined neurologically before and after filling of their cranial bone defects. By means of conventional X-ray the size and position of the skull defect was measured from the frontal and lateral projection pictures. The size was determined from the projection in which it appeared to be largest.

CSF Hydrodynamic Investigations

CSF hydrodynamics were studied using the method of Ekstedt and Fridén[1, 2, 6, 7]. With the patient in the supine position the sagittal centre of the skull was used as the zero reference level. In the sitting position the midpoint between the horizontal level through bregma and inion was chosen as zero reference level. Both reference levels are at the height of the foramen Monroi. The

Table 1. *Normal Reference Values for the Measured CSF Hydrodynamic Variables*

	Mean SD	Dimension
1. Lumbar resting pressure (Pclr) I supine	1.38 ± 0.19	kPa
II sitting	no reference values	
2. Conductance of CSF outflow pathways (Gop)	17.98 ± 4.06	$mm^3kPa^{-1}s^{-1}$
3. Sagittal sinus pressure (Pss)	1.00 ± 0.20	kPa
4. CSF formation rate (Qf)	6.67 ± 1.39	mm^3s^{-1}
5. Pressure difference across outflow pathways (Pdop)	0.37 ± 0.10	kPa
6. Volume at elastance minimum [Vc(Emin)] (—)	19.20 ± 5.50	cm^3
7. "Buffer volume" (V buff)	15.30 ± 4.50	cm^3
8. Pressure at minimum elastance [Pc(Emin)]	0.96 ± 0.17	kPa
9. Elastance minimum value (Ec min)	0.02 ± 0.007	kPa cm^{-3}
10. Elastance at resting pressure [Ec(Pclr)]	0.066 ± 0.032	kPa cm^{-3}
11. Pressure at minimal pulsations [Pc(PcPmin)]	0.90 ± 0.25	kPa
12. Pulse variations at minimum pulsations (PcPmin)	0.06 ± 0.04	kPa
13. Pulse variations at resting pressure (PcPr)	0.24 ± 0.12	kPa

reference values for the variables are shown in Table 1. Due to technical difficulties, it was not possible to measure all variables before and after cranioplasty in all patients. The statistical significance of the differences was evaluated using a t-test for paired samples.

Results

Clinical and Radiological Studies

After cranioplasty the patients have been classified into the following four groups:

Group I. "True ST patients". Fourteen patients with symptoms included in the definition of ST aggravated during the Valsalva manoeuvre or from changes in body position. Their symptoms were not present before craniectomy and were reduced or relieved after repair of the cranial bone defect.

Group II. "Partial ST patients". Eight patients with symptoms of ST not present before craniectomy. The symptoms were more or less relieved after cranioplasty but were unaffected by the Valsalva manoeuvre or positional changes.

Group III. "Other patiens". Twelve patients with neurological deficits that could be related to their primary disease and the surgical procedure.

Table 2. *Relationship Between Clinical Symptoms and the Location of Cranial Bone Defect*

Patient group	Total	Frontal	Fronto-temporal	Temporal	Fronto-parietal	Parietal	Parieto-occipital	Occipital
Group I "True ST patients"	14	3	1			5	5	
Group II "Partial ST patients"	8	1	2		2	3		
Group III "Other patients"	12	1	6	1	2		1	1
Group IV "Symptom-free patients"	6	1	3			1	1	
Total	40	6	12	1	4	9	7	1

Table 3. *Clinical Symptoms Related to Positional Changes and Cranioplasty*

Clinical and neurological symptoms	Total no. of symptoms pre-operative	Group I "True ST patients" (14) Symptoms aggravated by changes in body position				Group II "Partial ST patients" (8) Symptoms not aggravated by changes in body positions			
		Before craniectomy	Relieved postop.	Improved postop.	Unchanged postop.	Before cranioplasty	Relieved postop.	Improved postop.	Unchanged postop.
Headache	10	6	3	3		4	1	3	
Vertigo	4	4	4						
Discomfort	2	2	2						
Irritability	2					2			2
Memory disturbance	11					11	2		9
Dysphasia	8	1	1			7	2	2	3
Dyspraxia	2					2			2
Hemi-monoparesis	17	4	4			13	2	4	7
Visual defect	2	1	1			1	1		

Group IV. "Symptom-free patients". Six patients without ST or neurological deficits.

In *group I* 79 percent of the patients were relieved of their symptoms and 21 percent were improved after cranioplasty. All these patients had flaccid skin flaps that became concave in the upright position. Ten of these patients had their defects located in the parieto-occipital region (Table 2).

All patients in *group II* were improved after cranioplasty. Seven of these patients had flaccid defects, and one patient had a rigid skin flap. The defects was located in the fronto-parietal region in $^5/_8$ cases. Patients in *groups III and IV* usually had rigid skin flaps, predominantly in the frontal and temporal regions. In these groups, those defects extended over the midline.

The mean size of the defect (cm^2) was 44 ± 24 in group I, 40 ± 24 in group II, 52 ± 21 in group III and 51 ± 27 in group IV. All patients with headache, vertigo, or a feeling of discomfort were improved or completely relieved of symptoms after cranioplasty, regardless of whether the symptoms were aggravated by changes in body position or not. Ten out of 17 patients with hemiparesis showed improvement or relief, as did $^2/_2$ patients with visual defects and $^5/_8$ patients with dysphasia. Memory disturbance was little affected by cranioplasty, and irritability and dyspraxia not at all (Table 3).

CSF Hydrodynamic Studies

The time between preoperative CSF hydrodynamic investigations and cranioplasty was 1–56 days, mean 6.5 days. The time between surgery and postoperative studies was 3 days to 3 years, mean 9 months. The results of the pre- and postcranioplastic CSF hydrodynamic measurements in the different groups of patients are listed in Table 4. The CSF resting pressure in the supine position was low in groups I, II, and III before cranioplasty, and significantly higher after. The CSF pressure in the sitting position showed a tendency to be higher before cranioplasty than after, but this could not be proved statistically. The sagittal sinus pressure increased significantly in the group I patients but was also normalized in the other groups after operation. Pulse variations at resting pressure were significantly increased (normalized) in group I patients after cranioplasty. The conductance and the pressure difference across the outflow pathways were unaffected by the operation in all groups of patients. The pressure/volume variables were mainly unaffected by the operation with some important

Table 4. *Results of CSF Hydrodynamic Measurements in the Different Groups of Patients.* The differences between the values before and after cranioplasty have been tested according to a t-test for paired samples. $P \leqslant 0.5$ is indicated by *, $P \leqslant 0.01$ by **. 1 kPa = 7.5 mm Hg = 102 mm H_2O. 1 $mm^3\ kPa^{-1}\ s^{-1} = 8 \cdot 10^{-3}$ ml $(mm\ Hg)^{-1}\ min^{-1}$

	Before cranioplasty				After cranioplasty			
	Group I	Group II	Group III	Group IV	Group I	Group II	Group III	Group IV
1. Lumbar resting pressure (Pclr)								
I supine	1.10 ± 0.38	1.19 ± 0.58	1.02 ± 0.46	1.38 ± 0.63	1.50 ± 0.30*	1.65 ± 0.44*	1.31 ± 0.32**	1.45 ± 0.55
II sitting	—1,09 ± 0.47	—0.96 ± 0.53	—1.16 ± 1.16	—0.37 ± 0.84	—1.47 ± 0.69	—1.61 ± 0.38	—0.62 ± 1.09**	—0.86 ± 1.08
2. Conductance of CSF outflow pathways (Gop)	21 ± 10	17 ± 8	9 ± 11	18 ± 7	20 ± 13	18 ± 10	16 ± 8	20 ± 8
3. Sagittal sinus pressure (Pss)	0.75 ± 0.32	0.80 ± 0.66	0.68 ± 0.31	0.88 ± 0.64	1.05 ± 0.28*	1.09 ± 0.39	0.93 ± 0.37	0.91 ± 0.43
4. CSF formation rate (Qf)	7 ± 2	8 ± 3	6 ± 1	8 ± 3	8 ± 2	7 ± 2	7 ± 3	8 ± 3
5. Pressure difference across outflow pathways (Pdop)	0.38 ± 0.16	0.46 ± 0.16	0.55 ± 0.42	0.51 ± 0.26	0.48 ± 0.23	0.56 ± 0.38	0.43 ± 0.19	0.47 ± 0.27
6. Volume at elastance minimum [Vc(Emin)]	8.92 ± 12.67	15.08 ± 42.88	11.18 ± 7.20	16.17 ± 2.36	14.19 ± 4.72	19.17 ± 3.52	18.83 ± 5.92	19.17 ± 3.79
7. "Buffer volume" (V buff)	31.03 ± 9.05	41.82 ± 42.68	21.68 ± 15.57	39.43 ± 27.53	19.19 ± 6.50*	12.0 ± 3.35	15.54 ± 5.80	15.57 ± 8.41
8. Pressure at minimum elastance [Pc(Emin)]	0.97 ± 0.26	0.71 ± 0.47	0.68 ± 0.29	0.65 ± 0.23	0.91 ± 0.23	0.81 ± 0.27	0.88 ± 0.14	0.90 ± 0.05
9. Elastance minimum value (Ec min)	0.018 ± 0.006	0.014 ± 0.008	0.016 ± 0.004	0.016 ± 0.011	0.023 ± 0.012	0.019 ± 0.004	0.014 ± 0.003	0.019 ± 0.005
10. Elastance at resting pressure [Ec(Pclr)]	0.025 ± 0.006	0.054 ± 0.060	0.049 ± 0.050	0.045 ± 0.051	0.067 ± 0.54*	0.090 ± 0.044	0.073 ± 0.028	0.082 ± 0.055
11. Pressure at minimal pulsations [Pc(PcPmin)]	0.085 ± 0.27	1.09 ± 0.17	1.06 ± 0.25	0.20 ± 0.18	0.83 ± 0.40	0.93 ± 0.38	0.62 ± 0.09	0.70 ± 0.14
12. Pulse variations at minimum pulsations (Pc(Pmin)]	0.06 ± 0.04	0.03 ± 0.03	0.08 ± 0.06	0.08 ± 0.01	0.05 ± 0.04	0.05 ± 0.03	0.05 ± 0.04	0.06 ± 0.28
13. Pulse variations at resting pressure (PcPr)	0.12 ± 0.09	0.15 ± 0.10	0.15 ± 0.10	0.22 ± 0.16	0.26 ± 0.21*	0.30 ± 0.19	0.17 ± 0.14	0.33 ± 0.28
Number of patients	14	8	12	6	14	8	12	6

exceptions: elastance at resting pressure was only half of the normal value in group I patients before operation but was normal after. The buffer volume was significantly higher preoperatively in group I patients but was also increased in the other groups as a reflection of the volume of the cranial defect per se.

Discussion

In the literature there are single or limited case reports describing the appearance of ST weeks and months after craniectomy, with aggravation of symptoms when changing body position, and relief or reduction of symptoms after cranioplasty[12, 16, 18, 19, 21, 23]. All these patients had large concave cranial defects. Starting from the assumption that many of the neurological symptoms present in the postcraniectomy patients stem from the presence of the bone defect and are not the result of surgery or the primary pathology which led to surgery, the observed changes in the CSF hydrodynamic parameters could be caused by the atmospheric pressure acting directly on the underlying cerebral tissue. Since the resting pressure is believed to depend primarily on the CSF formation rate, the conductance of outflow across the sagittal sinus and the sagittal sinus pressure, a change in resting pressure must be caused by changes in some of these components. The low resting pressure seen in our craniectomized patients was due to the low sagittal sinus pressure. The reason why the bone defect decreases the sagittal sinus pressure is unknown. The increase in resting pressure after cranioplasty can be explained by the rise in the sagittal sinus pressure. In the upright position the intracranial pressure is normally negative in a closed skull. If there is a cranial defect present the intracranial pressure will tend to equalize with the atmospheric pressure, which in turn will cause an increase in the intracranial pressure in the sitting position. The removal of a large bone segment will leave the cranium with a more or less flaccid area which should contribute to changes in elastance and volume variables. Such changes were identified in our patients. The size of the cranial bone defect was loosely related to the degree of abnormality in the volume and elastance variables. However, the rigidity of the tissue over the defect can greatly modify these measurements. True ST was found primarily in patients where there was a flaccid cranial bone defect in the region of the dural sinuses, particularly when this had been associated with a convexity or parasagittal meningioma (six cases in group I).

Resection of all or part of the sagittal sinus may render neighbouring cortical areas particularly susceptible to the influences of extradural (atmospheric) pressure on local cerebral blood flow. Postcraniectomy symptoms that were relieved by cranioplasty in our patients were mainly headache, vertigo, discomfort, and visual defects and to a lesser extent dysphasia, hemiparesis, and memory disturbance. In our opinion, only symptoms reduced or relieved by cranioplasty should be included in the definition of "the syndrome of the trephined".

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