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Cervical Diastematomyelia: A Case Presentation and Systematic Review

Jeff F. Zhang, MD[1] Oleksandr Strelko, BS[2,*] Oleksandr Komarov, MD[3,*] Viktoriia Kuts-Karpenko, MD[4] Jonathan A Forbes, MD[5] Ostap Fedorko, MD[4] Luke D. Tomycz, MD[6]

[1] Department of Neurosurgery, SUNY Upstate Medical University, Syracuse, New York
[2] Stritch School of Medicine, Loyola University, Maywood, Illinois
[3] Institute of Postgraduate Education, Bogomolets National Medical University, Kyiv, Ukraine
[4] Clinical Municipal Communal Emergency Hospital, Lviv, Ukraine
[5] Department of Neurosurgery, University of Cincinnati College of Medicine, Cincinnati, Ohio
[6] Epilepsy Institute of New Jersey, Jersey City, New Jersey

Address for correspondence Jeff F. Zhang, MD, Department of Neurological Surgery, 750 East Adams Street, Syracuse, NY 13210 (e-mail: zhangjef@upstate.edu).

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Abstract

Diastematomyelia is a rare congenital disorder characterized by the separation of the spinal cord by an osseocartilaginous or fibrous septum. While diastematomyelia has been reported to be more common in the thoracic and lumbar regions, the true incidence of cervical diastematomyelia is currently unknown. In this study, we conducted the most comprehensive systematic review to date of all other case reports of diastematomyelia to better characterize the incidence of cervical diastematomyelia and provide comprehensive statistics on the clinical characteristics of diastematomyelia generally. Ninety-one articles were included in our study, which comprised 252 males (27.9%) and 651 females (72.0%) (and one patient with unspecified gender). In 507 cases, the vertebral level of the diastematomyelia was described, and we recorded those levels as either cervical ($n = 8$, 1.6%), thoracic ($n = 220$, 43.4%), lumbar ($n = 277$, 54.6%), or sacral ($n = 2$, 0.4%). In 719 cases, the type of diastematomyelia was specified as either Type I ($n = 482$, 67.0%) or Type II ($n = 237$, 33.0%). Our study found that diastematomyelia has been reported in the cervical region in only 1.6% of cases, and we provide comprehensive data that this disorder occurs in female-to-male ratio of approximately 2.6:1 and Type I versus Type II diastematomyelia in an estimated ratio of 2:1.

Keywords
- diastematomyelia
- neurosurgery
- spinal cord malformation
- spine
- systematic review
- Ukraine

Introduction

Diastematomyelia (also known as split cord malformation [SCM] or diplomyelia) is a rare congenital disorder characterized by the separation of the spinal cord by an osseocartilaginous or fibrous septum. While the total incidence of spinal dysraphism is estimated to be one to three cases per 1,000 live births,[1] the true incidence of diastematomyelia is unknown, though thought to occur in approximately 5% of congenital spine abnormalities.[2]

SCM is classified into two types: Type I SCM, in which the two hemicords are contained within two dural sacs divided by an osseous or cartilaginous septum, and Type II SCM, in

* *Both contributed equally as co-second authors.*

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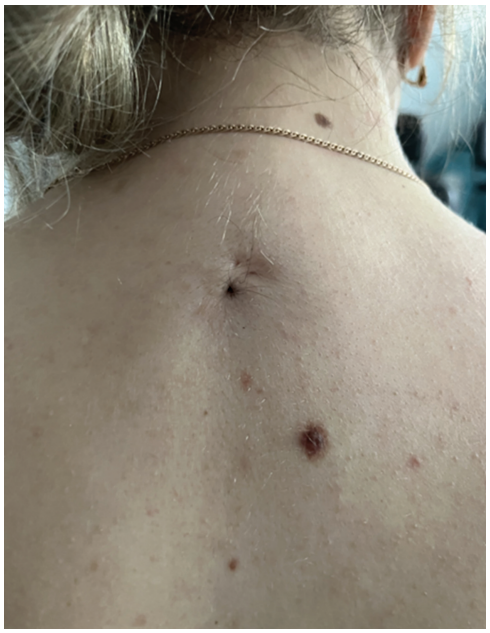

Fig. 1 Localized hypertrichosis on the patient's dorsal cervical region overlying the location of the diastematomyelia.

which a single dural tube contains both hemicords separated by a fibrous median septum.[3] Diastematomyelia is usually diagnosed in childhood and associated with other congenital spine deformities in 85% of cases, such as scoliosis, tethered cord, syringomyelia, spina bifida, Chiari 2 malformation, spinal lipoma, or dermoid cyst.[4] Type I diastematomyelia is more frequently associated with other congenital anomalies than Type II, and surgical intervention is commonly indicated for Type I patients due to symptom progression resulting from impingement of the rigid septum on the spinal cord, associated adhesions, and increasing scoliosis.[5] Patients with Type II diastematomyelia usually only require surgery when there is a significant change in scoliosis or neurological function, and symptoms in these patients tend to be milder due to the midline septum being fibrous in composition.[5]

While diastematomyelia has been reported to be more common in the thoracic and lumbar regions, the incidence of cervical diastematomyelia is thought to be extremely rare, with very few cases presented in the medical literature. In this case study, we report a patient who presented to neurosurgery clinic in Lviv Ukraine for upper extremity radicular pain during a US–Ukraine neurosurgery partnership mission (the Co-Pilot Project)[6] and was found to have cervical diastematomyelia on imaging. We also present a thorough systematic review of all other case reports of diastematomyelia in the medical literature in order to better characterize the incidence of cervical diastematomyelia.

Case Presentation

A 31-year-old female presented to the neurosurgery clinic with complaints of intermittent right shoulder pain with radiation down her arm and associated right upper extremity hypoesthesia. The patient noted that she had had these symptoms for many years but was concerned due to increased frequency and migration of the pain from her arm to the dorsal cervical region. The patient denied any medical conditions requiring medication or any family history of connective tissue or neurological diseases. The patient noted a surgical history of spina bifida treatment at 7 months of age (operative details for this surgery were unavailable), requiring 2 to 3 days of postoperative hospitalization and no complications at the time of discharge.

On physical examination, the patient had noted 4/5 right hand grip and 4/5 right arm extension weakness. Lower extremity motor and neurological functions were normal, but the patient noted that her right lower extremity was approximately 3 to 4 cm shorter than her left lower extremity. A small tuft of hair in the patient's dorsal cervical region was observed on examination (►**Fig. 1**), from which the patient reported her muscle spasms and pain originated.

CT myelography was performed and revealed a noncontrast-enhancing bony lesion splitting the spinal cord into two asymmetric hemicords at the C6 to C7 vertebral levels (►**Fig. 2**). No other structural anomalies were found on imaging.

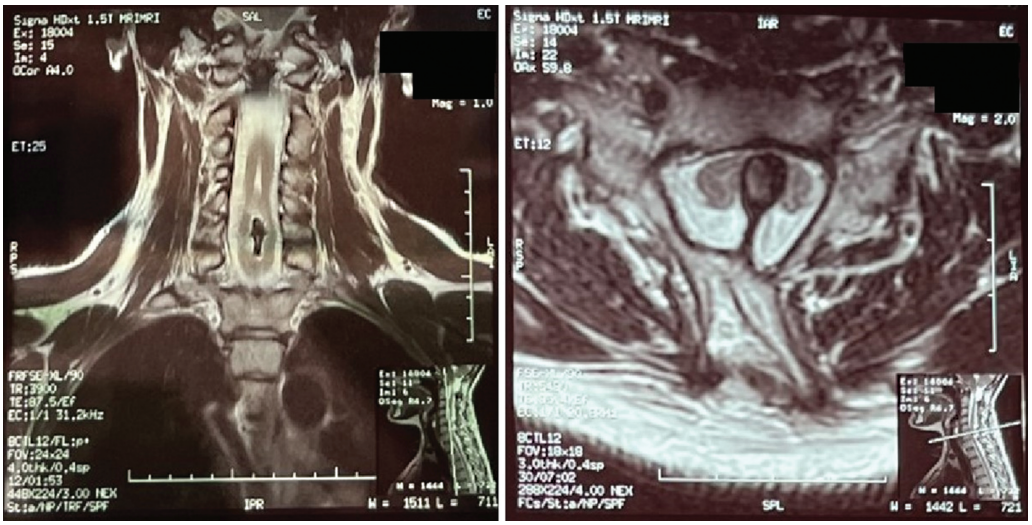

Fig. 2 Coronal and axial views of the cervical vertebrae at the level of the diastematomyelia lesion.

Due to the patient's symptoms being well-controlled with occasional use of NSAIDs, surgical intervention was not thought to be warranted at the time of the interview. The patient agreed with the course of action and was counseled to seek physical therapy and follow-up for any progression of her symptoms.

Methods

We conducted a systematic search using the PubMed database for all full-text reports in the English language describing patients with diastematomyelia. Searches were performed for all articles with the term "diastematomyelia" in their title. Studies were included in our review if patients described in the case report or series had a confirmed diagnosis of diastematomyelia on imaging. All relevant studies were reviewed and information related to the number of patient(s) described, their sex and age, the spinal level of the lesion, Type I versus Type II diastematomyelia, patient treatment, and clinical outcome were recorded. The search strategy used for study selection is represented by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram in ►**Fig. 3**.

Results

Two-hundred and fourteen results were provided by the PubMed database search. Of those 214 articles, 123 results were excluded due to the full article text not being available ($n = 55$), the article referring to a prenatal patient for whom a neurological examination was not possible ($n = 24$), the article not presenting a case report or series ($n = 21$), the article missing critical details ($n = 19$), or the article presenting a case of triplomyelia ($n = 3$) or diastematomyelia in a rachipagus twin ($n = 1$). Ninety-one studies matching the inclusion criteria described in the Methods were included for data collection and further analysis. These results are shown in ►**Table 1**.

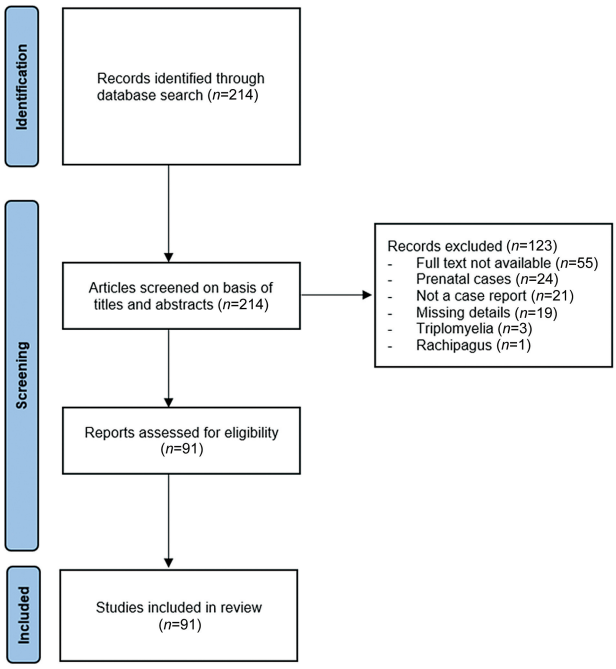

Fig. 3 PRISMA flow diagram describing the search strategy used for study inclusion in this systematic review.

The 91 articles included in our study comprised reports from 904 total patients with a mean age of 23.1 ± 22.0 years. A total of 252 males (27.9%) and 651 females (72.0%) (and one patient with unspecified gender) were included in our study. In 507 cases, the vertebral level of the diastematomyelia was described, and we recorded those levels as either cervical ($n = 8$, 1.6%), thoracic ($n = 220$, 43.4%), lumbar ($n = 277$, 54.6%), or sacral ($n = 2$, 0.4%). In 719 cases, the type of diastematomyelia was specified as either Type I ($n = 482$, 67.0%) or Type II ($n = 237$, 33.0%). Of 529 patients for whom follow-up data were available, 420 patients (79.4%) underwent surgical treatment for diastematomyelia, and 305 of those patients (72.6%) reported improvement in their neurological symptoms postoperatively. These findings are presented in ►**Table 2**.

Discussion

Consistent with previous reports of diastematomyelia found in the medical literature, the incidence of cervical diastematomyelia was found to be extremely rare, accounting for only 1.6% of all cases of diastematomyelia. The results of our comprehensive review of the literature also found that diastematomyelia has an approximately 2.5:1 predilection for females versus males and occurs as Type I versus Type II SCM in a 2:1 ratio.

The etiology of diastematomyelia is uncertain but thought to be related to abnormalities in the formation of the neural tube during the 4th week of development.[5] Adhesions between ectodermal and endodermal tissues lead to the formation of an accessory neurenteric canal in the midline of the neural tube, which results in the separation of the growing spinal cord into two hemicords as the notochord elongates rostrally.[7] These adhesions simultaneously prevent the complete involution of fibrous septations and developmental fistulas, resulting in the formation of cysts, lipomas, and fistulas[8] and cause disruptions in the associated development of the surrounding vertebrae, accounting for the high proportion of comorbid spinal malformations seen in diastematomyelia patients.[5] The accessory canal then forms the basis for the migration of mesenchymal cells which subsequently develop into the bony or cartilaginous septa seen in Type I SCM patients.[9]

While the majority of patients undergoing surgical treatment for diastematomyelia (72.6%) saw improvements in pain severity, motor function, and/or neurological symptoms, the performance of prophylactic surgery for patients with incidental findings of diastematomyelia (particularly patients with Type I SCM) on imaging is controversial.[8] Surgical removal of an osseous septum can cause damage to the spinal cord, especially in young children, and has been reported to result in postoperative worsening of neurological symptoms in a few cases.[10] Though other studies have shown that postsurgical prognoses for patients with Type I SCM are

Table 1 Systematic review of cases of diastematomyelia

Author	*n*	Sex/Age	Spinal level	Type	Treatment	Outcome
Ritchie and Flanagan 1969[11]	8	M: 2, F: 6 1 wk–9 y	T: 4 L: 3 U: 1	II: 8	Surgery: 8	Improvement: 2/8
Huang et al, 2013[12]	156	M: 47, F: 109 Mean = 4.5 y	C: 2 T: 82 L: 72	I: 123 II: 33	Surgery: 121 nonsurgical: 35	Improvement: (I: 96/123), (II: 0/33)
Kachewar and Sankaye 2014[13]	2	M: 2 (17 y and 1 y)	T: 1 L: 1	U	U	U
Sack and Khan 2016[14]	1	F: 1 (29 y)	T: 1	II: 1	U	U
Gbadamosi et al, 2022[15]	1	F: 1 (U)	L: 1	I: 1	Nonsurgical	U
Russell et al, 1990[16]	45	M: 12, F: 33 Mean = 37.8 y	U	U	Surgical: 24 Nonsurgical: 21	Improvement: (surgical: 23/24)
Tizard 1957[17]	1	F: 1 (3 y)	C: 1	U	Nonsurgical	U
Saini and Singh 2009[18]	1	M: 1 (22 d)	L: 1	I: 1	U	U
Maebe et al, 2018[19]	1	F: 1 (72 y)	L: 1	I: 1	Nonsurgical	U
Hao et al, 2022[20]	1	F: 1 (18 y)	L: 1	I: 1	Surgical	Improvement
Bekki et al, 2015[21]	1	F: 1 (14 y)	T: 1	I: 1	Surgical	Improvement
Ge et al, 2020[22]	1	M: 1 (36 y)	L: 1	I: 1	Surgical	Improvement
Albulescu et al, 2016[23]	1	U: 1 (45 y)	L: 1	I: 1	U	U
Cheng et al, 2012[5]	138	M: 34, F: 104 Mean = 15.7 y	U	I: 106 II: 32	Surgical: 112 Nonsurgical: 26	Improvement: (surgical: I: 91/96), (surgical: II: 8/16)
Constantinou 1963[24]	1	F: 1 (23 y)	L: 1	I: 1	Nonsurgical	U
Hamidi and Foladi 2019[25]	1	M: 1 (48 y)	L: 1	I: 1	Nonsurgical	U
Apostolopoulou et al, 2021[26]	1	F: 1 (5 y)	L: 1	I: 1	Surgical	Improvement
Kapsalakis 1964[27]	2	F: 2 (6 y and 5 y)	L: 2	I: 2	Surgical	Improvement: 1/2
Vissarionov et al, 2018[28]	20	M: 8, F: 12 Mean = 9.2 y	T:15 L: 5	U	Surgical: 17 Nonsurgical: 3	Improvement: (surgical: 17/17)
Hood et al, 1980[29]	60	M: 13, F: 47 Mean = 4.7 y	T: 24 L: 36	U	Surgical: 51 Nonsurgical: 9	Improvement: (surgical: 20/51)
Meena et al, 2018[30]	1	F: 1 (15 mo)	L: 1	I: 1	Surgical	No improvement
Lersten et al, 2017[31]	1	F: 1 (50 y)	L: 1	I: 1	Nonsurgical	U
Winter et al, 1974[32]	27	M: 6, F: 21 Mean = 6.5 y	T: 8 L: 19	U	Surgical: 22 Nonsurgical: 5	Improvement: (surgical: 5/19)
Srinivasan et al, 2020[33]	1	F: 1 (55 y)	C: 1	II: 1	Nonsurgical	Improvement
Kim et al, 1994[34]	5	M: 3, F: 2 Mean = 14.8 y	L: 5	I: 5	Surgical: 5	Improvement: 4/5
Singh et al, 2015[35]	1	F: 1 (3 mo)	T: 1	I: 1	Surgical	U
Mamo et al, 2021[36]	1	M: 1 (50 y)	T: 1	I: 1	Nonsurgical	U
McNeil et al, 2018[37]	1	F: 1 (3 y)	L: 1	I: 1	U	U
Hader et al, 1999[38]	1	F: 1 (16 y)	L: 1	I: 1	Surgical	U
Alimli et al, 2015[39]	1	F: 1 (4 y)	T: 1	I: 1	Surgical	U
Huang et al, 2014[40]	82	M: 17, F: 65 Median = 6 y	T: 50 L: 32	I: 82	U	U
Khurram et al, 2021[41]	1	M: 1 (38 y)	L: 1	I: 1	U	U
Shorey 1955[42]	1	M: 1 (12 y)	L: 1	I: 1	Surgical	Improvement

(*Continued*)

Table 1 (*Continued*)

Author	*n*	Sex/Age	Spinal level	Type	Treatment	Outcome
Azimi and Mohammadi 2013[43]	1	M: 1 (53 y)	L: 1	I: 1	Surgical	Improvement
Gavriliu et al, 2014[44]	2	M: 1 (12 y), F: 1 (7 y)	T: 1 L: 1	I: 2	Surgical	Improvement: 1/2
Scotti et al, 1980[8]	21	M: 8, F: 13 Mean = 7.5 y	L: 21	I: 5 II: 15	Surgical: 15 Nonsurgical: 6	U
Kansal et al, 2011[45]	1	M: 1 (1.5 y)	L: 1	I: 1	Surgical	No improvement
English and Malthy 1967[46]	2	M: 1 (48 y), F: 1 (32 y)	L: 2	U	Surgical: 1 Nonsurgical: 1	Improvement: (surgical: 0/1) No improvement: (nonsurgical: U)
Patankar et al 2000[47]	1	F: 1 (5 y)	T: 1	I: 1	Surgical	Improvement
Gan et al 2007[48]	17	M: 8, F: 9 Mean = 3.4 y	T: 5 L: 12	I: 17	Surgical: 17	Improvement: 5/17
Yamanaka et al 2001[49]	1	U: 1 (6 d)	L: 1	I: 1	Surgical	Improvement
Beyerl et al 1985[50]	1	M: 1 (34 y)	C: 1	I: 1	Surgical	Improvement
Sandhu et al, 2021[51]	1	M: 1 (25 y)	T: 1	II: 1	Nonsurgical	Improvement
Chembolli 2015[52]	1	F: 1 (16 y)	T: 1	I: 1	Surgical	Improvement
Elmaci et al 2001[53]	1	M: 1 (42 y)	L:1	I: 1	Surgical	Improvement
Xu et al 2023[54]	1	F: 1 (17 y)	L: 1	I: 1	Surgical	Improvement
Zaleska-Dorobisz et al 2010[55]	1	F: 1 (78 y)	L: 1	I: 1	U	U
Sheehan et al,2002[56]	1	F: 1 (38 y)	T: 1	I: 1	Surgical	Improvement
Kanbur et al 2004[57]	1	M: 1 (12 y)	L: 1	I: 1	Nonsurgical	No improvement
Tubbs et al 2004[58]	1	F: 1 (18 y)	T: 1	I: 1	Surgical	No improvement
Shivapathasundram and Stoodley 2012[59]	1	F: 1 (8 y)	T: 1	I: 1	Surgical	Improvement
Parmar et al 2003[60]	1	F: 1 (34 y)	L: 1	I: 1	Surgical	Improvement
Senkoylu et al 2019[61]	1	F: 1 (4 y)	L: 1	I: 1	Surgical	Improvement
Tsitsopoulos et al 2006[62]	1	F: 1 (44 y)	L: 1	I: 1	Nonsurgical	No improvement
Pettorini et al 2007[63]	1	M: 1 (2 y)	T: 1	I: 1	Surgical	Improvement
Lewandrowski et al 2004[64]	1	F: 1 (44 y)	L: 1	I: 1	Surgical	Improvement
Ross et al 1988[65]	1	M: 1 (63 y)	S: 1	I: 1	Nonsurgical	No improvement
Porensky et al,2007[66]	1	F: 1 (54 y)	T: 1 L: 1	I: 2	Surgical	Improvement
Filippi et al 2010[67]	3	M: 1 (67 y), F: 2 (53 and 49 y)	T: 1 L: 2	I: 1 II: 2	U	U
Senel et al 2008[68]	1	F: 1 (14 mo)	T: 1	I: 1	Surgical	U
Shen et al 2016[69]	214	M: 61, F: 153 Mean = 14.2 y	U	I: 73 II: 141	Surgical: 214	U
Sharma et al 1997[70]	1	M: 1 (15 y), F: 1 (9 y)	T: 1 L: 1	I: 2	Surgical: 2	U
Sgouros 2010[71]	1	F: 1 (3 y)	L: 1	I: 1	Surgical	Improvement
Wenger et al, 2001[72]	1	F: 1 (38 y)	L: 1	II: 1	Nonsurgical	Improvement
Kilickesmez et al, 2004[73]	1	F: 1 (7 y)	L: 1	I: 1	Surgical	Improvement
Kaminker et al 2000[74]	1	M: 1 (38 y)	L: 1	I: 1	Surgical	Improvement
Ak et al, 2014[75]	1	F: 1 (10 y)	L: 1	I: 1	Surgical	Improvement

Table 1 (*Continued*)

Author	*n*	Sex/Age	Spinal level	Type	Treatment	Outcome
Ohwada et al, 1989[76]	1	M: 1 (29 y)	C: 1	I: 1	Surgical	Improvement
Morelli and Shalick 2011[77]	1	F: 1 (29 y)	L: 1	I: 1	Surgical	No improvement
Macht et al 2012[78]	1	M: 1 (57 y)	L: 1	I: 1	Nonsurgical	U
Kanagaraju et al 2016[79]	1	F: 1 (15 y)	L: 1	I: 1	Nonsurgical	Improvement
Boussaandani et al 2011[80]	1	F: 1 (33 y)	L: 1	I: 1	Nonsurgical	U
Armstrong et al, 2016[81]	1	F: 1 (49 y)	L: 1	I: 1	Nonsurgical	U
Giordano et al 2016[82]	1	F: 1 (43 y)	T: 1	II: 1	U	U
Sharma et al 2005[83]	1	F: 1 (18 mo)	L: 1	I: 1	Surgical	Improvement
Sedzimir et al, 1973[84]	1	M: 1 (22 mo)	L: 1	I: 1	Surgical	Improvement
Callari and Arrigo 2009[85]	2	F: 2 (80 and 59 y)	L: 2	U	Nonsurgical	Improvement: 2/2
Kramer et al, 2009[86]	1	F: 1 (54 y)	L: 1	I: 1	Surgical	No improvement
Roche and Vignaendra 2006[87]	8	M: 3, F: 5 Mean = 53.8 y	L: 7 S: 1	U	U	U
Uzumcugil et al 2003[88]	18	M: 2, F: 16 Mean = 20 mo	T: 9 L: 9	U	Surgical	U
Burnei et al 2015[89]	1	F: 1 (18 y)	L: 1	I: 1	Surgical	Improvement
Hung et al 2010[90]	1	F: 1 (2 d)	L: 1	I: 1	Surgical	U
Turgut and Doger 2008[91]	1	M: 1 (1 d)	L: 1	I: 1	Surgical	U
Bale 1973[92]	1	F: 1 (6 d)	T: 1	I: 1	Surgical	U
Korinth et al 2004[93]	1	M: 1 (2 y)	C: 1	I: 1	Surgical	No improvement
Mendez et al, 2009[94]	1	F: 1 (88 y)	L: 1	I: 1	Nonsurgical	Improvement
Yamada et al,1996[95]	1	F: 1 (2 y)	L: 1	I: 1	Surgical	Improvement
Lourie and Bierny 1970[96]	1	F: 1 (7 y)	T: 1	I: 1	Surgical	U
Ugarte et al, 1980[97]	2	F: 2 (1 d and 1 d)	T: 2	U	Surgical	Died: 2/2
Okada et al 1986[98]	1	M: 1 (19 y)	C: 1	I: 1	Nonsurgical	U
Azhar et al, 1996[99]	1	M: 1 (35 y)	L: 1	I: 1	Surgical	Improvement

Note: Systematic review of case reports and series describing patients with diastematomyelia. "*n*" number of patients described in the study, the sex (M = male, F = female, U = unknown) and age of included patients, the spinal location of the lesion (C = cervical, T = thoracic, L = lumbar, U = unknown), Type I versus Type II spinal cord malformation, treatment, and clinical outcome were recorded.

significantly improved compared to patients with Type II SCM,[5] these qualifications of "improvement" versus "no improvement" compared to preoperative status are frequently complicated in studies by patients who had minimal symptoms prior to surgery (especially in patients with Type II SCM). Additionally, it has been suggested that the symptoms characteristic of diastematomyelia are related to some intrinsic myelodysplasia resulting from abnormal development rather than the presence of a bony spur in itself, as the location of a spur (resulting in the asymmetric compression of one hemicord) is not in itself predictive of the laterality or severity of symptoms.[8] This is also corroborated by reports of patients with Type II SCM who did not have any spur detected at all on imaging but nonetheless complained of significant neurological symptoms.[8]

Limitations of our study include an inability to confirm diagnoses of diastematomyelia from an independent review of imaging in all of our included cases, and the difficulty to adequately distinguish surgical outcomes for patients with Type I versus Type II SCM due to inconsistent reporting of results across studies and the often mixture of these two patient populations in the studies that did report surgical outcomes. The decision for surgical treatment is currently based on symptom severity or when necessary in the context of correcting concurrent spinal deformities. The establishment of clearer guidelines for surgical intervention for diastematomyelia requires further studies and trials beyond the scope of this present review.

Conclusion

Cervical diastematomyelia is an extremely rare condition, accounting for 1.6% of all cases of diastematomyelia. Clinical correlations for establishing more rigorous guidelines related

Table 2 Patient demographics

	Population ($n = 904$)
Age (mean ± SD)	23.1 ± 22.0
Gender	
Male (*n*, %)	252 (27.9)
Female (*n*, %)	651 (72.0)
Unspecified (*n*, %)	1 (0.1)
Vertebral level	507 (56.1)
Cervical (*n*, %)	8 (1.6)
Thoracic (*n*, %)	220 (43.4)
Lumbar (*n*, %)	277 (54.6)
Sacral (*n*, %)	2 (0.4)
Pang criteria	719 (79.5)
Type I (*n*, %)	482 (67.0)
Type II (*n*, %)	237 (33.0)
Surgical treatment	420 (79.4)
Improvement (*n*, %)	305 (72.6)
No improvement (*n*, %)	115 (27.4)

Abbreviation: SD, standard deviation.
Note: Patient Demographics. Determinations of clinical improvement following treatment were calculated from the $n = 529$ patients for whom follow-up data were provided.

to surgical intervention in cases of diastematomyelia require further studies to clarify best practices.

Authors' Contributions

Conceptualization: All authors.
Data curation: J.F.Z.
Formal analysis: J.F.Z.
Funding acquisition: N/A
Investigation: All authors.
Methodology: All authors.
Project administration: J.A.F., O.F., and L.D.T.
Resources: N/A
Supervision: J.A.F., O.F., and L.D.T.
Validation: All authors.
Writing—original draft: J.F.Z., O.S., and O.K.
Writing—reviewing and editing: All authors.

Informed Consent

Written and verbal consents were obtained from all patients or their health care proxies for all aspects related to this report and prior to any procedures which were performed.

Data Sharing

Data supporting the findings of this study will be made available by the corresponding author upon request.

Conflict of Interest

None declared.

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