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Research Article

Diagnosis and Surgical Management of Chiari Malformation Type I

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Abstract

Introduction: Chiari malformation type I (CM I) is an abnormality of the craniovertebral junction affecting both adults and children. Its genesis is not clear however a reduced volume of the posterior fossa looks to play a significant role. We aim to report our experience in the diagnosis and the surgical management of CM I.

Methods: A prospective study was conducted in our department over three years. We have included 50 patients diagnosed with CM I and surgically managed. We reviewed different criteria; epidemiological, clinical, and radiological before and after surgery to evaluate the efficiency of surgery in our patients. The data were analyzed using the Epidata software.

Results: The most encountered preoperative symptoms were headaches majored by Valsalva in 100 % and sensitive disorders (suspended sensory disorders in 32% and thermo-algesic dissociation in 32 %). Preoperative imaging showed that 66% have Chiari type I, 28% have Chiari type 1.5, and 06% have Chiari type 0. The bony and dural opening was performed in all patients. In 70% of patients, cerebellar tonsils were coagulated in 58% and resected in 12% of cases unilaterally. The thenar and hypothenar amyotrophies improved only in 10% of cases, and thermo-algesic dissociation improved in 63%.

Conclusion: Surgical management for CM I is a safe and efficient procedure to relieve preoperative symptoms. Bony and dural is considered a good surgical technique. Additional maneuvers on tonsils should be added according to the preoperative finding after arachnoidal opening.

***Keywords:** Chiari, PCF, Tonsils.*

Introduction

The congenital Chiari type 1 (CM-I) malformation is an abnormality of the posterior cerebral fossa (PCF) and was first described by an Austrian pathologist HANS CHIARI (2,3) in 1891. CM-I affects the cerebellum, the brainstem, and the spinal cord, it is specifically marked by the descent of the cerebellar tonsils of + 5mm below the foramen magnum (1) and occurs as much in children as in adults. The history of this malformation is interesting with additional research projects performed every year on its pathophysiology.

With the advent of more sophisticated imaging modalities such as MRI, more cases were reported and described (1). On one hand, it allowed a precise description but on the other hand added new controversies on treatment modalities: who to treat? When to treat? And how to treat it? In short, the advent of MRI has swung the clinical pendulum of the CM-I, we have gone from the state of rare patients with major clinical signs to the state of many patients presenting few clinical symptoms.

The treatment of Chiari malformation is one of the most controversial topics in neurosurgery; there is disagreement regarding the definition, symptoms, natural history of this malformation, and the appropriate timing of treatment, A wide variety of surgical techniques has been proposed: should we only do a bony decompression of the posterior cerebral fossa? If we open the dura mater, should it be closed, and how to proceed? Should we reduce the size of the tonsils? Can a combination of several techniques be proposed? There is therefore no codified therapeutic course of action reported in the literature, for a precision specialty such as neurosurgery.

We aim in this work to present the main clinical and paraclinical parameters of this condition. The intraoperative findings were also reviewed and added to the surgical results.

Methods

A prospective analytical study of the surgical treatment of CM-I was carried out at Lamine Debaghine University Hospital in Algiers on a general population, from December 2017 to June 2020. The study was conducted on 50 patients. The MRI allowed us to make the diagnosis, verify the presence or absence of a syringomyelic cavity, and visualize the specific morphological characteristics (the containing and content) of the PCF. Flow MRI was performed in patients with CM-I without syringomyelia to confirm obstruction of CSF flow at the level of the PCF. Data entry and analysis were performed with EPIDATA software. We evaluated the different criteria; epidemiological, clinical, and radiological (before and after surgery), to evaluate the efficiency of surgery in CM-I.

We have defined several variables in each studied group, for which we have calculated the frequency, the mean, and the standard variation. The comparison between the quantitative variables was carried out using the STUDENT test, the degree of significance retained is $p<0.05$. (P. Value). The correlation between two variables was carried out using the Pearson index and the comparisons between qualitative variables were carried out by a test of Chi 2.

Result

Patient presentation

- All of our patients (100%) presented symptoms related to the obstruction of the flow of the CSF at the level of the PCF.

- Occipital headaches majored by the Valsalva maneuver in 100% of cases.

- Visual disturbances marked by visual blurring in only 4% of cases. however visual acuity and fundus were normal.

- 40% of patients present signs due to the compression of the brainstem and stretching of the cranial nerves :

* Swallowing disorders in 22%, vertigo in 18%, nystagmus in 14%, sleep apnea in 6%, diplopia in 4%, snoring in 2%, and vocal cord paralysis in 2%.

- 38% of patients showed cerebellum compression-related symptoms that were grouped as follows :

* A static cerebellar syndrome in 34%.

*A kinetic cerebellar syndrome in 20%.

*Cerebellar ataxia in 2% .

- 84% of patients presented symptoms related to spinal cord compression and were using the JOA score, and we have found a minimum of 08, a maximum of 18, and an average of 15 in our patients.

- Sensory disturbances and thermo-algesic dissociation were found in 64% of patients.

- Urinary incontinence in 4% of cases.

- Decreased libido in 2% of cases.

Associated pathologies

- Among the studied population, the pathologies associated with CM-I were; basilar impression in 12% (non-surgical), cervico-dorsal scoliosis in 8%, atlas assimilation in 6%, passive hydrocephalus in 4% and pseudotumor cerebri in 2%.

Family history of CM-I

A family history is found in 4% of patients.

Imaging results

Preoperative imaging showed that 66% have Chiari type I, 28% have Chiari type 1.5, and 06% have Chiari type 0. A tonsillar herniation of at least 05mm under the foramen magnum was found in 94% of our patients (excluding Chiari types 0), with a minimum of 7mm, a maximum of 45mm, and an average of 17.57mm. the average PCF volume is 166.22 cm3, with a minimum of 100 cm3 and a maximum of 260 cm3.

The extension of the syringomyelic cavity concerns at least: 01 vertebra, at most: 18 vertebrae, and on average: 8.9 vertebrae, The Vaquero index of the syringomyelic cavity had an average of 0.56, a minimum of 0.3, and a maximum of 0.9.

In 94% of our patients, the lower end of the cerebellar tonsils is pointed.

Flow MRI was only performed in 30% of our patients and this was specifically for patients with symptomatic CM-I without syringomyelia or if CSF flow obstruction was not evident on conventional MRI.

Particular association

- The study of the relationship between tonsillar ectopia and the JOA score found a correlation coefficient (r) of -0.04, so there was no relationship between the degree of tonsillar ectopia and the clinical impact.

- The (r) between tonsillar ectopia and syrinx extension is -0.22, which means that the degree of tonsillar ectopia does not influence the extension of the syringomyelic cavity.

- The (r) between PCF volume and tonsil descent degree is -0.22, so there is no relationship between PCF volume and tonsil descent degree.

Surgical results and intraoperative findings

The size of the suboccipital craniectomy has an average of 3.85 cm +/- 0.47 cm in height and 4.14 cm +/- 0.37 cm in width. The ablation of the posterior arc of C1 was done in all patients. The posterior arc of C2 and C3 were resected in 09, and 02 patients respectively.

A compressive fibrous band at the level of C1 was found in 87% of patients. We have noticed asymmetric tonsils in 92%, and usually, the left amygdala was lower than the right amygdala.

Arachnoid adhesions retaining the tonsils at the bulbomedullary junction were found in 78% of patients. In 80% of our patients, we found a free Magendie foramen, and in 20% of cases it was obstructed by an arachnoid veil, this obstruction was found especially in Chiari type 0.

In 70% of patients, cerebellar tonsils were coagulated unilaterally in 58% and resected unilaterally in 12% of cases. We have looked for a relationship between these maneuvers and the type of Chiari that showed a P. Value of 0.41. Consequently, the type of Chiari does not influence surgical maneuvers on the tonsils.

The dural defect was repaired by an autologous plasty in all patients. It was harvested from the occipital galea. When closing the dura, we maintained the arachnoid lifted to the dura to avoid postoperative arachnoiditis.

Postoperative clinical results

The majority of clinical signs improved postoperatively, except for thenar and hypothenar amyotrophy which improved in only 10% of cases, and thermo-algesic dissociation which improved in 63%. The JOA score in CM-I overall postoperatively had a minimum of 14, a maximum of 18, and an average of 16.6+/-0.9. Nearly half of the patients had a JOA score of 17 postoperatively.

Table 1: Post-operative clinical study.

Symptoms	Total number pre op	Number of improvement	Percentage of improvement	Number of stabilizatio	Percentage of
headaches	50	50	100%		
visual disturbances	1	1	100%		
diplopia	1	1	100%		
nystagmus	8	8	100%		
dysphagia	11	9	82%	2	18,18%
sleep apnea	4	4	100%		
snoring	1	1	100%		
vocal cord paralysis	1	1	100%		
vertigo	8	6	75%	2	25,00%
cerebellar syndromes kinetics	10	10	100%		
static cerebellar syndrome	17	14	82%	3	17,65%
ataxia	1	1	100%		
suspended sensory disorders	32	26	81%	6	18,75%
thermo-algesic dissociation	32	20	63%	12	37,50%
urinary incontinence	2	1	50%	1	50,00%
decreased libido	1	1	100%		
neuralgia	23	22	96%	1	4,35%
thenar amyotrophy	30	3	10%	27	90,00%

Table 2: The JOA score postoperative.

JOA Score	Percentage
14	2,4%
15	9,5%
16	26,2%
17	47,6%
18	14 ,3%

By comparing the JOA score before and after surgery, a significant P. Value of 0.0000001 was noticed, confirming the improvement of the JOA score after the surgical procedure.

Postoperative radiological results

All of our patients underwent a cerebro-medullary MRI between 15 days and 1 month post-operatively. where we noted:

-An ascension of cerebellar tonsils was noticed in all patients.

-A creation of a neo-large cistern was visible in all patients.

-A modification of the tonsil's shape was observed in all patients. The lower extremity of the tonsils became rounded post-operatively.

In our series, the extension of the syringomyelic cavity post-operatively concerned a minimum of 01 vertebrae, a maximum of 16 vertebrae, and an average of 6.76 vertebrae. By comparing the extension of the syringomyelic cavity pre- and postoperatively, the P.

Value is 0.002, so this proves that the surgical procedure was efficient and reduced the extension of the syrinx.

The postoperative vaquero index had an average of 0.25, with a minimum of 0.1 and a maximum of 0.6. We have noticed in 9.30% of cases, a total resolution of the syringomyelic cavity.

Complications

During our study, we did not have any CSF fistula cases. Only 1 patient presented a wound infection that was well managed after antibiotic treatment.

Illustrative Case

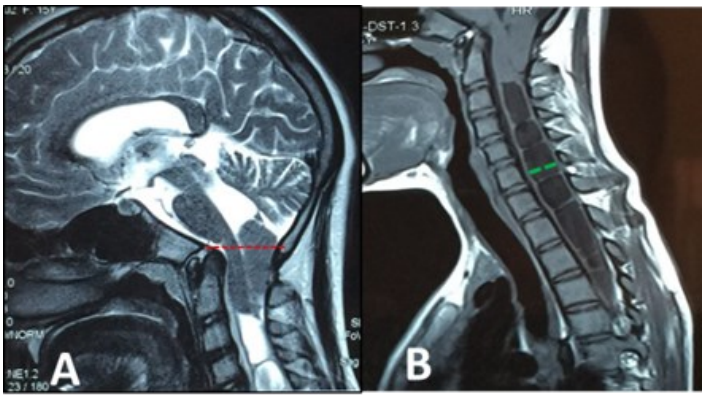

Figure 1 : *(A) Preoperative MRI sagittal T2 sequences, showing a CM-I type 1.5, with a significant descent of the brainstem and tonsil beneath Macrae line (red dots). (B) cervical MRI showing an important syrinx (green marks) extending to the dorsal medulla.*

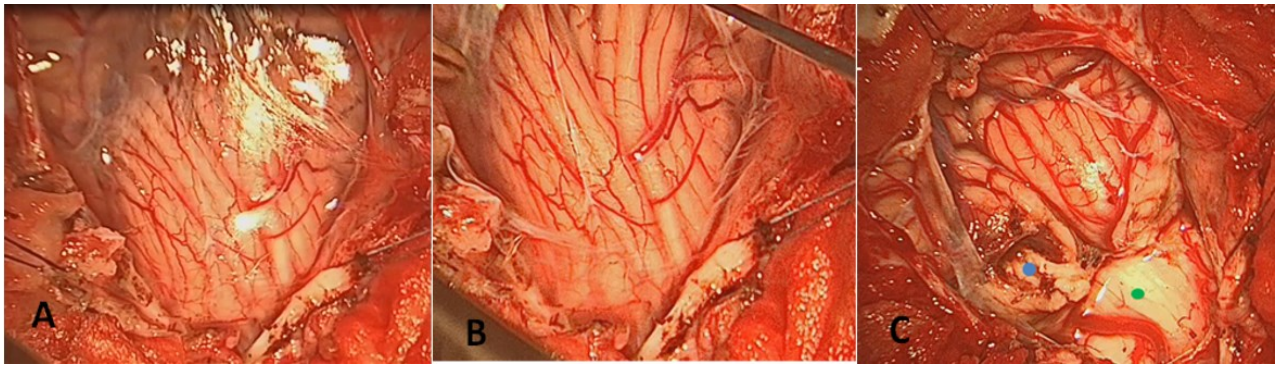

Figure 2*: (A) Preoperative microscopic view of the tonsils after dural opening with an intact arachnoid. (B) a view of the tonsils after arachnoid opening and before their resection. (C) superior displacement of the tonsils after a subpial and partial resection (blue dot) and a good visualisation of the brainstem (green dot).*

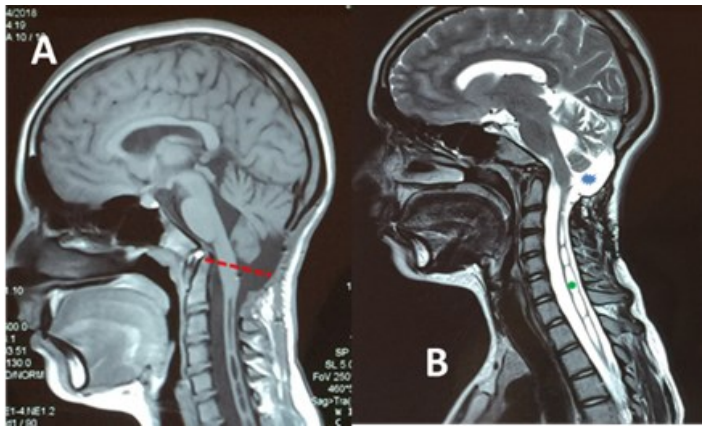

Figure 3: *(A) postoperative T1 sagittal MRI, showing a significant ascension of the tonsils above the Macrae line. (B) sagittal T2 MRI showing a good resolution of the syrinx (green star) and the neo-large cistern (blue star).*

Discussion

Pathogenesis

Chronic tonsillar herniation in CM-I could be related to an abnormal development of the occipital bone, therefore the cerebellum will have an abnormal growth in a small PCF. The absence of cisterna magna in the PCF was found in 100% of our patients, the same findings have been reported by Milhorat et al (7). The tonsillar herniation was 05mm at least under the foramen magnum. This was found in 94% of our patients, and it was 91% in the series by the same authors (7). Considering the volume of the PCF, Quantitative measurements of the volume showed a significant reduction in this compartment. In our study, the average volume of the PCF is 166.22 cm3 while in normal subjects this volume is 236.565 cm3 +/- 40. These Results showed that a small PCF plays a fundamental role in the genesis of CM-I. These findings have been observed in the literature, notably in the work of Tubbs (8), the study of Milhorat (7) where the average volume of the PCF is 124+/-21.4 cm3, and the series of Noudel (10) where the average volume preoperatively was 156.68 ± 16.0 cm3, as well as the study by Schardy (11) which found that the volume of the PCF is 23% smaller in patients with CM-I compared to the normal subject.

Epidemiology

In our series, women were far more affected than men with a sex ratio of 0.51, This female predominance is found in the literature among the majority of authors, the sex ratio in Milhorat's series (7) is 0, 32, and 0.65 in the Alzate study (12). This predominance tends to be reversed in the pediatric population. The average age of the patients in our series was 40 years, in the study by Milhorat (7) that concerned the adult population, the average age was 35.9 years. Tubbs reported that in the pediatric population, the average age was 11 years (8), and finally in the Alzate series (12) which included both adults and children, the average age was 15 years.

Our results regarding clinical presentation were compatible with the large studies in the literature. For Tubbs (8), the encountered symptoms were headaches in 100% of cases, neck pain in 15%, the lower cranial nerves deficit in 10%, dysphagia in 2%, sleep apnea in 5%, dysphonia in 1.8 %, neurological deficit in 2.4% and cerebellar ataxia in 3.8% of cases. In Milhorat's work (7), he also found that the most frequent clinical signs are sub-occipital headaches majored by the Valsalva maneuver found in 81% of patients. The cranial nerves and the brainstem-related symptoms were present in 52% especially dysphagia, sleep apnea, and dysarthria.

An unexpected data in this work is the presence of signs of spinal cord compression in 42.85% of CM-I without syringomyelia, this feature was observed in 66% of patients in the same group, it was also reported by Milhorat (7). These disturbances could be related to mechanical compression (herniation of cerebellar tonsils) of the corticomedullary junction. Another origin could be, can be evoked represented by the effect of the systolic pulsation wave in the spinal canal and its capacity to produce symptoms similar to those of syringomyelia (8).

We also found that the symptoms related to the compression of the brain stem and the stretching of the cranial peers predominate in Chiari type 1.5 with syringomyelia, the P. Value is 0.0152. This observation has not been noted in the literature. This predominance is explained by the fact that Chiari type 1.5 is associated with a descent of the brainstem, hence the high frequency of the attack of the cranial pairs.

CM-I and syringomyelia

In our series, 86% of our patients had a syringomyelic cavity. In the study by Milhorat (7) 65.38% of cases presented syringomyelia, Tubbs (8) identified 57% of syrinxes associated with CM-I, and Alzate (12) reported 51.51% of syringomyelia associated with CM-I.

We therefore note that the rate of syrinx in our series is slightly higher than what is described in the literature. This could be explained by the radiological investigation performed mainly for patients presenting significant symptoms, present in patients with extensive syringomyelia.

CM-I and scoliosis

8% of our patients presented scoliosis associated with CM-I. Tubbs (8) reports a rate of 18% of scoliosis in and. Milhorat 41% (7).

Preoperative radiological findings

We found a tonsillar herniation of at least 05mm under the foramen magnum in 94% of our patients (excluding Chiari types 0). We noticed a minimum of 7mm, a maximum of 45mm, and an average of 17.57 mm. Milhorat (7) reported 91.2% tonsil descent of 5 mm or more.

Tubbs (8) reported that all patients had a tonsillar herniation of at least 05 mm below the foramen magnum.

There was no relationship between the descent of the tonsils and the severity of the clinical signs or the extent of the syringomyelic cavity, this was demonstrated by calculating the correlation coefficient (r) between these different parameters, which is -0.04 and -0.22. This was also noted by Tubbs (8) and Milhorat (7).

The pointed shape of the lower end of the tonsils preoperatively, according to Tubbs (8), would play a role in the formation and extension of the syrinx by increasing the piston-like effect. In his study he noted this particularity in 97% of cases, in fact during our study the same observation was established, 94% of our patients have a pointed shape of the lower end of the cerebellar tonsils preoperatively.

The absence of a large cistern was noted in all our patients, this particularity is common for all types of Chiari (1,1.5 and 0), it is described in the majority of series, notably Tubbs (8, 6, 9), Milhorat (7), Alzate (12), McGrit (14).

The surgical procedure

The average duration of the intervention is 164 min +/- 36 min. This duration tends to decrease over time. We also note that 2/3 of the time is spent closing the dura mater using duralplasty. In the literature, the average duration of the intervention is between 95 and 120 min, Tubbs (8) noted an average duration of 95 min. This difference in the duration of the procedure compared to other authors is due to the additional time (2/3 of the total intervention duration) that we devote to the watertight closure of the dura mater.

As for the majority of studies, the surgical indication in our series is proposed for symptomatic patients with tonsillar ectopia of 5 mm or more below the foramen magnum, associated or not with syringomyelia. In addition to these criteria, Tubbs (8) recommended operating on even asymptomatic patients in whom the syrinx vaquero index is greater than 0.5.

The size of our suboccipital craniectomy is on average a height of 3.85 cm +/- 0.47 cm, and a width of 4.14 cm +/- 0.37 cm with systematic removal of the posterior arch of C1, the posterior arch of C2 in 18% of cases and of C3 in 4% of cases. In Tubbs reports (8) the craniectomy was 4 to 5 cm, with the ablation of the posterior arch of C1 in all patients, rarely the one of C2 (0.4%), because the coagulation of the tonsils is often unnecessary. Coagulation of the tonsils allows their adequate raising. The size of the craniectomy is fairly standard, between 3 and 5 cm, because if it exceeds these values, a risk of postoperative cerebellar ptosis may be associated with that. A fibrous and adherent band, compressing the extradural bulbo-medullary junction is found in the majority of our patients, after removal of the posterior arch of C1. This data has been also described by Tubbs (8).

Anatomical and technical parameters for the dural opening option

- In 78% of our cases there was no pulsation of the cerebellar tonsils after dural opening and were generally retained by arachnoid adhesions (78%), the frequent presence of these adhesions has been confirmed by several authors, notably Tubbs (8), Milhorat (7) and McGrit (14).

-In 20% of our patients, there was an arachnoid veil obstructing the foramen of Magendie. This was described by Tubbs (130) in his study where 12% of cases presented an obstruction of the foramen of Magendie. In 58% of cases of our patients, coagulation of the tonsils was necessary to elevate the cerebellar tonsils, and in 12% of cases, a subpial resection of the tonsils was necessary. Alzate (12), achieved coagulation of the tonsils in 59%, and in 9.76% of cases a subpial resection. All these arguments cited previously concerning intra-dural exploration make opening the dura mater capital in CM-I.

An autologous dural plasty was performed in all our patients, Tubbs (8) also reported the same procedure unlike Alzate (12) who used a synthetic dura for all his patients. We preferred an autologous plasty, because there was less risk of infection and is free of cost. During the closure of the dura, we maintained the arachnoid suspended to the dura to avoid postoperative arachnoiditis as it was described by Tubbs (8).

Complications

We did not notice any case of CSF fistula in our series. 01 patient presented an infection of the operative wound well managed with antibiotics. We didn't have cases of septic meningitis, cerebellar ptosis, pneumocephalus, instability of the occipital-vertebral junction, or hydrocephalus. Tubbs (8), in his study which concerned 500 patients with CM-I, only 01 case presented a CSF fistula, 01 infection of the operative wound, 04 hydrocephalus and hematoma in 02 patients. McGrit (14) described 3% of CSF fistula in 393 patients. The absence of CSF fistula in our study is related to the significant time devoted to closing the dura mater (2/3 of the total duration of the procedure), and the frequent use of biological glue. We also note that no patient underwent surgical revision during a follow-up period of around 30 months.

Tubbs (8) reported that 3.2% of patients underwent reoperation, 2 months after the first surgery, because of the persistence of clinical signs and syringomyelia. In these patients, the dura mater was not opened during the first surgery. He noticed the presence of an arachnoid veil, obstructing the flow of CSF through the Magendie foramen.

Clinical results

Suboccipital headaches, cerebellar syndrome, cranial nerve, and motor deficits, had an improvement rate approaching 100%. Unlike thenar and hypothenar amyotrophy which only improved in 10% of cases, and thermo-algesic dissociation which improved in 63% of patients.

Tubbs (8) noted that the clinical symptoms that recovered after surgery were Valsalva-induced suboccipital headaches, motor deficits, and sleep apnea. Indeed, in his series, suboccipital headaches were relieved immediately postoperatively in 83% of cases, while non-occipital headaches only improved in 15% of cases. McGrit (14), noted that only 40% of frontal headaches improved, and apnea and vertigo were less likely to improve.

Radiological results

We noticed that the lower end of the cerebellar tonsils became rounded postoperatively, in all patients, whereas preoperatively it was pointed. The change in the shape of the lower extremity of the cerebellar tonsils, before and after surgery, was noted by Tubbs (8), it was even considered as an effectiveness criterion of the surgical procedure. The comparison of the longitudinal extension of the syringomyelic cavity before and after surgery in our patients (P. Value of 0.002) proved the effectiveness of our surgical procedure on the extent of the syrinx.

The vaquero index pre and postoperatively of the syringomyelic cavity was also evaluated (P. Value of 0.033) confirming the efficiency of our surgical procedure. In 9.30% of cases, we noticed the total disappearance of the syringomyelic cavity.

The majority of the series agree that surgery in CM-I significantly reduces the volume of the syringomyelic cavity, Tubbs (8) noted resolution of the syrinx in 95% of patients, Zhang (15), noted a reduction in the size of the syringomyelia at 3 months in 30% of cases, and 85% at 6 months. Alzate (12), notes a disappearance of the syrinx in 22.72% of cases, and a significant reduction of the latter in 77.28% of cases.

Conclusion

CM I is a disabling pathology affecting the patient and his quality of life by several clinical symptoms. MRI is the gold standard of the diagnosis, however complete functional evaluation of the CV junction is mandatory with other imaging modalities. The pointed shape of the tonsils has a significant role in the pathophysiology of this disease. Surgery is an efficient and safe option for symptomatic patients or asymptomatic with syrinx and a vaquero index superior to 0.5. thenar and hypothenar amyotrophy don't resolve after surgery in most cases. The scoliosis association with CM I and its appropriate management could be an interesting research field in the future.

Conflict of Interest

The authors declare no conflict of interest.

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