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Surgical Procedure And Future Treatment Options For Posttraumatic Syringomyelia: A Systematic Review

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Abstract

Background: Syringomyelia related trauma is uncommon case compared to other etiology such as Chiari Malformation type 1. The management of PTS is remain unclear and debatable. The aim of this study is to review PTS regarding surgical procedure, outcomes, and potential future treatment strategies.
Methods: This study uses a PRISMA flowchart as structured analyses. Document selection using the keywords "Postraumatic Syringomyelia" AND "Syringomyelia" and based on publication (10 years). A total 17 studies met the eligibility criteria. We recorded treatment strategies, level of syrinx, severity of injury, injury to symptom duration and outcomes.
Results: A total 264 participants with PTS from 15 observational studies and 2 clinical trials were reviewed. We recorded 68% of participants were treated surgically (n=181). The most common procedure is arachnoid lysis and duraplasty (32,5%, n= 59) followed by syrinx shunting (15,4%, n=28) and subarachnoid-subarachnoid bypass (11%, n=20). We found that the syrinx shunting was the most successful procedure with symptoms improved in 82% of patients postoperatively. Beside of surgical strategy we also found another therapeutic option that use mesenchymal stromal cells (MSCs) that injected into the syrinx. We recorded a 100% success rate from this procedure (n=7)
Conclusion: Although the overall outcomes of the surgical procedure for PTS are still unsatisfactory, the syrinx shunting procedure seems to be considered as the most effective surgical procedure. Other therapeutic strategies such as MSCs is considered promising but still require further researches with larger sample sizes.

Keywords : Syringomyelia, Syrinx Shunting, Cells Therapy, Arachnolysis

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Introduction

Syringomyelia is a disease that produces fluid-containing cavity in the spinal cord. Based on the etiology, syringomyelia divided into traumatic and non-traumatic syringomyelia.[1] The most common cause is Chiari Malformation type 1 which contributes more than 50% as non-traumatic cause of syringomyelia. In recent years, there are several publication that presents various surgical technique for syringomyelia related Chiari Malformation type 1, in other hand management for syringomyelia related trauma is still lack of literature and debatable [1][2].

The cause of posttraumatic syringomyelia rremains incompletely understood, but there are certain risk factor that can increase the predisposition to developing syringomyelia, including arachnoiditis, cord compression, and kyphotic deformity [2][3][4]. The clinical appearance of posttraumatic syringomyelia is progressive neurological deterioration including motor function, sensory, and otonom. The prevalence of symptomatic posttraumatic syringomyelia is approximately 4% among patients with spinal cord injury, whereas the prevalence of asymptomatic posttraumatic syringomyelia is approximately 28% [5].

Diagnosis must always be derived from clinical suspicion. Syringomyelia is usually suspected in patients with stable spinal cord lesions or a history of neurological deficit, secondary to spinal cord processes or surgery, who display sudden, progressive clinical deterioration, and whose symptoms progress at a slower pace than at onset. MRI is currently the most widely used imaging technique and is complemented by cine MRI [6][11].

All theories to explain syrinx formation and extension have focused on cerebrospinal fluid (CSF) circulation and its disturbance as the origin of posttraumatic syringomyelia. Surgical management is indicated in patients with progressive neurological deterioration. Surgery may be classified into 2 types, depending on the purpose of the intervention : symptomatic surgery, aimed at emptying the syrinx by draining the CSF into other cavities through tubes; and aetiological surgery, which aims to identify the exact location of the obstruction and restore normal CSF circulation [5][6][9].

Recently, clinical studies show that cell therapy with mesenchimal stromal cells (MSCs) improves the outcomes of spinal cord injury patients. Rhen cell therapy is applied to patients with syringomyelia, a strategy may be the intalesional or intrathecal administration of MSCs. This type of cell therapy reported safe and effective to syrinx reduction and clinical improvement in individual patients [17][18].

Methods

The retrieval of studies was performed in PubMed, Cochrane Library, Scopus, and JNS using the combined filter with predefined search term “Syringomyelia” AND “Posttraumatic Syringomyeia”. All records were screened based on title and abstract independently by first author. All studies describing treatment and outcomes of treatment of posttrumatic syringomyelia from 2012 to 2022 were included in this research. There are no limitation on study type or sample size. Animal studies were not included in this study.

This systematic review was prepared in accordance with Preferred Reporting Items for Systematic Review and Meta-Analyses (PRISMA). Relevant articles were screened based on their title and assessed for the their eligibilty by reading the abstract and full text if necessary. Finally, we recorded treatment strategies, level of syrinx, severity of injury, injury to symptom duration and outcomes.

Results

Study Selection

We found 709 papers that match with our search term, all papers in english languages. We remove 7 papers before screening due to duplicate reason. We identified and screened 702 papers by titles and abstracts and exclude 669 papers because they didn't study posttraumatic syringomyelia and didn't perform treatment strategies, and outcomes of surgical procedure. The full texts of the remaining 33 papers were analyzed comprehensively and we decided to exclude 14 papers that didn't meet inclusion and exclusion criteria such as syringomyelia with nontraumatic etiology, no follow up after treatment, and lack an outcomes. 2 studies were animal study and excluded. Finally, we have 17 studies with 15 observational studies and 2 clinical trials with a total 264 participants.

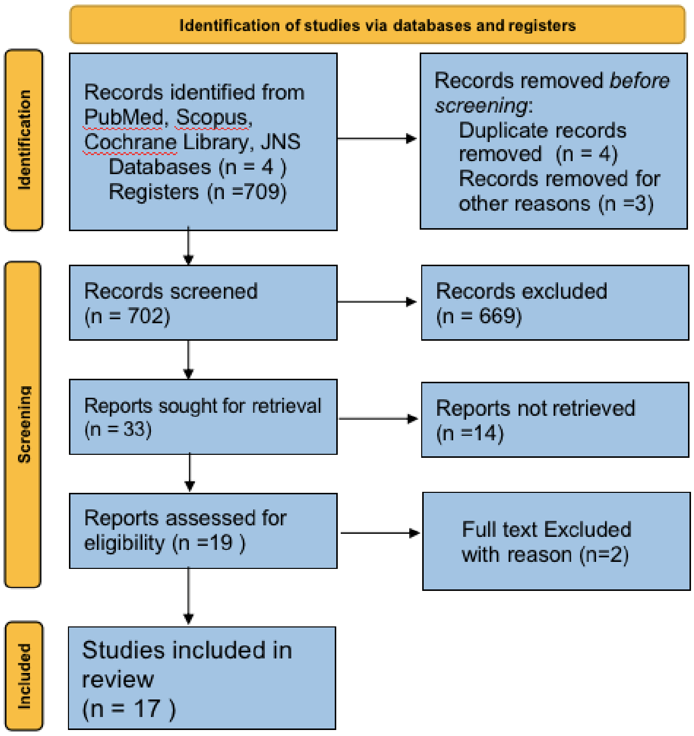

Figure 1. PRISMA flow diagram of the screening process

Sample Characteristics

A total 264 participants were included in this study with male as the most frequent patient with posttraumatic syringomyelia. The most frequent level of syrinx were thoracic region (53%) followed by lumbar and cervical (22,3% and 17,4%) respectively. The most common symptoms was pain and motor and sensory disturbance that depends on severity. We also recorded 68% of participants were treated surgically, 28% treated conservatively, and 2% treated with an Injection of mesenchymal stromal cells (MSCs).

Table 1 Sample Characteristics

No	Author and Study Type	Sample Size	Duration of Injury	Severity of Injury	Level of Syrinx	Surgical Technique / Treatment Strategy	Follow Up	Outcomes
1.	Srikantha et al Observational Study	n = 3 Male : 3	24-36 month	n.a.	CV Junction - D11; n=1 C3 to D2 and D3 to L1; n=1 C2 to D3; n=1	Syringo-Subarachnoid Shunt	1 year	Signficant Symptom relief and preserve neurological status; 100%
2.	Vincenzo et al Observational Study	N= 1 Female	7 years	Right hemisoma paresthesias and pain, D5 sensory level	D10 - L1	Syringo-Subarachnoid Shunt	2 years	Improved No Complication
3	Yuping D. Li et al Observational Study	n = 24 Male : 19 Female : 5	1.5-50 month	AIS (A) n=12 AIS (B) n=0 AIS (C) n=2 AIS (D) n=9 AIS (E) n=1	Cervical = 10 Cervicothoracal = 8 Thoracal = 5 Thoracolumbal = 1	Fusion = 10 Laminectomy = 5 Fenestration = 4 Lysis of adhesion = 1 Duraplasty = 1 Shunt Placement : 4	21 months	Syrinx Lengths; Increase = 4 Decrease = 10 Stable = 10 Syrinx diamter ; Increase = 6 Decrease = 17 Stable = 1
4	J.Vaquero et al Clinical trial	N = 6 Male	5.75-27.68 years	AIS (A) n=3 AIS (B) n=2 AIS (D) n=1	D5 ; n=2 D3; n=1 D4; n=1 D8; n=1 L1; n=1	Administration of 300 millions mesenchimal stromal cells (MSCs) inside the syrinx	6 months	Improvement observed in the studies of urodynamic, neurophysiology, neuroimaging and anorectal manometry for all patient.

5	Seon-Hwan Kim, M.D. et al Observational study	N = 1 Male	16 years	Loss of sensory sensation and motor weakness	L1	Syringo-subarachnoid-peritoneal shunt using T-Tube	2 years	Improvement of motor weakness and sensory function and reduction in the size of syrinx
6	Hyun Gon Kim, et al Observational study	N = 9 Male	3-44 years	Motor weakness; n=5 Sensory disturbance; n=4 Bladder disturbance; n=1	Cervical; n = 2 Cervicothoracal, n=3 Thoracal; n = 2 Cervicolumbal; n= 2	Syringo-subarachnoid shunt; n=3 Arachnoidolysis + Duraplasty; n=4 Syringopleural shunt; n=2	2-17 years	Deteriorated; n=4 No change; n=4 Improved; n=1
7	Yasuyoshi Miyao et al observasional study	N = 1 male	2 months	ASIA A	C1 - T3	Syringo-subanachnoid shunt	2 years	Improvement of motor weakness
8	Ali Fahir Ozer, Hosein Jafari Marandı, Mehdi Sasanı observasional study	N = 1 male	16 years	ASIA 0	T4 - T5	Syringopleural shunt and bilateral subanarchnoid to subarachnoid catheters	6 months - 3 years	Partial regression of the syrinx and no clinical symptoms

9	HP Leahy, AA Beckley, CS Formal observational study	N = 6 male	104 - 2028 weeks	AIS A = 2 AIS B = 1 AIS D = 3	Cervical; n=2 Thoracal; n=4	Lysis of adhesions = 1 Shunt placement = 1 Syringo-peritoneal shunt = 1 Syringo-plural-shunt = 1 Syringo-subarachnoid-shunt = 2 Fusion = 1	N/a	Improvement; n = 0 Decline; n = 6
10	Idris Amin, Gavriil Ilizarov, Nayeema Chowdhury observational study	N = 1 male	30 years	N/a	C1 - T12	Syringo-subarachnoid-shunt	N/a	improve
11	Jack M. Leschke, Michael L. Mumert, Shekar N. Kurpad observational study	N = 1 male	6 months	Complete loss of motor function	C2 - C7	Syringo-subarachnoid-shunt using myringotomy tube	2 months - 6 months & 1 year	Improvement of strength and sensassion
12	Mengchun Sun, MD, Benzhang Tao, MD, Gan Gao, MD observational study	N = 28 Male = 12 Female = 16	N/a	N/a	Lumbo sacral; n=9 Thoracolumbo-sacral = 7 Whole spinal cord = 12	Terminal ventriculostomy-associated "V" - type ostomy	3, 12, 24, 36 month	90% of patients had achieved "markedly improved" and "improved" outcomes

13	UKachukwu et al Observational study	N = 2 male	N/a	N/a	Cervical ; n=1 Multilevel; n=1	Syringo-subarachnoid-shunt Syringo-myelotomy	N/a	Improved ; n=2
14	Jörg Klekamp, M.D Observational study	N = 137 Male=101 female=36	135 months	ASIA A+D; n=45 ASIA C+D; n= 55 ASIA E, n = 37	Cervical; n=10 Thoracal; n=91 Lumbar; n=36	conservative (n=76) surgery (n=61) - arachnolysis + duraplasty; n=58 - chord transection; n=7	67 months	Conservative : Stable; n=51 Surgery ; - ASIA A+B: improved; n=12 Stable; n=8 - ASIA C+D: improved; n=12, stable; n=9, deteriorated; n=3 - ASIA E: Improved; n=6 Stable; n=9 deteriorated; n=2
15	Tetsuo Hayashi, M.D., Takayoshi Ueta, M.D., Ph.D., Masahiro Kubo, M.D. Observational study	N = 20 male : 19 female : 1	126 months	Complete loss motor function; n=11 Incomplete loss of motor function; n=9	Cervical; n=4 Thoracal; n=11 Lumbar; n=5	Subarachnoid-subarachoid bypass; n= 20	48 months	Improved; n=12 Stable; n=4 Deteriorated; n=4

16	Nejat Isik 1 , Ilhan Elmaci 2 , Nihal Isik	N=19	24 months	n.a.	n.a.	Syringostomy; n= 2 Syringosubarachnoid shunt; n=5 Syringopleural shunt; n=12	108 months	Improved; n= 16 Stable; n= 1 Deteriorated; n=2
17	Jesús Vaquero, Rasha Hassan, Cecilia Fernández Clinical Trial	N=1	26 years	Complete paraplegia	C2-T4	Injection of Mesenchimal Stromal Cells (MSCs) into the syrinx without aspiration or drainage	1 year	ASIA score improve from 148 to 178

Clinical Outcomes

Patients were broadly grouped into 3 groups based on the therapy they received. all studies follow up with an average time span of 36 months. For the patients who underwent surgery, we subdivided them based on the type of surgical procedure. Arachnolysis and duraplasty are the most frequently performed procedures for the treatment of postraumatic syringomyelia. There were 59 patients (32.6%) who underwent surgery with this procedure and 29 patients (49,1%) had clinical improvement, 25 patients (42,4%) did not experience any changes, and 5 patients (8,5%) experienced worsening. Syrinx shunting and terminal ventriculostomy were the second most common procedures in this study, with 28 patients in each procedure. However, in terms of operating results, syrix shunting become the most successful procedure to improve clinical outcomes. From a total of 28 patients, there were 20 patients who experienced clinical improvement or about 71.4%. As for terminal ventriculostomy, patients who experienced clinical improvement were 50%. Other surgical procedures in this study were subarachnoid-subarachnoid bypass and fusion. The success rate of the subarachnoid-subarachnoid bypass procedure was 60% in a total of 20 patients. For the fusion procedure the success rate was about 54% out of a total of 11 patients.

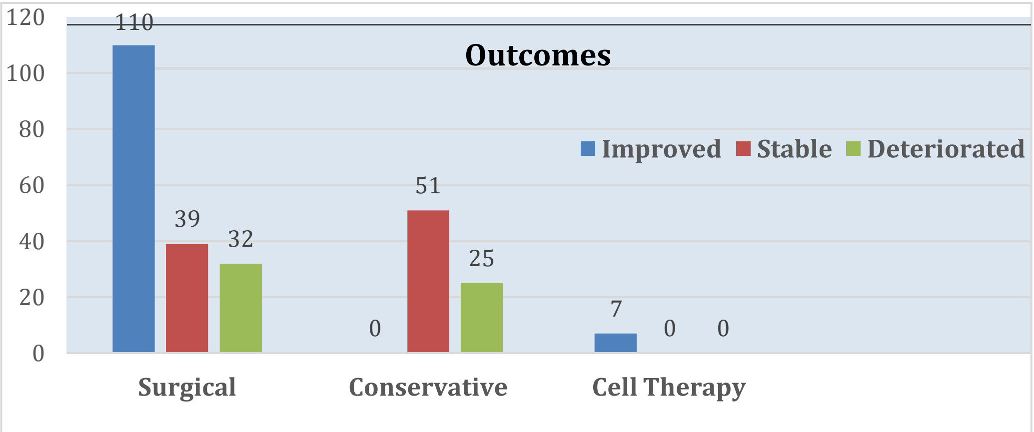

Figure 2. Outcomes from each procedure for posttraumatic syringomyelia

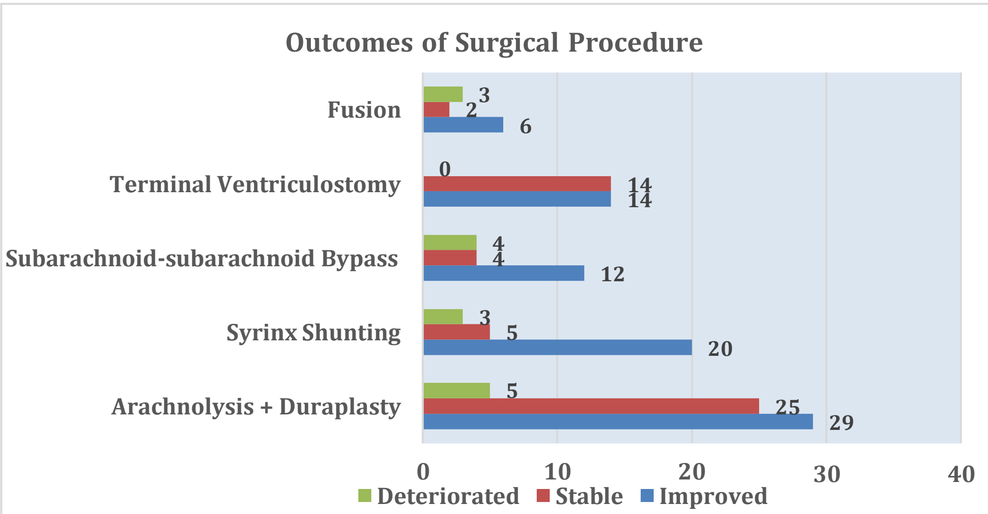

Figure 3. Outcomes of surgical procedure

Discussion

Multiple treatment options and strategies have been described; these include conservative treatment, symptomatic treatment, and symptomatic or aetiological surgical treatment. Most studies support aetiological treatment in view of the high rate of poor outcomes and recurrence associated with symptomatic treatment, despite initially positive results.[5]

CSF shunting to the pleural cavity or the subdural space is the most widely used symptomatic surgical technique. CSF shunting to the pleural cavity was first performed by Abbe in 1892. Although in the field of neurosurgery CSF has traditionally been diverted to the peritoneal cavity, it is usually diverted to the pleural cavity in the context of syringomyelia as this space is closer and the procedure is easier to perform. Several studies report favorable short-term outcomes for symptom management and patient function. CSF may also be diverted to the subdural space. This procedure has been used for many years as it is easier to perform than other techniques; the literature reports good immediate outcomes, with improvements in 72.5% of patients and a low morbidity rate. It does have limitations, however, as it is not possible to ensure proper CSF flow through the catheter or the absence of flow reveral. [5][6][10][11]

In this study we found that syrinx shunting as the most successful procedure for posttraumatic syringomyelia. it is based on clinical improvements such as postoperative improvements in sensory, motor and autonomic functions. However, some literature reported a high rate of recurrence at long-term follow-up in posttraumatic syringomyelia patients who undergo syrinx shunting procedures. In recent years, these techniques have been criticised for their high recurrence rates (up to 92% at 3 years31) and the need for additional surgical procedures and frequent follow-up consultations. Studies comparing the efficacy of shunting and such other techniques as arachnolysis place much emphasis on this issue. Ghobrial et al. 32 reviewed articles published until 2015, finding a total of 410 surgery patients. The authors concluded that older age and the type of intervention (patients undergoing shunting are 7 times more likely to experience recurrences than those treated with arachnolysis) are significantly associated with symptom recurrence.[13][14][15]

On the other hand, although arachnoid lysis and duraplasty were the most common procedures in this study, the results suggest otherwise. Of the total 59 patients, only 49.1% of patients experienced clinical improvement on short-term follow-up. Unfortunately, there is no data in the journal about the long-term outcome for this procedure. Another series published by Klekamp focused on post-traumatic syringomyelia; several techniques were used in the same centre, as the surgeon decided to replace shunting with arachnolysis. That series shows that patients undergoing shunting usually need closer follow-up and confirms the clinical effectiveness of both interventions; which show differences in the duration of the effects This explains why shunting is now only used as rescue therapy when arachnolysis cannot be performed or has failed.[2][5][14][17]

At present, various surgical procedure such as syrinx shunting, subarachnoid bypass, arachnolysis with duraplasty, and cordectomy have been widely used in posttraumatic syringomyelia, however the efficacy still questionable and the rate of recurrency was high. On the other hand, cell therapy with MSCs seems to provide new promise for patients with SCI and it is necessary to know the selection of patients who can get benefit from these new techniques. In this study, we recorded 100% success rate of 7 patients who undergo cell therapy with Injection Mesenchymal stromal cells intra lesion. In any case, new studies with a greater number of cases are required. [17][18]

Conclusion

We analyze outcome for each procedure such as surgical, conservative, and also cell therapy. Surgical approach for posttraumatc syringomyelia has various technique but the efficacy and success rate remain questionable in long-term follow up. Athough the overall outcomes of the surgical procedure for PTS are still unsatisfactory, the syrinx shunting procedure seems to be considered as the most effective surgical procedure. Other therapeutic strategies such as MSCs is considered promising but still require further researches with larger sample sizes.

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