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SPECIAL ANNUAL ISSUE

Bony decompression vs duraplasty for Chiari I malformation: does the eternal dilemma matter?

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Abstract

Purpose The management of Chiari I malformation (CIM) still raises the problem of the optimal surgical treatment, with special regard to the "eternal dilemma" of the posterior fossa bony decompression alone (PFBD) or with duraplasty (PFBDD). The goal of the present review is to update the results (outcome and complications) of both techniques to better understand the correct indication for each of them.

Methods A review of the literature has been performed, focusing on the articles and the meta-analyses specifically addressing the problem of PFBD vs PFBDD. Also, the personal authors' experience is briefly discussed.

Results PFBD (usually with C1 laminectomy, often with delamination of the external dural layer) is the most commonly used technique in children, especially if syringomyelia is absent. It ensures a high success rate, with > 80% clinical improvement and about 75% reduction of the syringomyelia, and a very low risk of complications, hospital stay, and costs. A certain risk of recurrence is present (2–12%). PFBDD (with autologous tissues or dural substitutes), on the other hand, is mostly used not only in adults but also in children with large syringomyelia. It is burdened by a higher risk of complications (namely, the CSF-related ones), longer hospital stay, and higher costs; however, it warrants a better clinical improvement (> 85%) and a lower risk of reoperation (2–3.5%). Eight meta-analyses of the literature (three on pediatric series and five in adult series) and one prospective study in children, published in the last decade, largely confirm these findings.

Conclusion PFBD and PFBDD are different techniques that are indicated for different types of patients. In children, PFBD has been demonstrated to represent the best choice, although some patients may require a more aggressive treatment. Therefore, the success in the management of CIM, with or without syringomyelia, depends on the correct indication to surgery and on a patient-tailored choice rather than on the surgical technique.

Keywords Chiari I malformation · Posterior cranial fossa · Craniectomy · Decompression · Duraplasty

Background

The management of Chiari I malformation (CIM) still keeps on raising the problem of the optimal surgical treatment. Namely, the "eternal dilemma" of the posterior fossa bony decompression alone (PFBD) or with duraplasty (PFBDD) is continuously re-proposed and discussed in many original studies and meta-analyses of the literature. According to Mazzola and Fried, the main causes for revision surgery for CIM decompression are the conservative or inadequate initial decompression and/or the occurrence of postoperative complications and/or the appearance/persistence of syringomyelia [45]. Therefore, they should be regarded also as the main reasons for the still open debate on PFBD versus PFBDD. In addition, however, another aspect has to be considered, that is a (possible) wrong indication to surgery. Actually, as the diagnosis of CIM has become quite common, the spectrum of symptoms and signs attributed to it is wider and wider and, sometimes, confusing (this problem is addressed specifically in this Annual Issue), thus making hard the decision process about the indication to treatment. The matter is further

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complicated by the increasing frequency of clear psychiatric disorders diagnosed among the CIM pediatric population (attention deficit hyperactivity disorder in 22% of cases, anxiety about 13% and depression in 10.5%) [31]. As a result, in the clinical practice, a certain proportion of patients may remain symptomatic just because of a "wrong" indication rather than because of an "unsuccessful" surgical technique.

In fact, in spite of many observations in favor of both a benign course of CIM in children and a subsequent low proportion of patients needing surgery [35, 53, 57], the real rate of surgical treatment is continuously increasing (at least in the USA, where this problem was specifically analyzed) [73]. Information on the current epidemiological trend, concerning PFBD vs PFBDD in the USA, has been provided by Shweikeh et al. [65]. According to their analysis, based on 1593 children treated with PFBD and 1056 children treated with PFBDD in the 2000–2009 decade (USA Kids' Inpatient Database), patients undergoing PFBD were slightly younger (9.8 vs. 10.9 years, $p = 0.001$), less likely White (75.6 vs 81.2%, $p = 0.04$), more frequently admitted in emergency (13.8 vs 8.4%, $p = 0.007$), less likely to have reoperations (0.7 vs 2.1%, $p = 0.01$) and complications (0.8 vs 2.3%, $p = 0.003$), hospitalized for a shorter period (3.8 vs 4.4 days, $p = 0.001$) and with lower charges (USD 31.483 vs 35,321, $p = 0.01$). Although this study is limited by data on preoperative presentations and long-term outcomes, the authors concluded that PFBD is more favorable for CIM patients because it is more economical and requiring fewer hospital resources and because PFBDD is burdened by a higher rate of complications and immediate reoperations.

A second problem that can justify the repeated analysis of the PFBD versus PFBDD policy is the absence of guidelines on the management of CIM. Such a phenomenon mainly results from the still missing clear data on the pathophysiology and symptomatology of the disease, other than on the evidence of different subsets of patients composing the CIM population. A third problem is the lack of standardized methods for the assessment of the postoperative outcome [20]. Nevertheless, these aspects indicate that each CIM patient deserves a personal indication based on the age, the clinical picture, and the radiological characteristics rather than a continuous update on the pros and cons of PFBD vs PFBDD.

PCF bony decompression (PFBD)

Rationale, indications, and techniques

The rationale of PFBD is to re-create the cisterna magna indirectly. Actually, the removal of the suboccipital squama, together with the opening of the foramen magnum and, in many cases, the removal of the posterior arch of the atlas is demonstrated to provide enough space to accommodate the expansion of the dura mater and to allow the CSF to be forced again into the subarachnoid spaces by the cerebellar pulsations [28, 59]. Such an indication is supported by the evidence that, independently from the causative pathophysiological mechanisms of the hindbrain herniation, the hindbrain is deformed but not malformed [58]. This finding would justify a poorly aggressive policy in the surgical management of the condition other than the use of the term "Chiari I deformity" instead of CIM. Actually, though some authors advocate the use of PFBDD in children [16, 41], the majority of pediatric neurosurgeons utilizes PFBD in most of the cases (except for the recurrences) because of the low rate of complications (especially, CSF leakage) and the virtually nil risk of surgical mortality [6, 19, 27, 29, 34, 37, 51].

The reason of the success of PFBD in the pediatric population should be identified in the PCF progressive enlargement due to the physiological growth of the skull. Subsequently, and differently from adults, PFBD can enhance a spontaneous bone enlargement, thus resulting in an effective procedure in giving space to the neural structures. Therefore, the extent of the PCF opening should be tailored according to the patients' characteristics and age. For example, in infants and young children, the suboccipital craniectomy should not be excessively large, to reduce the risk of cerebellar ptosis, nor too small, to reduce the risk of bone regrowth (that we found in some cases in our personal series) [44]. Based on our experience, we suggested a bony opening, centered on the foramen magnum, not exceeding 2.5/3 × 2.5/3 cm in this sub-population [43]. Moreover, because the opening of C1 seems to be necessary in the majority of cases of the analyzed series, it is advisable to include this step in the PFBD procedure (independently from the age of the patient), to increase the decompression surface and to decrease the risk of CIM recurrence. In older children and adolescents, where the bone is thicker and its growth slower, the extent of the bony opening should be tailored on the severity of the clinical and radiological picture. Furthermore, intraoperative ultrasounds can be successfully used to get this goal [44, 49, 76]. Indeed, the direct visualization of the re-opened cisterna magna and/or the newly pulsating tonsils can help the surgeon in assessing the correct extension of the craniectomy/laminectomy and, at the same time, in evaluating the need of dural opening. In the personal experience, we usually find a good improvement of intraoperative ultrasounds only after the removal of the epidural "ring" at the bulbo-cervical junction represented by a thickened atlanto-occipital membrane remnant (Figs 1 and 2). A limit of intraoperative ultrasounds is represented by the position of the tonsils lower than C1, since their pulsations and/or the ventral and dorsal CSF space re-expansion could be not appreciated adequately (although this could be a limit intrinsic to PFBD when it is not enough to reopen the CSF spaces or allow the tonsils to pulsate) [46]. The spontaneous re-expansion of the cisterna magna in prone position could be a further limit of the strategy based on intraoperative ultrasounds, at least in some cases. Bond et al.,

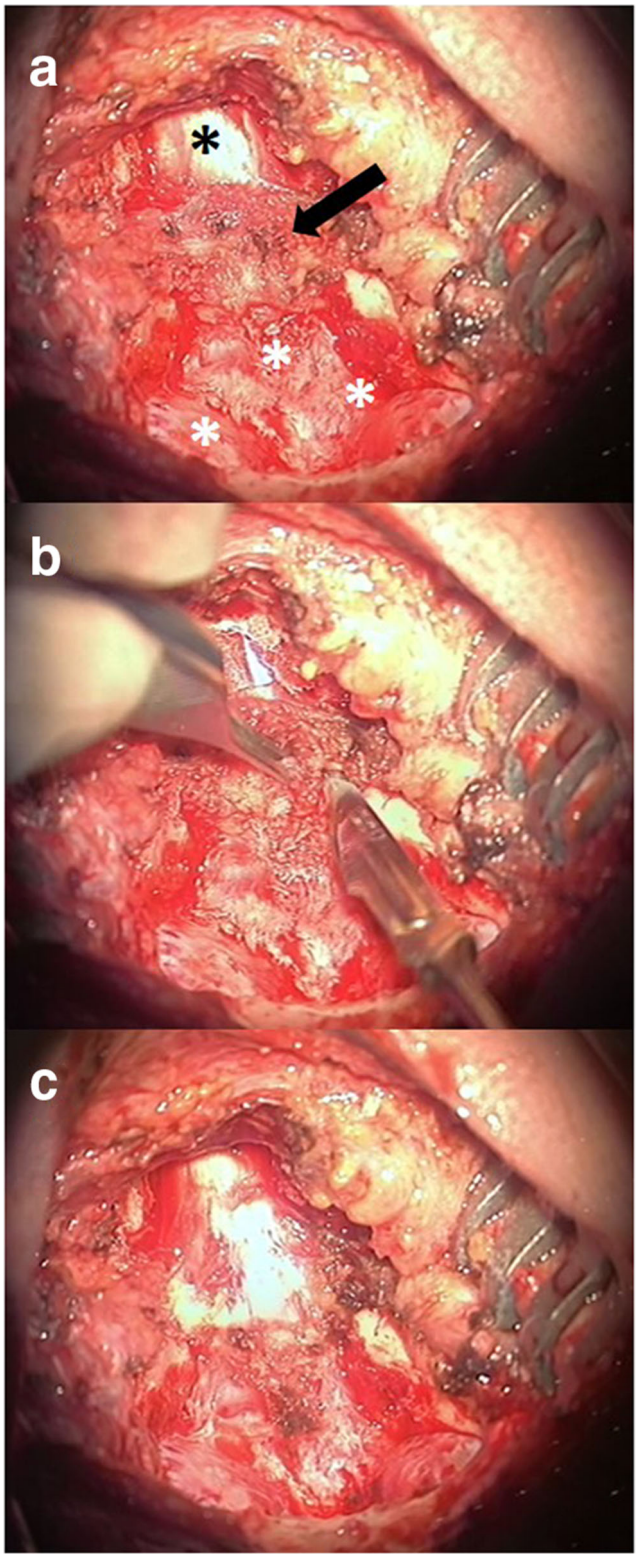

Fig. 1 **A** PFBD after midline suboccipital craniectomy (white asterisks) and C1 laminectomy (black asterisk). Note the dense epidural ring below the foramen magnum (arrow). **B** Sharp dissection of the epidural ring. **C** Intraoperative view after its removal (the dural sac of the bulbo-cervical junction appears)

indeed, reported on the improvement of the CSF flow dorsal to the cerebellar tonsils, at intraoperative MRI, just after the prone position (before starting surgery) in 14 out of their 15 adult patients [5]. However, this experience has not been replicated elsewhere yet and it seems to be in contrast with the obvious intraoperative evidence of the crowded PCF in children in spite of the prone position.

Outcome analysis in the clinical series

PFBD resulted to be extremely effective in reducing the CIM symptoms (success rate 86–97%) and preventing the risk of reoperation (risk rate 6–7%) already in historical series [19, 25, 30, 47, 77]. The reasons of such an excellent result are mainly two: (1) a follow-up not enough long to exclude a late recurrence of symptoms, the average ranging from less than 1 to 4.3 years in the aforementioned series and (2) a careful selection of the patients (often without severe tonsillar ectopia/syringomyelia) to treat by PFBD. The second reason seems to be predominant. Indeed, our recent experience, based on the long-term outcome of a series of 42 children undergoing PFBD (mean follow-up 11.3 years) [44], confirms the results that we obtained in the previous series of 30 children (mean follow-up 4.3 years) published about 10 years ago [43]. For the patient selection, we considered for PFBD only symptomatic children with tonsillar herniation above C2 and/or with syringomyelia thickened less than 5 mm. The goal of the recent study was to provide an update about the old series taking into account the long-term results and the use of a scoring system to standardize them (Chicago Chiari Outcome Scale, CCOS) [1]. Accordingly, a complete resolution of the clinical picture was achieved in 76.5% of cases (vs 43.5% in the previous series), with a 98% overall improvement rate (vs 93%) and the need of reoperation in 7% of cases (vs 6.7%). The CCOS showed an excellent outcome in 69.5% of patients, a functional outcome in 28.5% and an impaired outcome only in 2%. As far as the CCOS is concerned, good results have been confirmed also by other authors, with scores comparable to PFBDD [33].

These observations have been confirmed by recent series based either on PFBD alone or on the comparison with PFBDD [9, 13, 18, 27, 33, 34, 37, 39, 68]. More in details: (1) Although the absence of standardized methods to assess the outcome should be taken into account as an important limitation, the success rate of PFBD in giving symptoms improvement/resolution remains high, ranging from 65 to 90%. In the largest pediatric series published so far (156 patients treated by PFBD), reported by Kenney and coworkers, 90% of children experienced a favorable clinical outcome (especially when the tonsillar ectopia was less than 8 mm) after a 32-month mean follow-up [27]. The mean duration of the hospital stay was 2 days and no significant complications occurred. Tonsils below C2 ($p = 0.037$) and motor weakness at diagnosis (low statistical significance, $p = 0.075$) were the

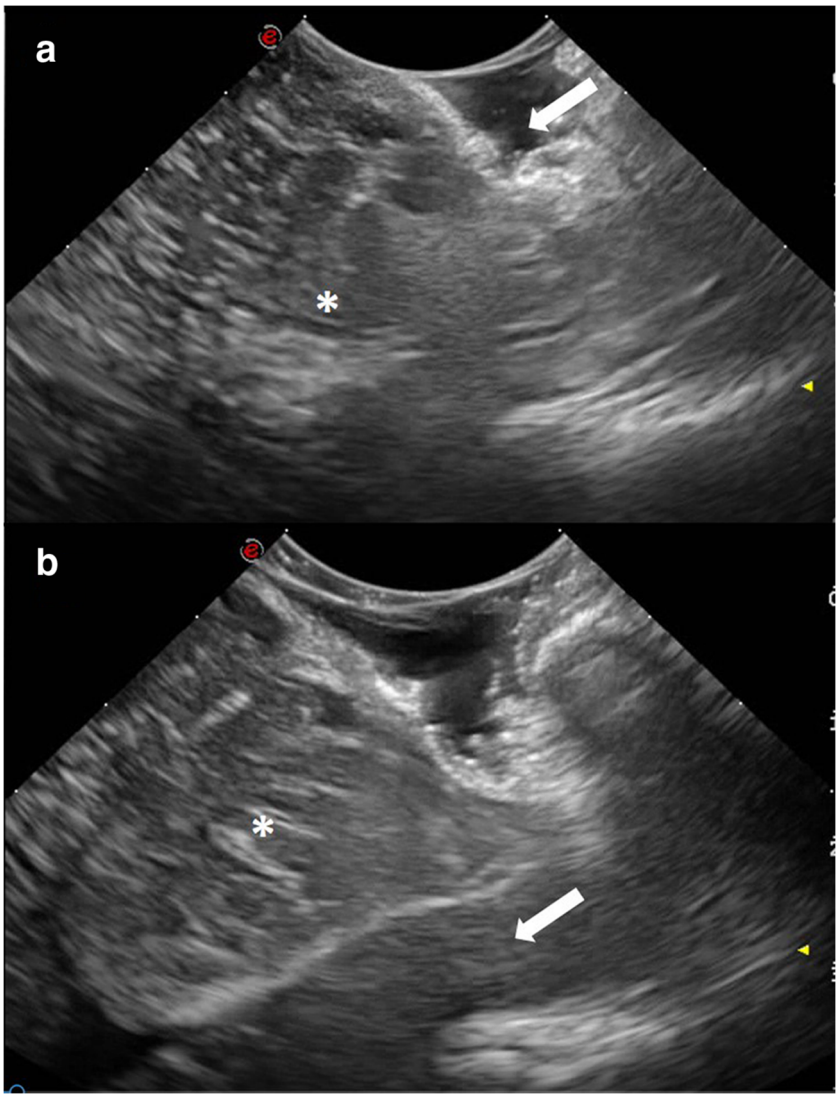

Fig. 2 Intraoperative US of the case in Fig. 1. **A** Before the removal of the epidural ring: the tonsil is not clearly recognizable (asterisk) and the CSF space is opened only above (arrow). **B** After the further epidural decompression, the tonsil clearly appears (asterisk) and the CSF is visible also below the tonsil (arrow)

main predictors for surgery failure (reoperation). (2) The postoperative re-expansion of the cisterna magna is a further as well as expected criteria to predict the success of PFBD. According to the observation of Quon et al., the greater the increase of the cisterna magna, the higher the rate of postoperative improvement [59]. Such a finding was more evident in the patients with the smaller cisterna magna preoperatively. (3) The risk of reoperation with PFBD is higher than with PFBDD. Usually, it is around 7–15% but it can reach even a 50% rate in series where patients with syringomyelia are included [68]. (4) It is worth noting that the poor improvement of the syringomyelia, in some studies, could depend on the still short follow-up. In fact, in series where the follow-up was long enough (> 5 years) good results were recorded also about the syringomyelia improvement/disappearance [30, 34, 44]. This could be because the indirect cisterna magna recreation needs a long time to completely restore the correct CSF dynamics (Fig. 3). (5) The length of hospitalization and the costs of PFBD are significantly lower than PFBDD (unless syringomyelia is present), as demonstrated by Limonadi et al. [37]. The authors calculated an advantage of PFBD over PFBDD in terms of mean operative time (99 vs 169 min, $p < 0.001$), total operating room time (166 vs 249 min, $p < 0.001$), duration of hospitalization (3 vs 3.75 days, $p < 0.05$), perioperative charges (3615 vs 5538 $, $p < 0.001$), and overall hospital charges (7705 vs 9759 $, $p < 0.001$).

PCF bony decompression with duraplasty (PFBDD)

Rationale, indications, and techniques

The rationale of PFBDD is to re-create directly the cisterna magna by means of an expansion duraplasty and, if needed, removal of arachnoid adhesions. Indeed, the re-expansion of the cisterna magna and the relaxation of the PCF content seems to be the best radiological predictor of postoperative

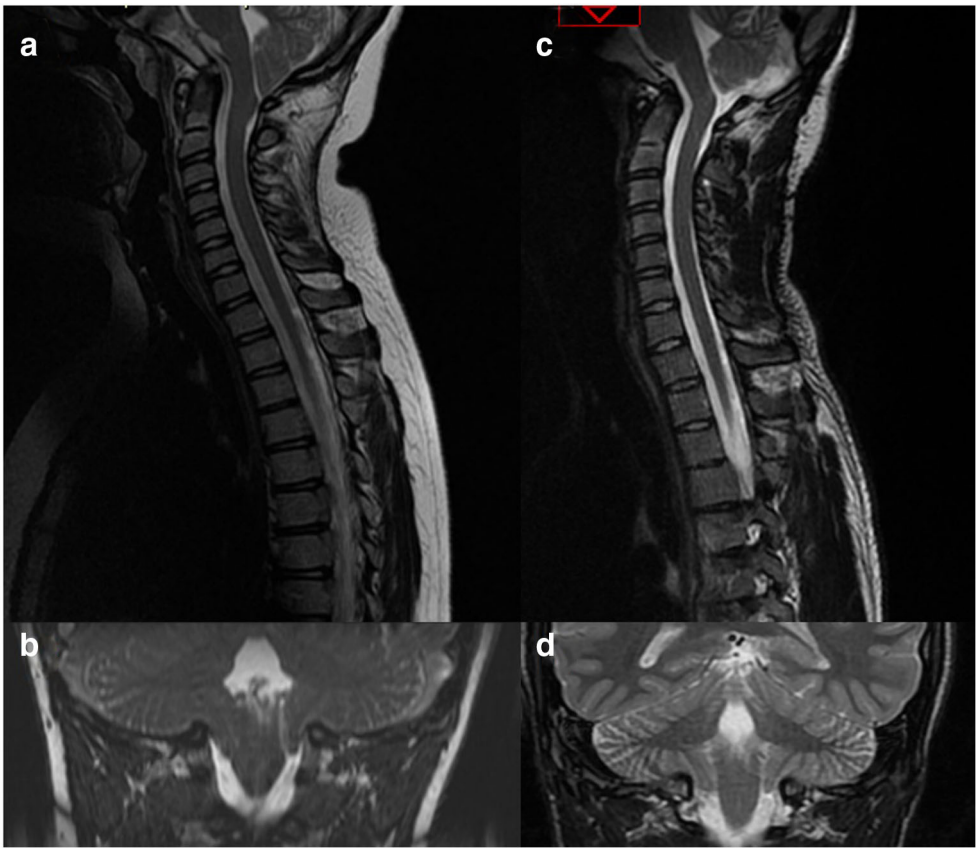

Fig. 3 **A**, **B** preoperative T2 sagittal and coronal MRI of a 10-year-old young boy with CIM without syringomyelia; **C**, **D**: postoperative MRI, 4 years after PFBD, showing the late reopening of the cisterna magna and the normalization of the position of the cerebellar tonsils

symptom improvement [59]. According to the volumetric analysis of the PCF/CSF spaces performed preoperatively and postoperatively by Khalsa and coworkers on 42 children undergoing PFBD/PFBDD, a statistically significant postoperative increase of the PCF and CSF spaces volume was associated to a significant improvement of clinical symptoms and a significant reduction of the tonsillar descent [28]. Moreover, a statistically significant increase of the caudal portion of the PCF volume (calculated below the line drawn between the cranial tip of the clivus anteriorly and the torcula posteriorly) corresponded to a significant reduction of both syringomyelia and cervicomedullary kinking. As expected, PFBDD was able to increase the volume of both PCF and cisterna magna significantly more than PFBD. Nevertheless, although children who underwent PFBDD showed a better clinical outcome, the difference with the clinical outcome of children treated by PFBD was not statistically relevant.

PFBDD is used prevalently in adult subjects, despite the higher rate of complication and the longer operation time than PFBD, thanks to the better outcome and the lower recurrence rates [15, 21, 62]. Based on the aforementioned considerations, PFBDD should be poorly used in children, where PFBD warrants very good results with shorter surgical times, reduced hospitalization length, and lower costs [33, 39, 51, 65]. However, according to the published clinical series, PFBDD is commonly used in children too, mainly because of the CIM-associated syringomyelia or the severity of the preoperative symptoms or, probably, just because of the surgeon's preference [12, 18, 33, 34, 39, 61, 65, 68].

The technique is quite well standardized, consisting of a PFBD plus a capacious and watertight duraplasty. The graft used for the duraplasty represents one of the well-known variants among the different centers. The ideal graft should be non-inflammatory, non-immunogenic, non-toxic, easy to be integrated by the native tissues, flexible, and strong enough to be easily suturable and watertight, durable and, possibly, cheap. Autologous tissues (pericranium, fascia lata, ligamentum nuchae) satisfy these required characteristics; however, in children, they are not routinely used because they are often insufficiently available for the entire patch other than burdened by longer surgical times, suboptimal surgical results, increased pain and scarring, and harvest-site complications [2, 32, 55, 69, 70]. Therefore, a rich spectrum of non-autologous dural graft have become available (allografts, xenografts, synthetic dural substitutes) which, in turn, carry several types of complications, like infection (bacteria, viruses, prions), foreign body reaction, systemic immune response, premature graft dissolution, pseudomeningocele, wound healing problems, and high costs [4, 11, 16, 42, 50, 52, 63]. Apart from the specific complications (higher risk with non-autologous grafts), no significant differences between autologous and non-autologous grafts have been pointed out as far as general

complications (namely, CSF leakage) and clinical outcome are concerned, so that the use of autologous patches is encouraged in PFBDD (especially in adults), when feasible [78]. Previous experiences with the posterior atlanto-occipital membrane [70] or pericranium [42] and recent experiences with the ligamentum nuchae [10] seems to support this strategy also in children in selected cases. In our center, we try to use autologous tissues as much as possible, sometimes adding a small synthetic dural patch if they are not enough (Fig. 4).

Several other technical variants, on the other side, have been designed to increase the effectiveness of PFBDD or, in particular, to decrease its rate of complications (which are the main disadvantage of PFBDD). For this purpose, for example, some authors proposed to increase the bony decompression with an extreme lateral suboccipital craniectomy (out of the level of the occipital condyles) coupled with duraplasty with autologous pericranium without manipulation of the arachnoid (in adults) [66]. Other authors confirmed the augmented risk of complications (without increased rate of success) in case of arachnoid manipulation (with or without coagulation of the tonsils), so discouraging such a procedure unless dense arachnoid adhesions and/or unsatisfying duraplasty occurs [72]. With regard to PFBDD without dural closure proposed several years ago [74], some authors noticed that this option in children is burdened by both worse clinical outcome and raised risk of complications if compared with PFBDD with dural closure and PFBD [18]. Finally, the dural splitting is a further, commonly used alternative technique, proposed to reduce the dural manipulation. Really, it is a variant of PFBD rather than PFBDD, consisting in the delamination of the outer layer of the dura mater realized to "weak" the dura, thus favoring its expansion without opening. Different sub-variants have been realized: only linear incisions of the external dural layer, Y-shaped opening of the external layer, opening and removal of the external layer, suture of the internal layer to the ligamentun nucahe (to enhance the expansion effect), and so on. Thanks to the elastic properties of the dura, this technique has been successfully used in children, where it can ensure similar results than PFBDD but with significantly lower surgical times, complications and costs [37, 39]. The experience with dural splitting has been replicated also in adults, obtaining results inferior than in children or with PFBDD, especially when syringomyelia was present [8, 13, 54].

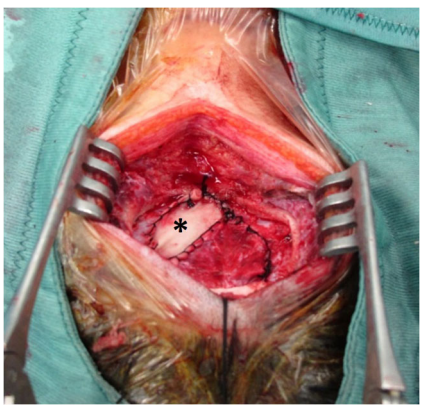

Fig. 4 Duraplasty after suboccipital decompression. In this case, a small synthetic dural patch (asterisk) has been added to the autologous dura to make the duraplasty more capacious and to reduce the use of non-autologous material

Outcome analysis in the clinical series

According to historical series, PFBDD warrants a rate of improvement of preoperative symptoms as high as 46% to 83% [23, 36, 47, 71], reaching even 85–97% of success when adjunctive surgical maneuvers (today poorly used) were utilized (tonsils resection, plugging of the obex, shunting of the IV ventricle, and/or syrinx) [3, 23, 56, 71]. Consequently, a relatively low rate of re-operations was reported (7–14%). Such a good outcome is confirmed by more recent studies, either based on a single PFBDD series [12, 16, 24, 41, 64, 66, 72] or on the comparison between PFBD and PFBDD [9, 13, 18, 33, 34, 39, 48, 51, 54, 65], with a reduction of the mean re-operation rate (3–5%). Overall, they show a good early post-operative improvement of CIM symptoms (57%), their relevant improvement/disappearance in the short-term follow-up in the great majority of cases (> 95%) and a stabilization around 85% of success (range 67–93%) in the long-term period. The clinical improvement measured by the Karnofsky disability scale is relevant as well, at least in adults, where it has been used as method for the outcome assessment [67].

As expected, the typical CIM headache shows a greater post-operative improvement than other headaches, thus stressing the importance of an accurate preoperative evaluation of the headache, based on the criteria of the International Headache Society [61]. As far as the associated syringomyelia is concerned, an early decrease (3–6 months after surgery) is reported in 65–80% of cases, while a late important decrease is observed in about 85% of cases (resolution in about 55–60% of cases) with a certain amount of recurrence in case of long-term follow-up [64].

The reason of such satisfying results depends on the direct enlargement of CSF spaces of the posterior fossa (Fig. 5). In addition, however, a further factor, which is the tonsil pulsatility, should be considered. Radmanesh and coworkers, indeed, added the cardiac-gated true FISP sequence and the phase contrast CSF flow imaging to the routine preoperative and postoperative MRI of 22 children undergoing surgery for CIM [60]. Both a qualitative and quantitative assessment of the tonsil pulsations were performed together with a qualitative evaluation of the peritonsillar CSF flow. After surgery, a general reduction of the degree of tonsil pulsatility was recorded together with an improvement of the peritonsillar CSF flow. No statistical differences between PFBD and PFBDD were found as far as the latter is concerned. Instead, the decrease of tonsil pulsatility was statistically superior in PFBDD, both qualitatively and quantitatively, if compared with PFBD.

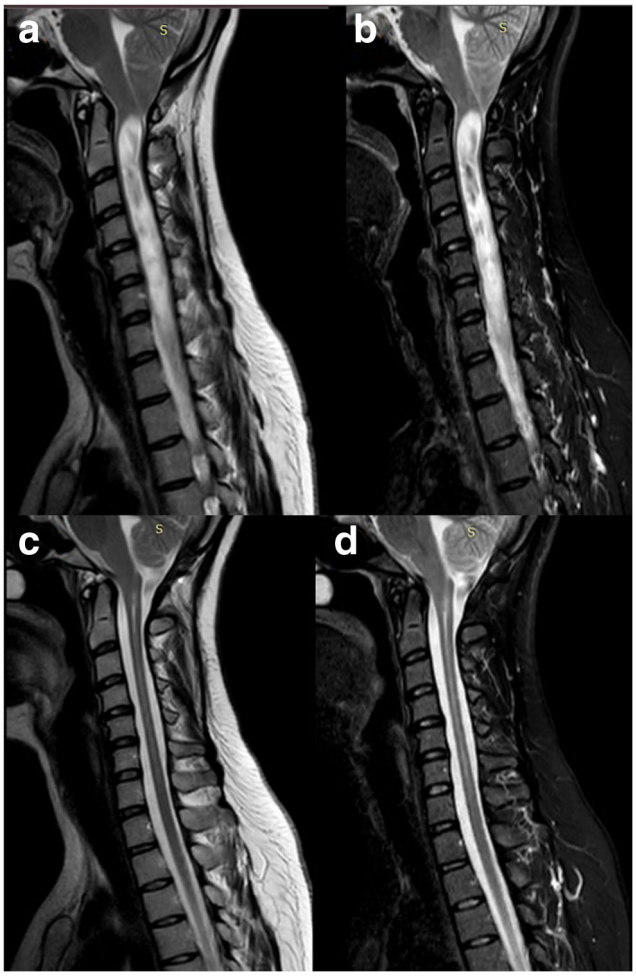

Fig. 5 **A**, **B** preoperative T2 sagittal MRI of a 14-year-old young girl with CIM and cervico-dorsal syringomyelia; **C**, **D**: postoperative MRI, 6 months after PFBDD, showing the reopening of the CSF spaces, the upper position of the cerebellar tonsils and the relevant improvement of the syringomyelia

As mentioned, the main disadvantage of PFBDD results from its relatively high rate of complications, especially if compared with PFBD. The surgical mortality rate is very low (< 1%) but not nil [29], as generally reported in PFBD. In spite of the usually absent major morbidity, the PFBDD complication rate is around 15%, ranging from 2 to 40% but increasing up to 70% in small series [9, 12, 13, 18, 24, 33, 39, 65]. CSF leakage, symptomatic pseudomeningocele, aseptic meningitis, hydrocephalus, and infection are the most common ones. It is worth noting that these complications not infrequently require a re-operation for their management.

PFBD vs PFBDD: review of the meta-analyses of the literature

The debate about PFBD vs PFBDD is so present that numerous meta-analyses continue to be published, even occupying two close issues of the same year in international journals [7, 38]. Why? In addition to the answers formulated in the background (unsatisfying outcome possibly resulting from a wrong indication, missing management guidelines, missing standardized outcome measurement), some other considerations can be done. Actually, in spite of the definition of "rare" disease, the diagnosis of CIM is more and more common in the clinical practice. Therefore, the large amount of patients to deal with justifies the efforts to find the optimal solution for such an increasing population. As a consequence, several authors are trying to test PFBD and PFBDD in larger and larger series to update their results. Moreover, the problem to differentiate the subpopulation of CIM patients still persists and then the need to find the optimal candidate for PFBD or PFBDD is still felt. Indeed, an important limitation of many of the aforementioned studies comparing PFBD and PFBDD is that patients with different clinical and radiological characteristics (and, sometimes, ages) are treated by these two different techniques. In addition, the poor weight of the patients' characteristics (age, type of symptoms, PCF size and morphometry, syrinx presence and extension) on the postoperative outcome [17] lead many authors to further investigate the role of the surgical technique on the final outcome. Finally, a last, relevant argument in favor of meta-analyses is the lack of prospective studies, which could validate the results reported so far [22].

In the last decade, eight meta-analyses have been published on the topic of the present article [7, 14, 15, 22, 38, 40, 75, 79]. A summary of their results is reported on Table 1. The main limitations concerning the reviewed articles were lack of standardization of the surgical techniques, lack of details on the surgical indication (assignment of patients to a specific surgical technique), lack of standardized outcome measurement (not blinded and often subjective), follow-up not ever specified, often missing level I or IIa evidence studies. Three meta-analyses have been specifically dedicated to the problem of PFBD vs PFBDD in children. The first one, provided by Durham and Fjeld-Olenec in 2008, was based on seven studies (five retrospective and two prospective) meeting all inclusion criteria and reporting on 582 children, 266 treated by PFBD and 316 with PFBDD [14]. PFBDD was superior in terms of clinical improvement and syringomyelia decrease but a significant difference among the two techniques was detected as far as only the need of reoperation was concerned (12.6% with PFBD vs 2.1% with PFBDD). Similarly, no statistical differences were reported among complications (wound infection, postoperative occipital neuralgia, bleeding), although they were less common with PFBD, except for CSF-related complications (1.8% with PFBD vs 18.5% with PFBDD). The second review, provided by Hankinson et al. [22], is based on the same studies than the previous one. Finally, the third review, realized by Lu et al., collected 3455 children, 1963 treated by means of PFBD and 1492 by means of PFBDD, from 12 studies [40]. A statistically significant

Table 1 Synopsis of the meta-analyses of the literature (the review by Hankinson et al. [22] is not reported here because, as far as PFBD vs PFBDD is concerned, it is based on the same studies than Durham and Fjeld-Olenec [14])

Authors/year (ref)	No. of papers	No of patients	Series' characteristics	Outcome	Complications
Durham and Fjeld-Olenec (2008) [14]	7	582	≤ 18-year-old Syrinx 28.5% PFBD 266 cases PFBDD 316 cases FU 2–120 months	PFBD Reoperation12.6% CI 64.6% SI 56.3% PFBDD Reoperation 2.1% CI 78.6% SI 87%	PFBD CSF-related 1.8% Infection 0.9% Neuralgia 1.8% Bleeding-related 0% PFBDD CSF-related 18.5% Infection 3.7% Neuralgia 1.5% Bleeding-related 0.7%
Forander et al. (2014) [15]	12	537	> 18-year-old Syrinx 62% PFBD 51 cases PFBDD 486 cases FU 3–89 months	PFBD Reoperation11% CI 78% SI 74% PFBDD Reoperation 2% CI 77% SI 78%	PFBD CSF-related 0% PFBDD CSF-related 7%
Zhao et al. (2016) [79]	18	1242 (including also tonsils coagulation and shunt)	< and > 18-year-old Syrinx 66.2% PFBD 216 cases PFBDD 721 cases FU: < and > 3 months	PFBD CI 73.6% SI 77.1% PFBDD CI 82.2% SI 83.3%	PFBD Recurrence 10.5% Overall comp. 4.5% PFBDD Recurrence 7.7% Overall comp. 20.7%
Lu et al. (2017) [40]	12	3455	≤ 18-year-old Syrinx 4.3% PFBD 1963 cases PFBDD 1492 cases FU < 1–55 months	PFBD Hospital stay 3.79 days Reoperation 2% CI 72.3% SI 60.9% ScI 44.4% PFBDD Hospital stay 4.46 days Reoperation 3.5% CI 88.1% SI 73.7% ScI 43.8%	PFBD CSF-related 9.9% Infection 1% Overall 11.8% PFBDD CSF-related 12.2% Infection 5% Overall 15.6%
Xu et al. (2017) [75]	12	841	< and > 18-year-old PFBD 417 cases PFBDD 424 cases FU > 6 months (max mean FU 3.6 years)	PFBD Operative time 97 min CI 70% PFBDD Operative time 112 min CI 86.3%	PFBD CSF-related 0% Infection 4.7% Meningitis 0% Pseudomenignoc. 0% Neurological 1.6% Overall 1% PFBDD CSF-related 8%

Table 1 (continued)

Authors/year (ref)	No. of papers	No of patients	Series' characteristics	Outcome	Complications
					Infection 5.1%
					Meningitis 6.8%
					Pseudomenignoc. 3%
					Neurological 8.7%
					Overall 5.7%
Chai et al. (2018) [7]	14	3666	< and > 18-year-old PFBD 2035 cases PFBDD 1631 cases FU 1–117 months	Reoperation and CI: no significant differences between PFBD and PFBDD SI: PFBDD better than PFBD	CSF-leak and aseptic meningitis: higher risk in PFBDD than PFBD Wound infection: no differences between PFBD and pFBDD
Lin W et al. (2018) [38]	13	3481	< and > 18-year-old PFBD 1593 cases PFBDD 1056 cases FU > 6 months (max mean FU 44 months)	Operative times: PFBDD longer than PFBD Recurrence: PFBD higher risk than PFBDD CI: no differences SI: PFBDD better than PFBD	Overall complications, CSF-related, aseptic meningitis, pseudomeningocele: PFBDD higher risk than PFBD Wound complications: no differences

FU follow-up, *CI* clinical improvement, *SI* syrinx improvement, *ScI* scoliosis improvement

difference between the two techniques was noticed about clinical improvement (72.3% of cases with PFBD vs 88.1% with PFBDD), length of hospital stay (3.79 days with PFBD vs 4.46 days with PFBDD), overall incidence of postoperative complications (11.8% with PFBD vs 15.6% with PFBDD), CSF-related complications (9.9% with PFBD vs 12.2 with PFBDD), and infections (1% with PFBD vs 5% with PFBDD). As shown, all findings were in favor of PFBD except for the clinical improvement. No relevant differences were found as far as estimated blood loss, revision surgery, syrinx, and scoliosis improvement were concerned. The remaining meta-analyses, either based on adult [15] or mixed series [7, 38, 75, 79], largely confirm that: (1) The main advantages of PFBD are the short duration of surgery and hospital stay, the very low complication risk, and the low costs, while its main disadvantage is the risk of reoperation resulting from the missing clinical improvement. (2) On the other hand, the pros of PFBD are the good rate of clinical improvement and the low rate of revision surgery, while the cons concern the risk of complications with subsequent lengthening of the hospital stay and increased costs. (3) No significant differences between children and adults occur as far as the clinical improvement and the syringomyelia decrease are concerned (according to the studies specifically addressing this comparison) [7, 38]. However, adults have a lower risk of re-operation, although no obvious associations were found in children. The rate of preoperative syringomyelia is usually higher in adults than in children.

These findings have been confirmed by the prospective study recently published by Jiang et al. on 82 adolescents (mean age 13.8 years), of whom 40 underwent PFBD and 42 PFBDD [26]. The clinical outcome and the syrinx improvement were similar between the two techniques. PFBDD, as expected, showed longer operation times and longer hospital stay and higher risk of complication. Therefore, the authors advised PFBD as the best option in children with CIM. Similar results and conclusions were obtained by Limonadi and Selden about 15 years before on 24 children [37].

Conclusions

This review of the literature definitively confirms that the success of the management of CIM, with or without syringomyelia, depends on a correct indication to surgery and on a patient-tailored choice of the surgical technique rather than on the surgical technique itself. Actually, both PFBD and PFBDD have been proved to be effective and to be burdened by some disadvantages at the same time. PFBD results to be the best treatment for children, especially if they are without syringomyelia, although the presence of syrinx is not a contraindication to PFBD. On the other hand, PFBDD seems to be indicated in adults, in particular, if they harbor a

syringomyelia. A center dedicated to CIM should be able to deal with both techniques and to offer the best option to each patient. A final word on this topic will be said only after a randomized, controlled trial on a homogeneous population.

Compliance with ethical standards

Conflict of interest No conflict of interest to disclose.

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