



Surgical treatment of brainstem cavernous malformations: an international Delphi consensus

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OBJECTIVE Indication for surgery in brainstem cavernous malformations (BSCMs) is based on many case series, few comparative studies, and no randomized controlled trials. The objective of this study was to seek consensus about surgical management aspects of BSCM.

METHODS A total of 29 experts were invited to participate in a multistep Delphi consensus process on the surgical treatment of BSCM.

RESULTS Twenty-two (76%) of 29 experts participated in the consensus. Qualitative analysis (content analysis) of an initial open-ended question survey resulted in 99 statements regarding surgical treatment of BSCM. By using a multistep survey with 100% participation in each round, consensus was reached on 52 (53%) of 99 statements. These were grouped into 4 categories: 1) definitions and reporting standards (7/14, 50%); 2) general and patient-related aspects (11/16, 69%); 3) anatomical-, timing of surgery-, and BSCM-related aspects (22/37, 59%); and 4) clinical situation-based decision-making (12/32, 38%). Among other things, a consensus was reached for surgical timing, handling of associated developmental venous anomalies, handling of postoperative BSCM remnants, assessment of specific anatomical BSCM localizations, and treatment decisions in typical clinical BSCM scenarios.

ABBREVIATIONS BSCM = brainstem cavernous malformation; CCM = cerebral cavernous malformation; DVA = developmental venous anomaly; SH = symptomatic hemorrhage.

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CONCLUSIONS A summary of typical clinical scenarios and a catalog of various BSCM- and patient-related aspects that influence the surgical treatment decision have been defined, rated, and interpreted.

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KEYWORDS brainstem cavernous malformations; Delphi consensus; surgery; vascular disorders

BRAINSTEM cavernous malformations (BSCMs) are rare, accounting for approximately 30% of all cerebral cavernous malformations (CCMs),¹ which have a prevalence of about 0.5% in the general population.² Due to the eloquent location of BSCMs, symptomatic hemorrhages³ (SHs) of BSCMs are more frequent and cause more severe disability compared to supratentorial CCMs.^{1,4,5} These factors make patients with BSCMs potential candidates for neurosurgical treatment, which is performed in approximately 20%–25% of cases according to larger series.¹ Because such treatment is associated with significant risks for short- and long-term morbidity and impairment of health-related quality of life,^{5–9} the indications for surgery remain controversial.¹⁰ The anatomical complexity of the brainstem and the clinical heterogeneity of BSCMs in terms of lesion characteristics (such as symptoms, specific location, past clinical course and frequency of SHs, size, time point of SH, etc.) in combination with the relative rarity of the disease make comparative clinical investigations with a randomized controlled approach in BSCM very challenging.

Current treatment decisions are therefore mainly based on the individual surgeon's experience, with medical evidence limited to many case series, few comparative studies, and no randomized controlled trials.^{5,11} This limited evidence may lead to high levels of variation in the management and outcome of BSCM among clinicians.^{1,5,12} Guidelines for clinical management of CCM¹⁰ only give very general recommendations in this regard: the authors of the guidelines believe offering a resection after a second SH is "reasonable" (class IIb evidence, level B¹³), while an indication for resection after a single disabling SH is seen as more controversial (class IIb evidence, level C¹³). Clinical decision-making and selection for BSCM surgery requires considering more variables than merely the history and frequency of SHs.

Therefore, we sought to perform a survey using the Delphi method on multiple aspects of surgical treatment and management of BSCM among international experts to investigate the extent of current clinical consensus that could be achieved and identify questions and ambiguities that may drive future (randomized controlled) trials. The Delphi method is a structured, multiround communication technique in which an expert panel identifies and evaluates key aspects of a complex problem to reach a consensus.^{14–16} This method, among others, is recommended as a tool to help develop guidelines for rare diseases with low medical evidence.^{17–20}

Methods

Delphi Survey

An international group of experts in the surgical management of BSCM was selected by a steering committee (M.T.L., K.S., and U.S.). The selection process included the following steps: First, the number of potential panel

members was set at 20–30 according to survey extent, number of survey rounds, and comparable surveys.^{21–24} Second, inclusion criteria for panel members were defined as follows: clinical experience with BSCM management > 10 years (leading to an assumed minimum of 50 BSCM consultations), and/or contribution in PubMed (NCBI)–listed cohort studies or clinical series on BSCM (2005–2018), and/or prominent participation in clinical CCM research. The steering committee finally unanimously designated 29 international candidates based on their assumed willingness and availability to participate. The selection process was therefore performed consistent with those in comparable Delphi surveys.^{21,24}

All candidates were invited to take part in a basic Delphi survey²⁰ to identify and evaluate crucial aspects of surgical decision-making and management of BSCM. Data were collected using web-based surveys (SurveyMonkey) that were electronically distributed in 2018 and 2019. The 3 authors who designed the survey rounds (P.D., M.T., and B.S.) did not participate in the consensus process. Three panel members confirmed the eligibility of the first survey (M.T.L., K.S., and U.S.) and participated in the consensus process. An overview of the survey process is given in Fig. 1.

In the first round, participants were invited to answer 47 open-ended questions (see Supplemental Digital Content 1). The first survey was segmented into 5 sections: patient-, symptom-, cavernoma-, and intervention-related, and general questions. Additionally, self-reported background characteristics of each panel member were evaluated and reviewed, including the number of consulted BSCM cases, number of BSCM surgeries performed, and proportion of cases in which an indication for surgery and/or postoperative outcome was uncertain or unclear. A content analysis of the answers to the 47 open-ended questions, consisting of 4 stages (decontextualization, recontextualization, categorization, and compilation),²⁵ was performed (P.D., B.S., M.D.O., L.R., A.N.S.). Based on this analysis, 99 categorized statements were designed (see Supplemental Digital Content 2).

In the second round, these statements were grouped into 4 categories: 1) definitions and reporting standards (n = 14); 2) general and patient-related aspects (n = 16); 3) anatomical-, timing of surgery-, and BSCM-related aspects (n = 37); and 4) clinical situation–based decision-making (n = 32). These statements were then distributed to the group (see Supplemental Digital Content 3). Degree of agreement was rated according to a 5-point Likert scale (strongly agree, agree, neutral, disagree, strongly disagree, not my area of expertise [skip the statement]) and responses were anonymously collected (Fig. 2A). Responses with 80%–100% agreement (strongly agree, agree) or disagreement (disagree, strongly disagree) were considered acceptable to reach consensus. Responses with 70%–79% agreement (strongly agree, agree) or disagreement (disagree,

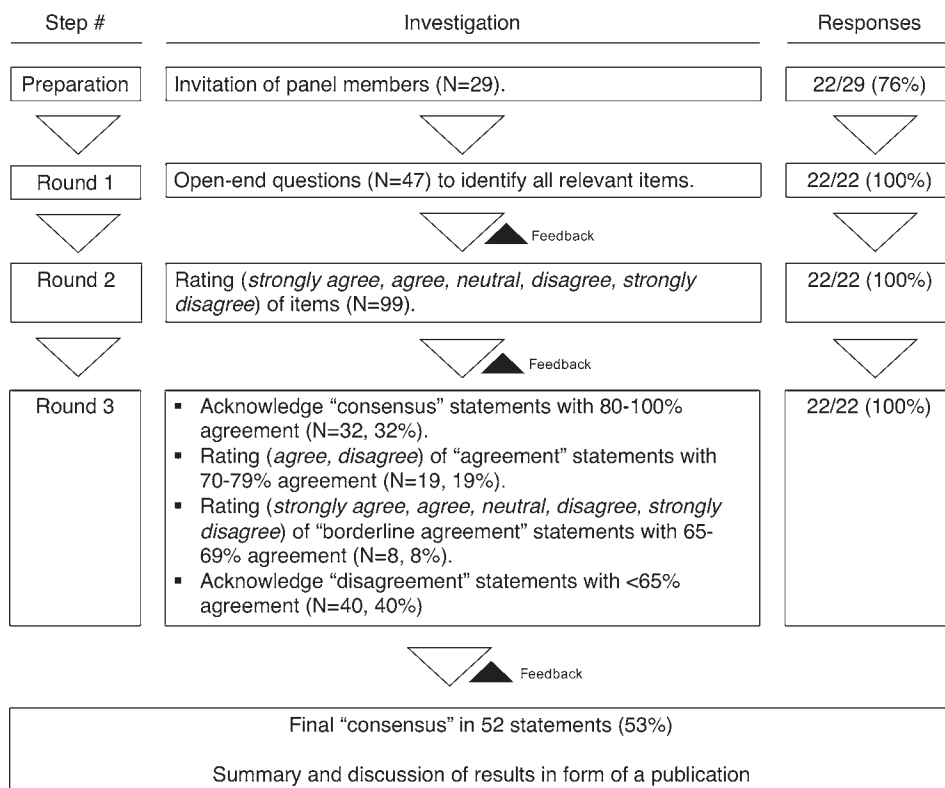


FIG. 1. Consensus study design. A group of 29 experts on BSCM were invited, 22 (76%) of whom participated in the Delphi consensus. In each round, participation and response rates were 100%.

strongly disagree) were defined as agreement. Responses with 65%–69% agreement (strongly agree, agree) or disagreement (disagree, strongly disagree) were considered borderline agreement. Based on the answers, statements were modified and distilled into 59 revised statements that functioned as feedback to the panel members.

A third survey was conducted to meet further consensus and revised statements were distributed to the group (Fig. 2B–D). Statements that reached consensus in the second survey were again confirmed (acknowledge, do not acknowledge). Statements that reached agreement in the second round were distributed to the panel members using a 2-point Likert scale (agree, disagree, not my area of expertise). Statements that reached borderline agreement in the second survey were again rated with a 5-point Likert scale (strongly agree, agree, neutral, disagree, strongly disagree, not my area of expertise). A final document including consensus statements was generated, in which statements were grouped to specific items, commented on regarding the clinical implications (P.D., M.T.L., K.S., U.S.), and reviewed by all participants (see Supplemental Digital Content 4, Tables 1–4).

Based on the consensus statements reached, a diagram covering typical clinical BSCM scenarios and their potential treatment options was designed (P.D., M.T.L., K.S., U.S.).

Results

Of the 29 invited experts, 22 (76%) responded and

agreed to participate (21/28 neurosurgeons [75%], 1/1 neurologist [100%]). All survey rounds were fully completed by all participants. Initial consensus (round 2) was reached on 33 statements. Initial agreement was reached on 19 statements, and initial borderline agreement on 9 statements. After reevaluation, final consensus (round 3) was reached on 52 statements (Fig. 1). Stratified by categories, consensus was reached on: 1) definitions and reporting standards statements (7/14, 50%); 2) general and patient-related aspects statements (11/16, 69%); 3) anatomical-, timing of surgery-, and BSCM-related aspects statements (22/37, 59%); and 4) clinical situation-based decision-making statements (12/32, 38%). For the first round, no median duration to complete the survey was recorded, whereas for the second and third rounds, 45.32 and 39.21 minutes were spent to complete the survey, respectively.

Consensus Statements

All 52 consensus statements were grouped into 4 categories, each consisting of several items: 1) definitions and reporting standards (5 items, Table 1); 2) general and patient-related aspects (11 items, Table 2); 3) anatomical-, timing of surgery-, and BSCM-related aspects (21 items, Table 3); and 4) clinical situation-based decision-making (9 items, Table 4). Information about several typical clinical BSCM scenarios merged into a diagram is found in Fig. 3.

Main Statements That Reached Consensus

The following selected, more-general statements are

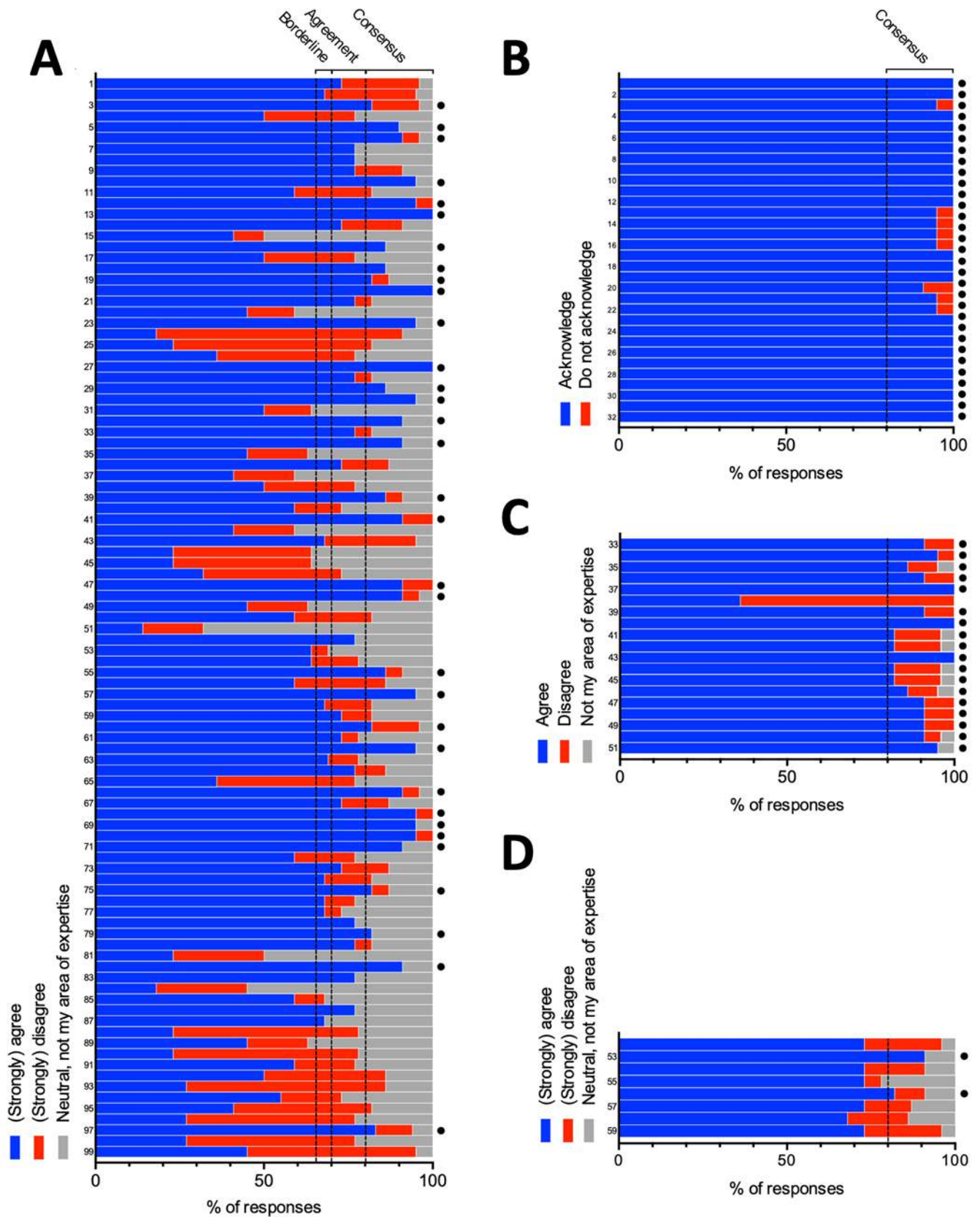


FIG. 2. Consensus process of second and third survey. **A:** Second survey results. Degree of agreement was rated according to a 5-point Likert scale (strongly agree, agree, neutral, disagree, strongly disagree). Responses with 80%–100% agreement (strongly agree, agree) or disagreement (disagree, strongly disagree) were considered consensus, and are marked with a circle. **FIG. 2. (continued)**→

FIG. 2. Responses with 70%–79% agreement (strongly agree, agree) or disagreement (disagree, strongly disagree) were considered agreement. Responses with 65%–69% agreement (strongly agree, agree) or disagreement (disagree, strongly disagree) were classified as borderline agreement. **B:** Third survey results. Statements that reached consensus (marked with a *circle*) in the second survey were again confirmed (acknowledge, do not acknowledge). **C:** Statements that reached agreement in the second round were distributed to the panel members using a 2-point Likert scale (agree, disagree). Responses with 80%–100% agreement were considered consensus and marked with a *circle*. **D:** Statements that reached borderline agreement in the second survey were again rated with a 5-point Likert scale (strongly agree, agree, neutral, disagree, strongly disagree). Responses with 80%–100% agreement (strongly agree, agree) or disagreement (disagree, strongly disagree) were considered consensus and marked with a *circle*. Figure is available in color online only.

considered to be clinically most important among the various statements upon which consensus was reached: symptomatic lesions are managed according to similar criteria in sporadic and familial cases; asymptomatic BSCM should not be treated, regardless of CCM or patient characteristics; a first severe SH with mass effect should undergo surgical treatment; recurrent SH with progressive neurological deficit and easy access of the BSCM should undergo surgical treatment; developmental venous anomalies (DVAs) should be spared during surgery and large DVAs can conflict the surgical approach and increase surgical risks; preferable timing for a resection is between 4 and 8 weeks after last SH event; and significant remnants identified on postoperative imaging should be directly resected, if accessible.

Controversies

Forty-seven items (47%) did not reach consensus, indicating controversial/neutral opinions among the panel members regarding these items (see Supplemental Digital Content 5). These items mainly concern the relevance of

intra- versus extralesional hemorrhage, follow-up imaging standards, interpretation of “growing” lesions (silent hemorrhages) on serial MRI, and the exact definition of a deep-seated lesion or difficult BSCM location.

Background Characteristics of Panel Members

Panel members reported consultations with an overall total of 5300 BSCM patients (median 200 patients). The number of reported surgeries was 1270 cases (unavailable in 3 panel members). The mean proportion of BSCMs treated was 27.4% (range 10%–55%). The proportion of uncertainty about expected postoperative outcome or indication for surgery in treated cases was 19.2% (range 5%–50%).

Discussion

This Delphi exercise achieved consensus (80%–100% agreement) for 52 (53%) of 99 statements relevant to surgery for BSCM, providing level III evidence¹³ (expert committee). Among other topics, panel members agreed

TABLE 1. Final consensus statements: definitions and reporting standards

Item	Statement	No. of Statements		Implication*
		Involved	Agreement	
Hemorrhage cluster	SHs that occur in an interval of days or (few) wks; increased risk for further SH	12, 13, 14, 37	90%–100%	Occurrence of cluster: pro surgery
Severe SH	A severe hemorrhage is an event significantly compromising the patient’s acute neurological condition &/or his/her daily life routines; this means the patient is dependent on others regarding personal care, etc., for a longer time (wks)	15	95%	In case of risk for (recurrent) severe SH, in case of past severe SH: pro surgery
Transient/permanent deficit	Transient = recovering during wks/mos, permanent = unchanged after 12–24 mos	16, 18	95%–100%	Recovery of symptoms can be expected & should be awaited: con surgery; a permanent deficit may offer a surgical route w/ decreased risk for neurological injury
Classification of SH	1) Life-threatening (coma, acute respiratory/cardiovascular depression); 2) severe w/ high risks of acute complications (deglutition, dysphagia, respiratory depression); 3) severe w/ major functional impairment (CN palsies, ataxia, hemiparesis); 4) mild w/ minor functional impairment (mild sensory deficits, minor weakness)	38	100%	SH 1–2: pro (immediate) surgery; SH 3: pro (planned) surgery; SH 4: con surgery; SH 1–2 are rather rare occasions, usually accompanied by larger size hematomas; SH 3 may also include isolated/mild CN palsies that argue for a watch & wait strategy
Intra-/extralesional SH	The intralesional hemorrhage supposes a lesion of the structures of the brainstem by compression; the extralesional hemorrhage disrupts the brainstem tissue	39	82%	Intralesional hemorrhages may have a better prognosis regarding neurological recovery, both spontaneously & after surgical removal

CN = cranial nerve; con = against; pro = in favor of.

* The item can influence the overall decision toward or against surgery.

TABLE 2. Final consensus statements: general and patient-related aspects

Item	No. of Statements Involved	Agreement	Implication*
Technical/organizational aspects			
Surgery for BSCM should be performed by a specialized dedicated team of neurosurgeons, neurophysiologists, & neuroanesthesiologists	19	100%	NA
Electrophysiological monitoring/mapping during BSCM surgery is mandatory	20	91%	NA
Neuronavigation for localization of BSCM/control of resection should be performed	40	82%	NA
To visualize (MRI) volume & age/consistency of intra- or extralesional hemorrhage is important for surgical planning	31	100%	NA
Patient-related aspects			
Age			
Age influences my decision as younger patients, having higher life expectancy, are at higher risk of recurrent hemorrhage (in patients presenting w/ hemorrhage) & tolerate surgery better; the opposite is true for older patients	2, 3, 50	90%–100%	Younger age: pro surgery; older age (>65 yrs): con surgery
Due to longer life expectancy/higher potential for recovery/more aggressive clinical course, I would be more “proactive” w/ surgery in pediatric patients compared to older patients in the same (CCM-related) clinical situation	33, 34	86%–96%	Pediatric population: pro surgery
Comorbidities			
Severe psychiatric/neurological disorders (depression, addiction, dementia) would prompt me to be more reserved w/ a surgical treatment	35	91%	Severe psychiatric/neurological disorder: con surgery
Severe comorbidities w/ a life expectancy <5 yrs would prompt me to be more reserved w/ a surgical treatment	6	100%	Life expectancy <5 yrs: con surgery
Meta-aspects			
Fear of a hemorrhage & thus decreased QOL may be a factor in considering a surgical treatment of a symptomatic lesion	4	100%	Decreased QOL/problems coping w/ fear of SH: pro surgery
Patient attitude toward life & risk, ability to cope w/ potential deficits & life-stage status is influencing my treatment decisions	5	100%	NA
Familial vs sporadic BSCM			
In a patient w/ multiple lesions (familial disease), the symptomatic lesions are dealt w/ in a manner similar to & based upon criteria associated w/ single symptomatic lesions in other patients	1	100%	NA

NA = not available; QOL = quality of life.

* The item can influence the overall decision toward or against surgery.

on the definition of so-called hemorrhage clusters and a classification of SH severity. Necessary technical and organizational requirements for surgical treatment of BSCM were outlined. Patient characteristics such as age, comorbidities, psychological aspects, and familial disease, as well as various BSCM-specific aspects (MRI morphology and location), were evaluated regarding their possible influence on an operative indication. A preferable time point of surgery after the last SH (4–8 weeks) and handling of postoperative BSCM remnants was agreed upon. Finally, the clinically most relevant basic BSCM scenarios (frequency and severity of SHs, location of BSCM) were rated regarding indication for surgery.

How to Read the Results?

The results of this work are condensed expert opinions reflecting personal experiences with the treatment and un-

derstanding of the currently available body of literature on BSCM. Given that absolute indications for surgery in BSCM appear to be rare (e.g., emergency situations with SH level 1–2; Table 1), and most of the time a very complex balancing of risks and benefits is necessary, this detailed evaluation of relevant clinical aspects regarding an indication for surgery is believed to function as an information baseline to drive future (randomized controlled) trials. As long as recommendations with higher levels of evidence do not exist, the extensive details of this work on different aspects of surgical treatment and indications may also comprehensively support current individual decision-making. The authors have deliberately refrained from establishing or proposing a grading scale with specific cutoff values, because such a scale would not reach the necessary level of detail. Validated grading scales based on a more limited number of parameters to estimate postoperative outcome are described elsewhere.^{6,26,27} However, based on

TABLE 3. Final consensus statements: anatomical-, timing of surgery-, and CCM-related aspects

Item	No. of Statements Involved	Agreement	Implication*
Anatomical aspects			
Posterior midbrain location	51	82%	Pro surgery
Near longitudinal fasciculus location	47	91%	Con surgery
Ventral medulla location	46	91%	Con surgery
Pulvinar location	45	91%	Pro surgery
Ventral midbrain (near red nucleus) location	30	100%	Con surgery
Near corticospinal tract	29	100%	Con surgery
Lateral pons location	28	100%	Pro surgery
If or if not approach via the anatomically safe entry zone lateral brainstem is possible, strongly influences my surgical decision-making	21	95%	NA
In general, lesions in the midline & deep-seated lesions pose a higher operative risk	26	100%	Con surgery
In general, a BSCM presenting to a pial or ependymal surface lowers risks of resection	27	100%	Pro surgery
BSCM-related aspects			
General			
The specific relation of hematoma location/size/quality (liquid, solid) & CCM location/size is a key factor for the surgical strategy	23	100%	NA
Perilesional edema			
A perilesional edema alerts me to the fact that the surrounding parenchyma is much more friable	49	96%	Con surgery
CCM shape			
I look for surface/shape of BSCM; according to the MRI appearance, dissection/resection may appear more/less invasive	48	91%	Important to estimate periop morbidity
CCM size			
Large BSCM (>2–3 cm) may displace normal anatomy, making utilization of entry zones, identification of midline structures, etc., much more difficult	44	86%	Important to estimate periop morbidity
w/ a very small BSCM there is the risk of performing the surgery w/o being able to find it	25	100%	Con surgery
DVA			
DVAs should be spared during surgery	22	95%	NA
Large DVAs may increase risk of surgery	41	100%	Con surgery
Timing of surgery			
Generally speaking, in acute phase (2 wks) tissue is more vulnerable due to edema, hematoma is solid & sticky; in chronic phase (after 2–3 mos) hemorrhage may be totally reabsorbed, leading to attachments & making dissection from surrounding tissue more difficult	43	82%	Pro surgery in subacute phase
In general, I prefer to operate approximately 6 (4–8) wks after the last hemorrhage; in this subacute phase hematoma is “fluid,” edema is normally reabsorbed, patient had time to partially recover function	42	82%	Pro surgery in subacute phase
In cases of large hematomas w/ significant mass effect (especially when reaching to the surface) & progressive deficit, early treatment in the acute phase should be considered	24	100%	Pro surgery
In case of a significant suspected BSCM remnant in early postop imaging, the remnant should be directly resected if accessible	97	83%	Pro surgery of postop remnants

* The item can influence the overall decision toward or against surgery.

TABLE 4. Final consensus statements: clinical situation–based decision-making

Item	No. of Statements Involved	Agreement	Implication*
SH frequency, neurological condition, lesion location			
Multiple SHs			
In case of multiple SHs causing a progressive neurological deficit, I favor an “aggressive” surgical posture, especially if the lesion reaches to the surface or has straightforward access (“easy to access”)	10	100%	Pro surgery
In case of multiple SHs causing a progressive neurological deficit, I favor an “aggressive” surgical posture, even if the lesion is deep-seated (“difficult to access”)	36	100%	Pro surgery
In case of multiple SHs, even if only causing a mild &/or transient neurological deficit, if the lesion reaches to the surface or has straightforward access (“easy to access”), I favor a surgical treatment	11	91%	Pro surgery
First SH			
In case of a first SH causing a severe neurological deficit (coma) or a progressive deficit (progressive hemiparesis) due to a mass effect, I favor an “aggressive” surgical posture	7	100%	Pro surgery
In case of a first SH even if only causing a mild &/or transient neurological deficit, if the lesion reaches to the surface or has straightforward access (“easy to access”), I favor a surgical treatment (also depending on patient age, risk factors, comorbidities)	9	100%	Pro surgery
In case of a first SH causing a mild &/or transient neurological deficit, especially if there is an increased surgical risk of excision based on anatomic characteristics of the lesion (“difficult to access”), I favor a conservative strategy	8	100%	Con surgery
No SH			
An asymptomatic BSCM should normally not be treated, regardless of size, location, MRI features, & patient characteristics	32	91%	Con surgery
Miscellaneous			
Long-tract deficits caused by a larger/space-occupying bleeding would rather prompt me to be more aggressive w/ surgery	17	100%	Pro surgery
In cases of large hematomas w/ significant mass effect (especially when reaching to the surface) & progressive deficit, early treatment in the acute phase should be considered	24	100%	Pro surgery

* The item can influence the overall decision toward or against surgery.

the results of the consensus, an overview of typical clinical scenarios and their potential treatment options is proposed (Fig. 3).

Generalizability to the Average Neurosurgical Practice

Surgical treatment of a rare and surgically challenging condition such as BSCM should be reserved for specialized teams that have already accumulated significant expertise and experience regarding surgical management and indications for treatment. Conversely, outcomes and indications of such experts should be continuously evaluated and reviewed, by establishing national and international registries and patient identifiers.²⁸ Consequently, the generalizability of the consensus of this survey to the average neurosurgical practice is limited. We do not believe that the survey can replace higher-level evidence practice guidance, but it serves to condense currently available expert opinions. Of course, randomized controlled trials have the highest internal validity, providing the most reliable level of evidence guiding practice. However, these trials are limited regarding flexibility and generalizability to neurosurgical “real-world” practice and their necessary level of detail, especially in rare, heterogenous, and complex diseases such as BSCM.

Recently, the National Institute of Neurological Disorders and Stroke of the NIH has published recommendations on how to address such challenges of clinical research in neurosurgery in the future.²⁸

Strengths and Limitations

Among the strengths of this study is the extensive experience of the panel members (n = 22), reporting 5300 overall consultations and 1270 surgeries for BSCM. For comparison, the largest meta-analysis to date comprises 2493 surgical cases from 86 studies since 1986.⁵ The relatively typical reported mean proportion of patients with BSCM who underwent surgical treatment by panel members (27.4%) reflects the external validity of the panel members’ overall surgical “posture.” However, a proportion ranging from 10% to 55% also indicate a wide variability in surgical management among the panel members. It is surprising that despite this variability, an agreement of 80%–100% was reached on 52 (53%) of 99 statements. Methodological strengths of our paper are the conception of a three-round survey with panel member feedback (circulation of previous responses) in between all rounds, and a clear definition of a desired consensus (80% agreement on a Likert scale). Although all these aspects are recom-

Surgery for acute relief of mass effect

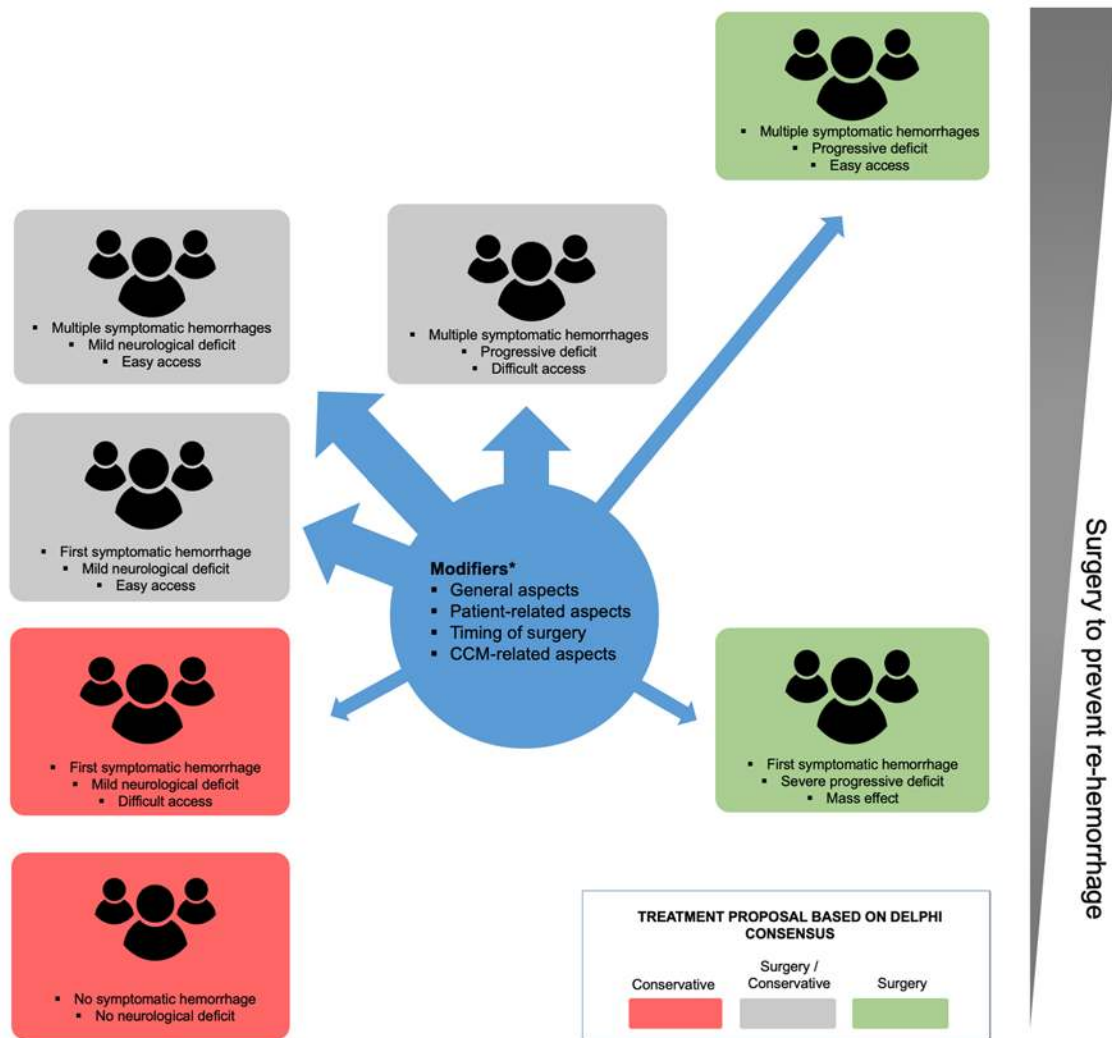


FIG. 3. Overview diagram according to consensus statements. Typical clinical scenarios of BSCM and corresponding treatment options reflecting the current Delphi consensus statements. Especially in cases in which both conservative and surgical treatment appear to be options (*gray fields*), the proposed modifiers (*asterisk*, as described in Tables 1–4) are relevant. While this diagram is believed to display general treatment tendencies, of course other scenarios are possible and should be addressed accordingly. Easy access is defined as superficial, reaching the pial or ependymal surface. Difficult access is defined as deep-seated, several millimeters of brainstem tissue to cross, no safe entry zone suitable. Figure is available in color online only.

mended for a Delphi survey, they are often not adhered to.²⁰ The number of panel members was only slightly above the average of that in other Delphi surveys (n = 17) as reported in a meta-analysis.²⁰ Because selection of panel members is crucial for the consensus technique to work properly,²⁹ we followed recommendations in panel member selection (expertise and willingness and commitment to participate)³⁰ and thus reached a response rate of 100% in each round. Overall, this Delphi consensus is in accordance with the Guidance on Conducting and Reporting Delphi Studies (CREDES),³¹ which is, however, a guideline for Delphi studies in palliative care.

Limitations of our work are the inclusion of terms that

are only roughly defined and leave space for individual interpretation, such as “easy” or “difficult” surgical access. However, such a scope for interpretation is due to the anatomical and clinical complexity of BSCM and seems ultimately unavoidable. Another limitation is the overrepresentation of neurosurgeons (n = 21) versus neurologists (n = 1, with extremely high expertise in clinical CCM research) as panel members. This survey was, however, specifically planned to evaluate the core surgical aspects and indications of BSCM management, consequently demanding a neurosurgical background. Another limitation is the exclusion of radiotherapy as a treatment option for BSCM, which, although discussed with controversy, is established

for the treatment of BSCM.³² However, the original aim of this work was to show consensus and controversy regarding selective surgical treatment in great detail. The balance between surgical, radiotherapeutic, and conservative treatment may also be a suitable topic for a Delphi consensus but would require a completely different structure of survey and panel members and should therefore be investigated in future studies.

Conclusions

We present a detailed summary and evaluation of relevant aspects for decision-making in the surgical treatment of BSCM. Due to lack of evidence from clinical trials, we performed a standardized multi-round Delphi survey with a large number of highly experienced experts to identify consensus and ambiguities. As a result, a summary of typical clinical scenarios and a catalog of various BSCM- and patient-related aspects that influence the surgical treatment decision have been defined and interpreted. We thus provide information to support BSCM-focused neurosurgeons in current clinical practice and inform clinical trial design, to ultimately improve the quality of the evidence for the management of BSCM.

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Author Contributions

Conception and design: Dammann, Schaller, Lawton, Sure. Acquisition of data: Dammann, Darkwah Oppong. Analysis and interpretation of data: Dammann, Santos, Rauschenbach, Darkwah Oppong, Schmidt, Schaller, Lawton, Sure. Drafting the article: Dammann. Critically revising the article: Dammann, Al-Shahi Salman, Santos, Rauschenbach, Darkwah Oppong,

Schaller, Lawton, Sure. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Dammann. Statistical analysis: Schmidt. Administrative/technical/material support: Dammann, Santos, Rauschenbach. Study supervision: Dammann, Couldwell, Ogilvy, Lawton, Sure. Delphi Consensus expert participant: Abla, Al-Shahi Salman, Andrade-Barazarte, Beneš, Cenzato, Connolly, Cornelius, Couldwell, Sola, Gomez-Paz, Hauck, Hernesniemi, Kivelev, Lanzino, Macdonald, Morcos, Ogilvy, Steiger, Steinberg, Spetzler, Schaller, Lawton, Sure.

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