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SPECIAL ANNUAL ISSUE

Which neuroimaging techniques are really needed in Chiari I? A short guide for radiologists and clinicians

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Abstract

Purpose To describe the most appropriate techniques and suggested protocols meant to address the various scenarios that clinicians and pediatric neurosurgeons may face in their day-to-day practice connected with Chiari I.

Methods Current literature related to image indications and findings in Chiari I has been reviewed. The authors focused on both standard and advanced techniques for clinical diagnosis and preoperative planning purposes.

Discussion and conclusion The complexity of providing neuroimaging guidelines for children investigated for Chiari I lies in defining the most appropriate neuroradiology tool to approach what is in fact a very heterogeneous condition with different etiopathogenetic mechanisms and associated abnormalities. Other variables that may influence the diagnostic strategy include the age of the patient, the presence of additional pathological conditions, the type of presenting symptoms, and the indication for surgical or conservative management. Although the average age at time of diagnosis is 10 years, the initial diagnosis may be done at any age, and the referral for neuroradiology workup may come from general practitioners/pediatricians, orthopedic surgeons, and endocrinologists following various baseline investigations including plain x-rays of skull and spine and/or CT head and/or MRI brain and spine.

Keywords Chiari I · Magnetic resonance imaging · Computed tomography · Surgical planning · Craniovertebral junction · Tonsils herniation

Introduction

So-called Chiari I (also known as Chiari type 1 malformation, Chiari I deformity, and low cerebellar tonsils) is characterized by clinical signs and symptoms related to a pathological tonsillar herniation below the foramen magnum, with consequent deformation of the hindbrain [1, 2]. Historically, the radiological diagnosis was made by measuring how far the tonsils protrude below the line drawn from the basion to the opisthion and measuring the inferior most part of the tonsils. The literature is rich in different thresholds for a definitive diagnosis of Chiari I; however, in adults any tonsillar herniation beyond 5 mm below the foramen magnum is generally considered suspicious and warrants further clinical, neuroradiological, and neurophysiological investigations [2].

The first important point for the radiologist is that this is somehow arbitrary and the presence of symptoms, associated spinal syrinx, and pointed shape ("peg-like") of the tonsils are more indicative of pathological condition than the extent of the descent per se.

Furthermore, in pediatric patients the diagnosis is even more challenging because the position of cerebellar tonsils is known to vary with age [3], with cerebellar tonsils that may reach their lowest point between 5 and 15 years of age, and then progressively ascend coming to rest at the level of the foramen magnum.

Overall, Chiari I is thought to be a group of different entities with similar cerebellar neuroimaging finding on midline MRI and discrepancy between content (brain parenchyma) and container (posterior fossa/cranial vault), and this is why the term "Chiari I deformity" rather than "malformation" has been recently proposed at least for some of the cases of Chiari I [2]. Beside all these situations, tonsillar herniation is also increasingly found by neuroimaging studies as an incidental

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finding in asymptomatic children simply because of the increasing prevalence of diagnostic imaging. In fact, on the one hand, the availability of better diagnostic tools allows for early identification of pathological findings and justifies the increasing number of children diagnosed with Chiari I worldwide, and on the other hand it also resulted in a growing number of futile referrals to pediatric neurosurgeons and neuroradiologists for additional scans.

Given the above, the complexity of providing neuroimaging guidelines for children investigated for Chiari I lies in defining the most appropriate neuroradiology tool to approach very different situations. The variables that may influence our strategy include the age of the patient, the presence of additional pathological conditions, the type of presenting symptoms, and the indication for surgical or conservative management. In this review, we will describe the most appropriate techniques and suggested protocols meant to address the various scenarios that clinicians and pediatric neurosurgeons may face in their day-to-day practice connected with Chiari I.

Teaching points, proposed protocols, and useful resources will be also highlighted.

Pathogenesis

The pathogenesis is variable, and details on it are outside the scope of this manuscript. However, it is relevant for radiologists to distinguish 4 groups of Chiari I patients with different pathogeneses and associated different roles of images:

1) Abnormal skull base (e.g., short clivus)
2) Cervical segmentation anomalies (such as patients with Klippel-Feil)
3) Small cranial vault and/or posterior fossa and consequent overcrowding
4) Excessive brain tissue

In view of these differences in the etiopathogenesis, Prof. Raybaud and colleagues suggested the use of "Chiari I deformity" for groups 1–2 and "tonsillar herniation" for groups 3–4 [2]. This distinction is not only semantic but also has relevance in terms of management and images.

Conventional X-ray investigation

Conventional X-rays are not indicated for diagnosis of Chiari I being not able to visualize the brain parenchyma directly. Nevertheless, the bony abnormalities associated particularly with group 1 and 2 Chiari deformity can be seen on radiographs.

The short clivus and abnormal skull base can be diagnosed on X-rays of the skull or cervical spine and MRI spine may be suggested in some of these cases to check for the position of the tonsils and other abnormal brain and spine findings.

Plain X-rays of the whole spine with long cassette are very common for children and adolescent investigated for spinal deformities; in these cases,

- segmentation abnormalities such as hemivertebra can be seen on conventional X-rays (Fig. 1)
- adolescents with idiopathic scoliosis (which may often be associated with Chiari 1) can be widely studied on plan films. In those patients, specific calculations of spinal balance are routinely performed, including sagittal vertical axis, lumbar lordosis, spinofemoral axis, Cobb angles, and obviously an assessment of the Risser's sign, a measure of the growth left in the spine which may help to determine the potential for progression of scoliosis hence guiding the management choices

CT head

The importance of the plain CT head is often limited in elective assessment of patients with Chiari I, obviously because of the paramount mandate to avoid unnecessary irradiation in developing children. Nonetheless, it is worth remembering that often this neuroimaging investigation represents the first step in the radiological workup of patients with clinical signs and symptoms such as headache, raised intracranial pressure, or other symptoms that may have tonsillar descent/Chiari I as related or incidental finding.

It is important to note, however, that the predictive accuracy of CT in diagnosis of Chiari I is not very high and, in a

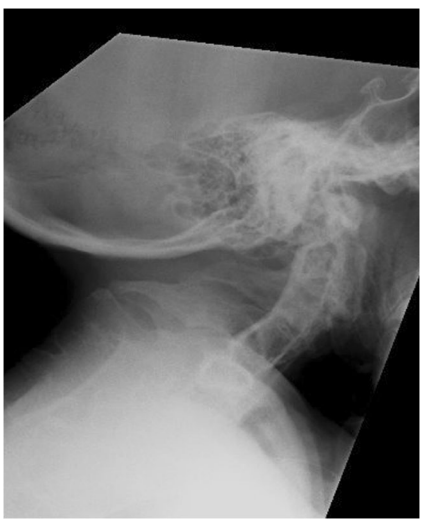

Fig. 1 Lateral plain film in a 15-year-old patient with Klippel-Feil shows fusion of multiple vertebrae bodies in the cervical spine

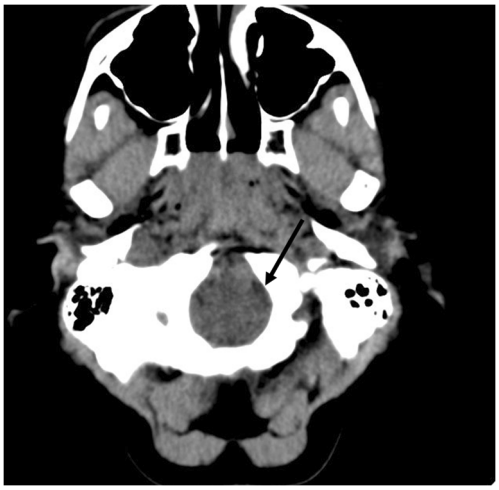

Fig. 2 Axial CT at the level of the foramen magnum showing low position of the cerebellar tonsils with crowding of the posterior fossa structures at this level (arrow)

prospective study, only 51.4% of Chiari I suspected on CT were confirmed on MRI [4] (Fig. 2). In this regard, it is very important to acquire a volumetric CT and to reformat the images on the sagittal plane to increase the rate of diagnosis.

Moreover, CT is the investigation of choice for further characterization of bone structures in skull base and craniocervical junction abnormalities such as platybasia, basilar invagination, assimilation of the arch of C1, and instability [5, 6]. The possible combination of each of those conditions in a significant proportion of Chiari I patients is well established [7–9].

In those scenarios, CT scan with 3D reconstruction and bone windows are of paramount importance in carefully assessing sutures in children considered for surgery or already operated (Fig. 3).

CT venograms may be useful in demonstrating anomalous intracranial venous anatomy which is a well-described phenomenon in patients with low cerebellar tonsils in a context of multisuture synostosis, and may have potential significance with respect to surgical morbidity.

The importance of assessment of the venous circulation at the skull base was recently confirmed in a Canadian study which reported a significant association of venous anomalies with Chiari I, elevated intracranial pressure, shunted hydrocephalus, and sleep apnea. In this cohort, an intraoperative complication rate of 7.3% was recorded, all due to venous anomalies [10]. So in this patient, CT venography or MR venography (CTV and MRV) should be considered in preoperative planning [11].

Figure 4 shows a patient with multisutural craniosynostosis and low cerebellar tonsils (group 3 Chiari I).

Teaching points for the use of X-rays and CT in children with Chiari I/low cerebellar tonsils

X-rays: X-rays do not show the brain parenchyma and are not indicated as a diagnostic tool for Chiari I; they can however visualize associated skull base and cervical spine abnormalities.

CT: CT can visualize the brain parenchyma and thus can be used to diagnose low cerebellar tonsils with a sensitivity that is inferior to MRI. It allows exquisite demonstration of the bony structures and is useful for preoperative planning (multiparametric reformats and 3D volume rendering).

Conventional and advanced MRI techniques

MRI is the modality of choice for the radiological diagnosis of Chiari I deformity and low cerebellar tonsils.

Despite the limits of MRI in emergency settings, especially in children requiring sedation or general anesthesia, conventional T1- and T2-weighted image (WI) MRI sequences can provide the highest-quality images regarding brain anatomy

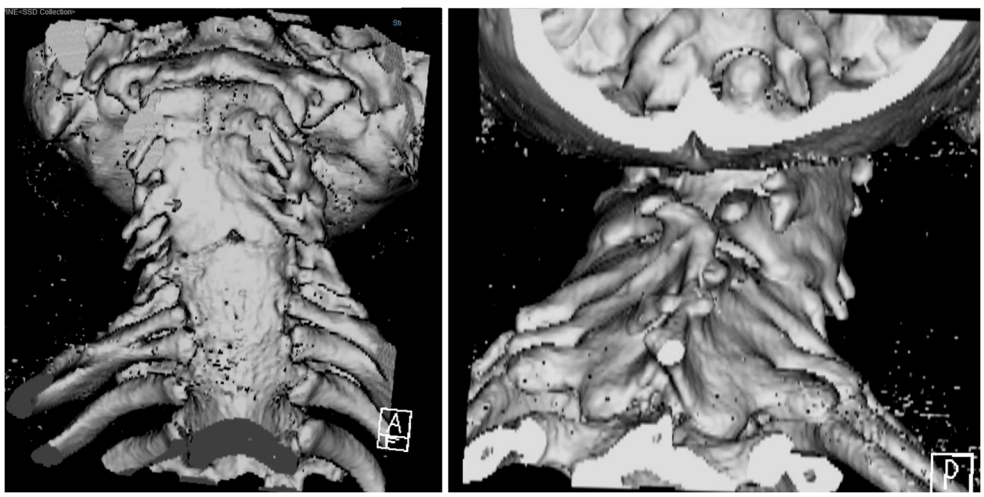

Fig. 3 AP (left) and PA (right) view of CT 3D volume rendering reformat of the cervical spine in a child with Klippel-Feil showing fusion of multiple cervical vertebral bodies

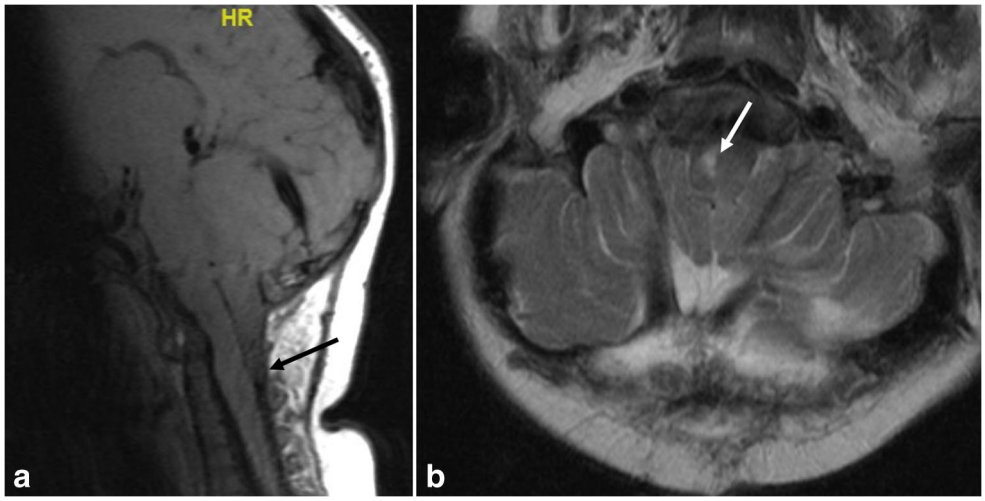

Fig. 4 **a** Sagittal T1 WI in a patient with multisutural craniosynostosis showing very low position of the cerebellar tonsils at the level of C2 (arrow). **b** Axial T2 WI in the same patient shows an area of abnormal signal at the level of the bulbo-medullary junction in keeping with cord edema as consequence of the crowding of the foramen magnum. Note the dysmorphic appearance of the posterior fossa which is related to syndromic craniosynostosis. Chiari I associated with small size posterior fossa is classified as type 3 (i.e., low cerebellar tonsils) according to Poretti and colleagues

and its variation from the physiological range. Usually, MRI is the elective investigation of choice performed in children suspected to have brain pathologies and, regardless of the indication for acquiring those scans, Chiari I may be found as an incidental finding or as a part of the pathological spectrum responsible for the symptoms.

Just to provide an exemplificative example, the presence of Chiari I represented over half (61%) of the brain anomalies identified by Hukki et al. in MRI scans performed in children with non-syndromic single suture craniosynostosis scheduled for cranial vault remodeling surgery [12].

Furthermore, conventional MRI can be used to assess the degree of cord compression in patients with Chiari I [7, 13–15]. Particularly, T2 WI shows intramedullary edema and T1 WI can be used to measure the narrowing of the craniovertebral junction (CVJ) (Fig. 4b).

Proposed MRI cervical spinal protocol for the study of CVJ in patients with Chiari I deformity/low cerebellar tonsils (Fig. 5)

- Sagittal spine-echo T1 WI (for the study of the anatomical position of the cerebellar tonsils and size of the foramen magnum)
- Axial and sagittal spin echo T2 WI (for the study of the cord signal—i.e., diagnosis of spinal cord damage)
- Axial gradient echo T2 (T2*) as a valid alternative if the spin echo axial T2 WI is affected by artifacts (this sequence is widely used in multiple sclerosis patients to visualize small cervical cord lesions)
- Coronal Fat-Sat region of interest in case of diastematomyelia, vertebral segmentation anomalies, and scoliosis
- Post-gadolinium sequences in case of syrinx (on first scan only to exclude associated tumor)
- Optional sequence as a 3D steady-state T2 (commercial names: DRIVE or CISS) to assess in more spatial details the CVJ, posterior fossa cisterns, and cranial nerves

A mobile app suggesting spine MRI protocol in children called *pedsMRI* is available on Apple Store and Google Play; it is developed by the Hospital for Sick Children in Toronto (Canada) and contains suggestions for MRI pediatric protocols not only in the spine but also in all body regions and for main pathological conditions. We do suggest the use of this app particularly for radiologists not specialized in pediatric images.

Of note, among the established metrics for the assessment of the CVJ, an interesting measurement proposed in patient with Chiari I is the so-called pBC2 on T1 WI, which relies on the maximum perpendicular distance to the basion-infero-posterior point of the C2 body on sagittal views [14].

The majority of patients with ventral compression can be treated by posterior decompression, realignment, and stabilization, reserving anterior decompressions for patients with profound, symptomatic brainstem compression [7]. The

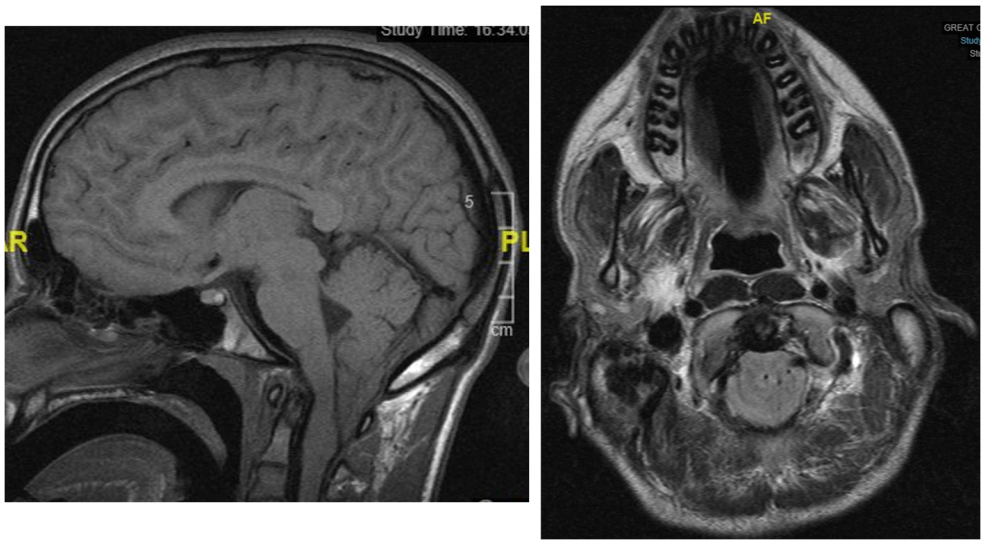

Fig. 5 Sagittal T1 WI (left) and axial T2 WI (right) in a patient with Chiari I due to horizontal and short clivus in the context of a dysmorphic posterior fossa (group 1)

post-operative status of the spinal cord (e.g., resolution of the cord edema) should be assessed again with MRI while the bony outcome is better visualized with CT.

MRI scan is also particularly useful to obtain insights on the possibly associated brain conditions, for instance, the study of the ventricular system and the flow of CSF which cannot be extrapolated from a simple CT head. For example, MRI can help in assessing periventricular edema in ventriculomegaly and overt hydrocephalus [16].

If needed, axial T2*-WI may be acquired for better detection of intracranial hemorrhage as a cause of hydrocephalus, whereas flow-sensitive MRI techniques and three-dimensional high-resolution sequences have been applied for functional and anatomical assessment of CSF flow dynamics, respectively [17–19].

A cine phase-contrast sequence can demonstrate the pulsatile flow of CSF at the level of the CVJ: the acquisitions are synchronized with the cardiac cycle generating images with velocity information providing the ability to calculate the stroke volume, mean velocity, and peak flow in the systole and diastole [20]. Hence, these sequences allow for a qualitative (i.e., monitoring the CSF flow through the aqueduct of Sylvius, and the basal subarachnoid spaces such as prepontine–premedullary cisterns) and quantitative (i.e., can estimate the volume and flow of the CSF) assessment in patients with Chiari I (Fig. 6). However, in our experience this technique can be challenging in terms of reproducibility and with limited clinical usefulness.

Recently, diffusion tensor imaging (DTI), an advanced MRI technique that allows the study of the internal neuroarchitecture in vivo by measuring the three-dimensional shape and direction of diffusion in the brain, was suggested as a suitable tool to evaluate qualitatively and measure quantitatively the microscopic integrity of white matter structures within the brain tissue in congenital and acquired disorders of the pediatric brain [21]. Very few studies have explored the use of DTI in patients with Chiari I: Eshetu et al. conducted a DTI assessment on a cohort of children to study the brainstem and cerebellar white matter tracts' architecture and integrity [22]. The regions of interest (ROI) were centered within the bilateral pontine corticospinal tract, medial lemniscus, and middle cerebellar peduncles: interestingly, their analysis revealed that axial diffusivity and mean diffusivity values were lower in middle cerebellar peduncles in symptomatic patients indicating microstructural white matter changes in these subjects compared with asymptomatic patients.

In light of the broad spectrum of management options available for asymptomatic, paucisymptomatic, and symptomatic patients, and given the lack of agreement on the safest and more effective surgical strategies, experts are now focusing on the potential advantages of advanced MRI sequences like DTI in patients with Chiari I, and future trials are warranted [23].

Preoperative planning and intraoperative imaging: future prospectives

Whereas in the previous sections of this article we have purely focused on the diagnostic protocols for Chiari I in the pediatric population, we considered appropriate to spend now a few words on how those information can be merged through high-tech surgical aid platforms meant to assist in elaborating preoperative planning or intraoperative neuronavigation protocols.

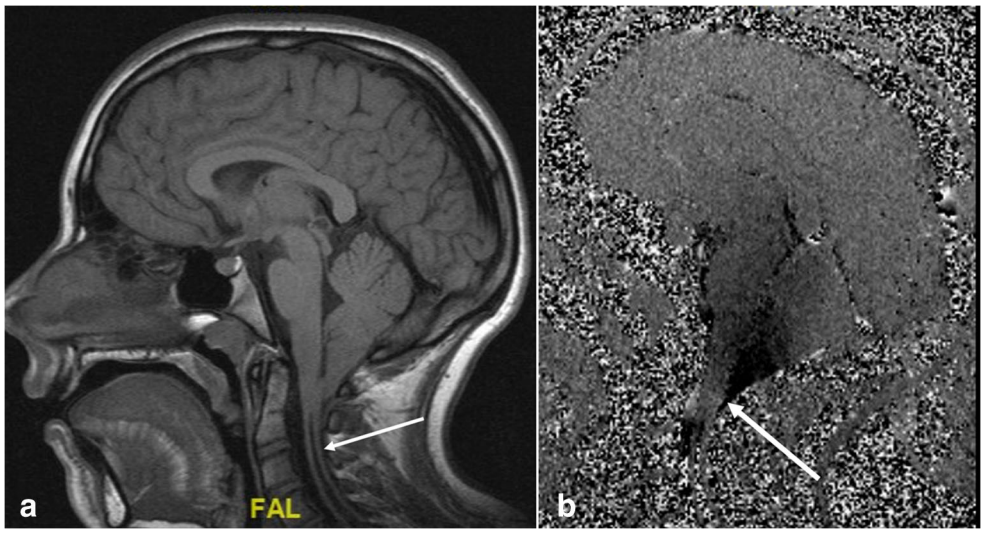

Fig. 6 A 15 year old female. **a** Sagittal T1 WI shows low cerebellar tonsils and associated syrinx (arrow). **b** CSF flow dynamic study (phase-contrast image) shows reduction of the flow posteriorly to the CVJ

Surgeons and scientists have attempted to come up with innovative solutions to address the multiple layers of complexity that several conditions including Chiari I might pose. Many translational advances have been recently applied in modern surgical planning; they range from virtual surgery to anatomical maps and are likely to transform the way the surgical team prepares for any elective operations [24, 25]. Among those translational advances, 3D models to evaluate the role of platybasia and clivus length in the development of Chiari I were proposed using a computer-aided design software based on DICOM files of standard preoperative CT and MRI scans. Using this method, Fernandes et al. simulated the challenges of approaching different clivus lengths and basal angles, and created an algorithm able to calculate the final

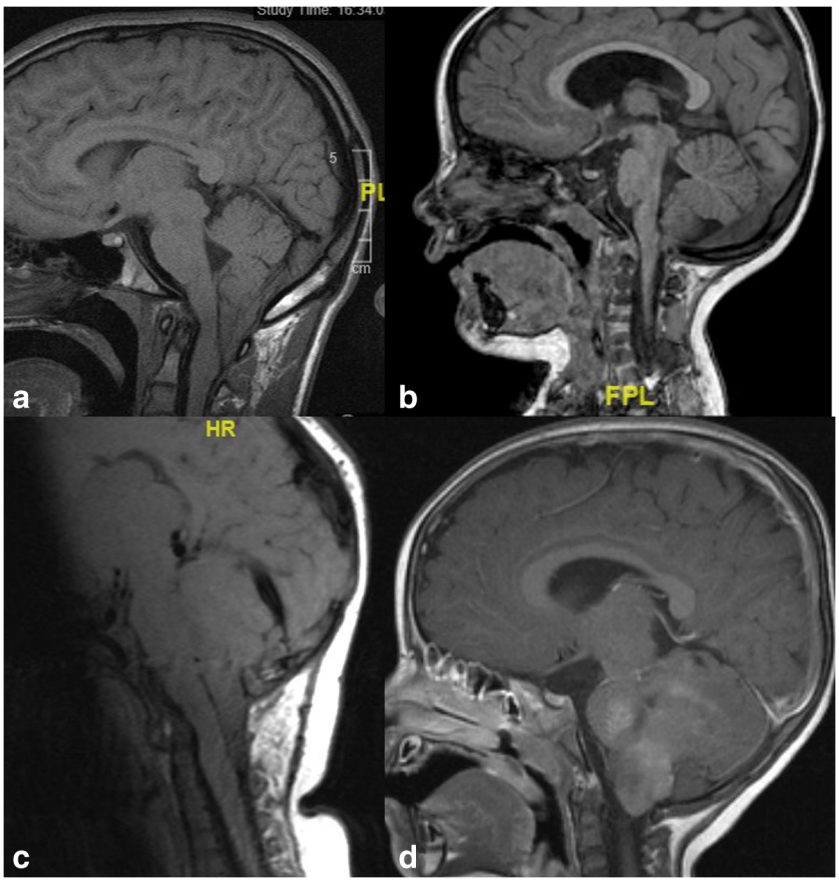

Fig. 7 Summary of the imaging characteristics of the four group of Chiari I. **a** Chiari I deformity due to dysmorphic skull base (group 1). **b** Chiari I deformity due to vertebral segmentation anomalies in a patient with Klippel-Feil (group 2). **c** Low cerebellar tonsils due to a small posterior fossa and associated overcrowding in multisutural synostosis (group 3). **d** Low cerebellar tonsils due to excessive soft tissue in a patient with Lhermitte–Duclos syndrome (group 4)

posterior fossa volume obtained following foramen magnum decompression for each variation and the percentage of the volumetric change achievable with different degrees of decompression [25].

The relevance of such approaches is based on the rationale that a greater increase in the postoperative posterior cranial fossa volume, and specifically an increase in the cisterna magna volume, is statistically associated with a greater likelihood of improvements in headache and tonsillar descent in patients with Chiari I [26–28]. These data have been confirmed in a recent study utilizing 3D methodologies based on a semiautomatic segmentation imaging program used to compare the pre- and postoperative volumes of the posterior cranial fossa and the CSF spaces (cisterna magna, prepontine cistern, and fourth ventricle) and correlated with functional outcomes in a cohort of pediatric patients with Chiari I [26]. Larger increases in the caudal portion of the posterior fossa volume were also associated with a greater likelihood of improvement in syrinx and cervicomedullary kinking.

Furthermore, the use of intraoperative imaging based on point-of-care portable ultrasound has been proposed as a reliable tool to assist neurosurgical teams at the time of foramen magnum decompression [29]. Intraoperative ultrasound can be considered to assess CSF flow patterns at the cranial-vertebral junction and guide the surgical decision-making process during the different stages of dural opening, arachnoid fenestration, and coartation of cerebellar tonsils.

Whereas patients undergoing posterior fossa decompression for Chiari I do not usually need any neuronagivation system, revision surgery and occipito-cervical fusion might warrant the use of all these methodologies ensuring increased accuracy. In these scenarios, neuronavigation systems based on preoperatively acquired and fused CT and MRI scan can help to plan the extent of decompression and the entry points/trajectories for plate/screw insertion. In a pediatric series including Chiari I patients requiring occipital condyle to axial or subaxial spine fixation, Kosnik-Infinger et al. described the remarkable impact on increased precision and reduced complication rate following insertion of polyaxial screw-rod constructs for occipital condyle to cervical spine fixation with intraoperative neuronavigation supplemented by a custom navigational drill guide [30].

Conclusion

We summarized the main indications of different radiological techniques in patients with Chiari I deformity/low tonsils and the impact of the new proposed etiopathogenetic classification on the radiological assessment of these patients and suggest MRI protocols and resources to guide radiologists and clinicians.

Figure 7 summarizes the image findings of the 4 groups of Chiari I (deformity: 1 and 2) and low cerebellar tonsils (3 and 4) recently proposed [2].

Compliance with ethical standards

Conflict of interest Authors have no funding or conflicts of interest to disclose.

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