Child's Nervous System (2022) 38:1977–1986
https://doi.org/10.1007/s00381-022-05578-5

ORIGINAL ARTICLE

Split cord malformation concomitant with spinal teratoma without open spinal dysraphism

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Received: 24 February 2022 / Accepted: 24 May 2022 / Published online: 10 June 2022

Abstract

Purpose Split cord malformation (SCM) presenting concomitant with spinal teratoma without any open spinal dysraphism has rarely been reported in the literature. We aimed to make a systematic review and qualitative analysis of the literature about the topic and present the first case of SCM concomitant with spinal teratoma harboring papillary thyroid carcinoma (PTC) component.

Methods Two big search tools (Pubmed/MEDLINE) and Scopus were used. The search strategy was compatible to Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA). An exemplary case of ours was also presented.

Results There were 30 patients (15 pediatric and 15 adult). Female and male distribution was even. Median age of the patients was 18 years (range = 0–66 years). The most common presenting symptoms were back pain and lower limb weakness. Spinal teratoma and SCM mostly presented at thoracic/thoracolumbar region in children and lumbar region in adults. Surgical outcome was better in the children compared to the adults.

Conclusion Thoracolumbar region is the most common location for such entity in children, whereas lumbar region for the adults. Surgical resection should be done as much as possible under neuromonitorization. The resected material should be evaluated thoroughly not to miss any malign pathology. Surgical outcome is better when it is done at an early age.

Keywords Spinal teratoma · Diastematomyelia · Split cord malformation

Introduction

Split cord malformations (SCMs) are congenital spinal cord malformations that bear cleft within the spinal cord together with bony or soft tissue spicules arising from the spine [1]. There are two types of SCMs: type I, divided hemicords via an osteocartilaginous septum; type II, divided hemicords via fibrous septum. Hemicords reside in two separate dura and one uniform dura in type I and type II SCMs, respectively [2].

Split cord malformations are rare and have been reported in 1 of every 5000 live births [2–4]. They have commonly been reported together with thickened filum terminale, low lying spinal cord, syringomyelia, and kyphoscoliosis. Rarely, SCMs were observed also with lipoma, neuroenteric cyst, dermoid cyst, and dermal sinus tract [2, 5, 6]. Concomitant presentation of SCMs with spinal teratomas without open spinal dysraphism has rarely been reported in the literature [7]. However, there is no one paper dealing with large case series including both pediatric and adult patients giving descriptive statistics of the pathology. We presented an exemplary case of papillary thyroid carcinoma (PTC) residing within spinal teratoma concomitant with SCM. To the best of our knowledge, this phenomenon has never been reported in the literature. We aimed to grab attention of neurosurgeons, dealing with congenital malformations of spine and spinal cord, to such a rare entity with a systematic review and qualitative analysis of the literature.

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Materials and methods

The present study is a systematic review and qualitative analysis of the literature. We used two big search tools (Pubmed/MEDLINE) and Scopus. The search was conducted through a timeline including inception of the relevant databases and up to April 2022. The search strategy was compatible to Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (Fig. 1) [8]. The search protocol included the following terms: "split cord malformation" OR "diastematomyelia" and "spinal teratoma."

A total of 643 citations were reached up following database search. Of those 643 citations, 310 were eliminated as

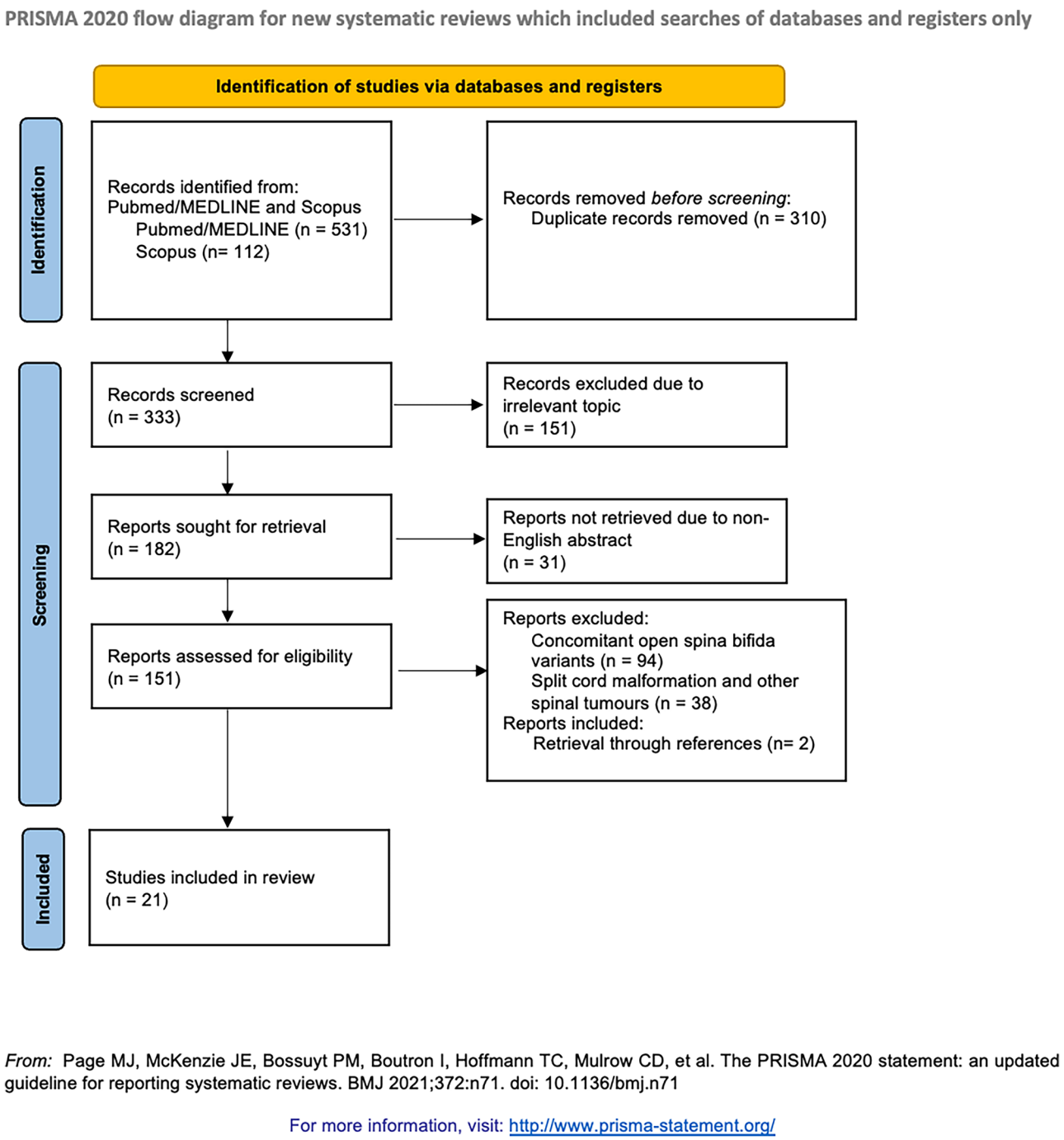

Fig. 1 Flow chart for the reviewing and data extraction of the literature

they were duplicate ones. In the present systematic review, we aimed to work on isolated SCM cases presented concomitant with spinal teratoma without any associated open spinal dysraphism (myelomeningocele, meningocele, lipomyelomeningocele, lipomeningocele). We excluded any cases of SCM presented with other kinds of spinal tumors (epidermod, dermoid, etc.) if there was no concomitant spinal teratoma. References of relevant citations were screened for any relevant, yet missed, citation through database search.

There were 21 citations about SCM presented concomitant with spinal teratoma, yet without open spinal dyraphism. Of those 21 citations, 9 (41%) were about pediatric, 11 (50%) were about adult, and 1 (9%) was about both pediatric and adult patients. We included one adult case of ours to the final qualitative analysis (Tables 1 and 2). Continuous and dichotomous variables were presented as median (with range) and absolute (with percentage) values. Fisher's exact test was used to make comparisons between dichotomous variables. Excel 2021 (for Mac, Microsoft, Redmond, WA, USA) was used for descriptive and qualitative statistics. An alpha value of < 0.05 was accepted as statistically significant.

Table 1 Pediatric patients with concomitant spinal teratoma and split cord malformation, presented in the literature

Author(s)/year	Age/gender	Clinical presentation	Location of teratoma	Location of SCM	Intervention	Outcome
Lemmen and Wilson/1951 [9]	11/F	Lower limb weakness	T6–T10, intradural	T6–T10	Subtotal removal	Improved for 2.5 months and died
Ugarte et al./1980 [10]	New born/F	Lower limb weakness	T2	T2	Total excision of the SCM and the tumor	Died due to systemic congenital anomalies
	New born/F	Dorsal and back mass	T6–L4	T6–L4	Excision of the tumor	Died due to systemic disease
Ersahin et al./1998 [11]	N/A	N/A	Cervicothoracic region	L2	Operated	N/A
Koen et al./1998 [12]	1.17/M	Decreased sensation in left lower limb	T4–T7, intradural	T4–T7	Total excision of the SCM and the tumor	N/A
Jarmundowicz et al./2004 [13]	13/F	Scoliosis	L1–L2	L1–L2	N/A	N/A
Uzum et al./2005 [14]	1/M	Lower limb weakness, constipation, hypertrichosis at lower back	L5–S1, intradural	L2–L4	Total excision of the tumor and the SCM	Complete recovery 2 years following the surgery
Suri et al./2006 [15]	8/M	Upper back pain, numbness below the clavicle, kyphoscoliosis	T2–T7, intradural	T2–T7	Total excision of the tumor and the SCM	Complete recovery 1.5 years following the surgery
Ye and Li/2008 [16]	9/F	Scoliosis	T10–T12, intramedullary	T9	Total excision of the tumor and the SCM	Complete recovery following the surgery
Sharma et al./2009 [17]	17/M	Right foot deformity, lumbosacral swelling	T12–L2, intramedullary	N/A	N/A	N/A
	8/M	Upper back pain	T1–T7, intramedullary	N/A	N/A	N/A
	13/F	Weakness of both lower limbs, frequency of micturition	T12–L2, intramedullary	N/A	N/A	N/A
	10/M	Low back pain	T11–L3, intramedullary	N/A	N/A	N/A
	1.4/M	Swelling back	L3–L5, intramedullary	N/A	N/A	N/A
Ersahin/2013 [18]	9/F	N/A	T12–L2, intramedullary	T12–L2	Total excision of the tumor and the SCM, with remnant of the lipoma	Complete recovery 15 years following the surgery

SCM split cord malformation, *F* female, *M* male, *N/A* not available

Table 2 Adult patients with concomitant spinal teratoma and split cord malformation, presented in the literature

Author(s)/year	Age/gender	Clinical presentation	Location of teratoma	Location of SCM	Intervention	Outcome
Rosenbaum et al./1978 [19]	49/M	Low back pain, bilateral lower limb pain, paresthesia & dysesthesia of left lower limb, decrease in sexual performance, difficulty in urination	T9, intramedullary	T9	Total excision of the tumor and the SCM	Recovery to near-normal status following the surgery
Garza-Mercado/1983 [20]	26/M	Increasing lower back and right lower limb pain, weakness & paresthesia of right lower limb for 1 year	L4, extradural with intramedullary epidermoid tumor	L2	Total excision of the tumor and the SCM	Recovery of most of the symptoms at the time of discharge
Conti et al./1984 [21]	24/F	Back pain, right lower limb weakness & paresthesia	L1–L2, intramedullary	N/A	Total excision	Improved
Elmaci et al./2001 [22]	42/M	Increasing pain & distal left lower limb weakness with neurogenic claudication for 1 month	L5, intradural	L5	Removal of the bony spur, cystic mass, and sectioning of the terminal filum	Partial resolution of the symptoms 1 year following the surgery
Tsitsopoulos et al./2006 [23]	44/F	Progressive lower limb weakness, gait disturbance, dysesthesia in the trunk & lower extremity	T8–T9, intradural & mostly intramedullary	L2–L3	Partial excision of teratoma, no intervention for the SCM	Better mobilization 1 year following the surgery
Mut et al./2007 [24]	34/F	Right lower limb weakness, left lower limb sensory deficit & urinary retention	L1–L2, intramedullary	L1–L2	Near total excision of the SCM and tumor, except the exophytic component	Fully recovery 3 months following the surgery
Sharma et al./2009 [17]	32/M	Lower limb pain & weakness, urinary incontinence	T10, intramedullary	N/A	N/A	N/A
	31/M	Numbness & unsteadiness of gait	T11, intramedullary	N/A	N/A	N/A
	30/M	Low back pain, difficulty in urination	L1–L3, intramedullary	N/A	N/A	N/A
Conti et al./2010 [25]	38/F	Low back & right lower limb pain, hypoesthesia on right hemithorax and right lateral side of the abdomen	L1–L2, intramedullary	L1–L2	Total excision of the tumor and the SCM	Full recovery 2 months following the surgery
Maiti et al./2010 [26]	18/M	Back pain, urinary problem, paresthesia in both lower limbs	L2–L3, intramedullary	L1	Partial excision of the tumor and excision of the SCM	Complete recovery 6 months following the surgery
Babu et al./2014 [7]	66/F	Back pain, lower limb stiffness & weakness, urinary & fecal incontinence	L3–L4, intradural extramedullary	L3	Total excision of the tumor	Complete recovery 1 year following the surgery
Kafadar et al./2016 [27]	48/F	Low back pain, lower limb weakness	L2–L3, intradural extramedullary	L2–L3	Patient refused surgery	N/A

Table 2 (continued)

Author(s)/year	Age/gender	Clinical presentation	Location of teratoma	Location of SCM	Intervention	Outcome
Ge et al./2020 [28]	36/M	Progressive weakness & paresthesia of left lower limb	L3, intradural	L3	Resection of SCM and the tumor	Improved near to normal 3 months following the surgery
Present case	34/F	Lower limb pain & weakness	L3–S1, intramedullary	L2–L3	Resection of the SCM and the tumor	Partial recovery 1 year following the surgery

SCM split cord malformation, *F* female, *M* male, *N/A* not available

Results

There were 15 (50%) pediatric and 15 (50%) adult patients in the final analysis. Age and gender were unclear for one pediatric patient. Median age of all the patients, pediatric, and adult patients were 18 years (range = 0–66 years), 8.5 years (range = 0–17 years), and 34 years (range = 18–66 years), respectively. There were 7 pediatric male (50%) and 7 pediatric female (50%) patients. There were 8 adult male (53%) and 7 adult female (47%) patients. Back pain, lower limb weakness and/or paresthesia, back mass, kyphosis, scoliosis, lower limb deformity, constipation, fecal incontinence, urinary problems, hypertrichosis at back, radiculopathy, neurogenic claudication, and sexual dysfunction were the main signs and symptoms reported in those patients with SCM concomitant with spinal teratoma. Back pain and lower limb weakness were the most common presenting findings.

Spinal teratomas presented mostly at thoracic region or thoracolumbar junction in pediatric patients ($n = 12$, 80%), whereas at lumbar region ($n = 10$, 66%) in adult patients. The difference between pediatric and adult patients in respect of teratoma location within the spine was statistically significant ($p = 0.03$). Intramedullary location was the most prevalent location for spinal teratomas ($n = 17$, 57%). Split cord malformation was mostly diagnosed at thoracic or thoracolumbar region ($n = 7$, 70%) in pediatric patients, whereas at lumbar region ($n = 10$, 91%) in adult patients. The difference in location of SCM between pediatric and adult patients was statistically significant ($p = 0.008$).

Treatment modality was unclear in 3 (10%) patients. One (3%) patient denied surgery. Outcome was unknown for 12 patients (40%). Three pediatric patients (20% of pediatric and 10% of all patients) died due to concomitant systematic diseases. Complete recovery was possible in all remaining pediatric patients ($n = 4/7$, 57%), whereas only 4/11 (27%) of the adult patients could manage to recover fully following the surgeries ($p = 0.63$) (Tables 1 and 2).

Exemplary case

A 34-year-old woman applied to our outpatient clinic with recent complaints of left hip pain and right foot weakness for the last 8 months. She began to feel weakness in her left foot, too. She had no urinary or defecation problem. She had motor deficits in her lower extremity most pronounced at her right side. Left S1 dermatomal region was hypoesthetic. Her lower extremity deep tendon reflexes were bilaterally hyperactive. There were no skin stigmata on the patient back.

On radiological examination, we observed SCM beginning at lower thoracic level. There was a bony septum at the L2–L3 levels. There was intradural lipoma infiltrating right hemicord, extending down to the back between the

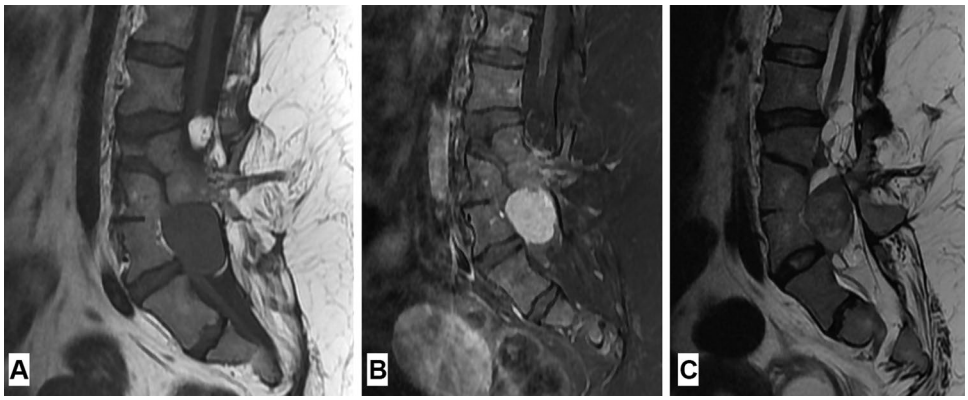

Fig. 2 T1-weighted (**A**), T1-weighted contrast enhanced fat suppressed (**B**), and T2-weighted lumbar spine MRI depicting intradural and extradural heterogenic mass lesion with vertebral formation and segmentation anomaly (**C**)

L3 and S1 levels. A homogenously enhanced isolated mass lesion with a dimension of 2 × 3.5 cm was observed within the defined area of interest (Fig. 2).

The patient was operated under surveillance of neuromonitorization. Laminectomies of L2 and L3 vertebrae were accomplished. Two hemicords splitted with a bony spur located between the L2 and L3 levels were observed. The bony spur was resected and durotomy of both hemicords was done to revise the dura as a whole piece. Fibrous bands tethering the spinal cord were released. Lipoma was observed as infiltrating posterior and medial aspects of the right hemicord. It was resected as much as possible leaving a thin remnant tissue, under guidance of neuromonitorization. Another mass lesion was arising from the right hemicord and filling whole spinal canal and pressing over the neural tissues. It was a paste-like, sandy colored mass and it was resected totally. The dura was closed primarily in watertight fashion. There was no abnormal signal or lost signal on neuromonitorization. She had no additional deficit following the surgery and she partly recovered under supervision of physical therapy sessions in 1 year.

Pathology confirmed the intradural intramedullary and extradural lipomatous mass lesion as benign fiprolipomatous tissue. The second biopsied, intramedullary lesion was observed to be a teratomatous lesion consisting of mature adipose tissue, fibrous tissue, and some glandular tissue resembling thyroid follicles, some of which lined with nonspecific columnar epithelium. Morphological and immunohistochemistry analysis confirmed the second mass lesion as papillary thyroid carcinoma developed in teratoma (Figs. 3, 4, 5 and 6). The patient was scanned with positron emission tomography (PET), and no primary or metastatic lesion of PTC was found. The patient was consulted to oncology and endocrinology departments and put on close surveillance for any recurrence of the disease. The patient was free of cancer at her 1st year control.

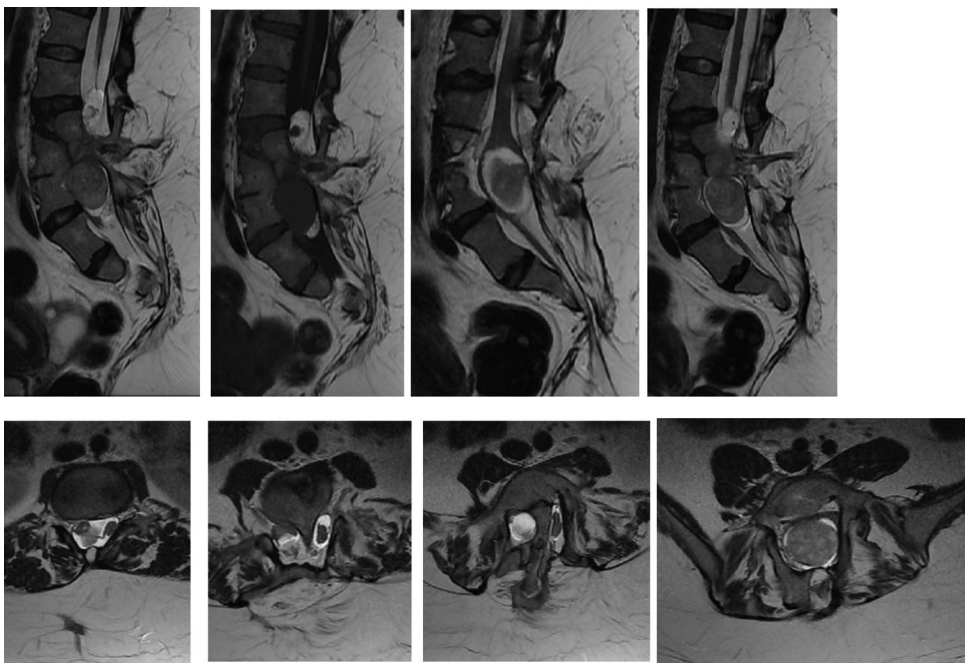

Fig. 3 Sagittal and axial views of SCM, spinal teratoma, and spinal lipoma

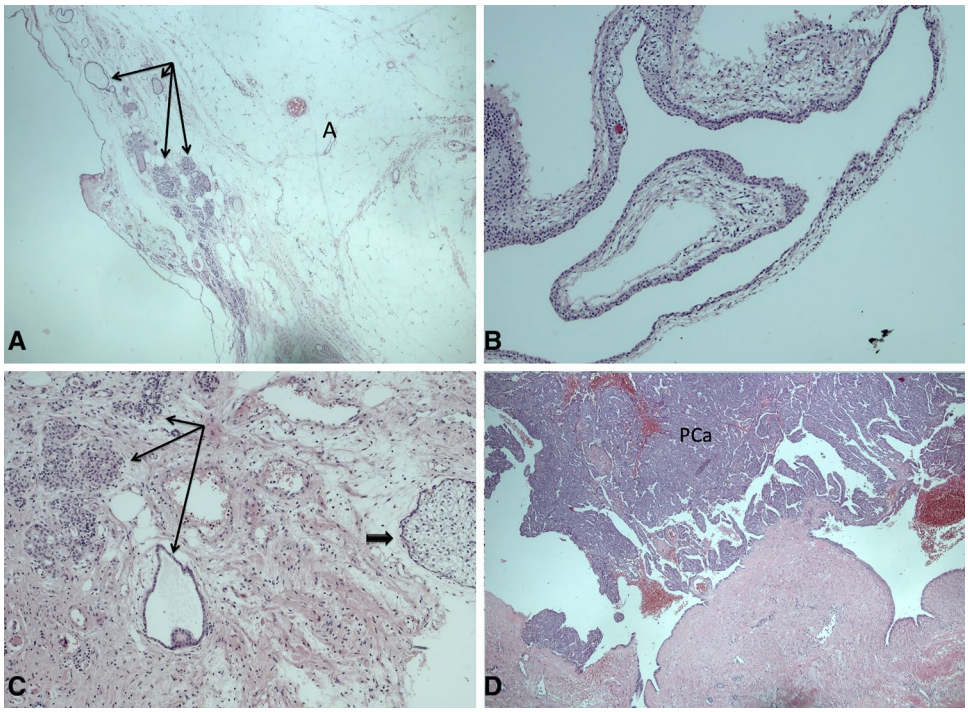

Fig. 4 Benign seromucinous glandular structures (arrows) and mature adipose cells (**A**) (**A**, H&E × 40 magnification), partially squamous cell-like structured epithelial cell lining of the benign cyst (**B**, H&E × 100 magnification), benign squamous glands (multiple thin arrows) and squamous epithelial cell clusters (thick arrow) of the cyst wall (**C**, H&E × 100 magnification), and papillary tumor (PCa) showing growing pattern within the cyst (**D**, H&E × 40 magnification)

Discussion

Split cord malformation or diastematomyelia is a congenital spinal cord malformation with two hemicords, splitted into two by a bony or fibrous tissue. The SCMs are classified as SCM-I or SCM-II depending on what type of tissue splitted the spinal cord: bony spur in SCM-I and fibrous band in SCM-II [2]. Split cord malformation presents early in human life, yet some rare cases presented in adults, even in elderly [29]. Traction on the conus determines the age of onset of symptoms in patients with SCM as a consequence of repetitive movements due to acute trauma or the aging process [29, 30].

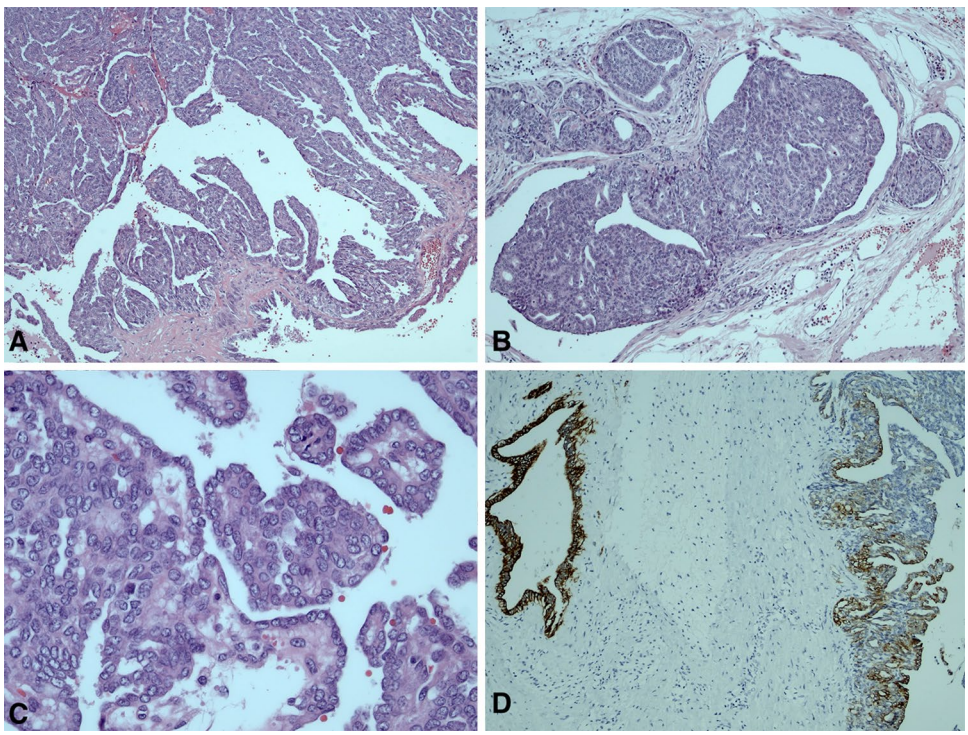

Fig. 5 Papillary tumor proliferation supported by fibrovascular stroma (**A**, H&E × 200 magnification), tumor part showing cystic-solid growing pattern within the cyst wall (**B**, H&E × 200 magnification), atypical epithelial cells with vesicular nucleus showing overlapping during tumoral proliferation (**C**, H&E × 400 magnification), and cytokeratin 19 positivity of the tumor (**D**, IHC × 100 magnification)

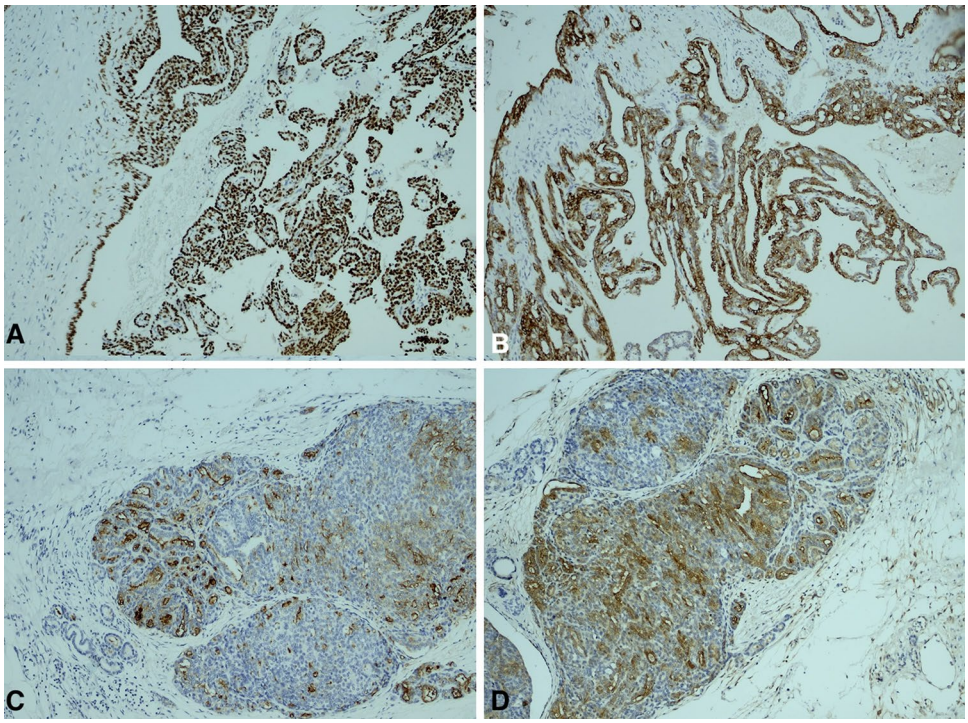

Fig. 6 Intranuclear TTF-1 positivity (**A**, IHC × 100 magnification), thyroglobulin positivity (**B**, IHC × 100 magnification), HBME-1 positivity (**C**, IHC × 100 magnification), and galectin-3 positivity (**D**, IHC × 100 magnification) within the tumor

Spinal cord lipomas are progressive congenital disorders of the spinal cord [31]. Asymptomatic lipomas have a likelihood of 33–40% to deteriorate over 9–10 years [32, 33]. Surgery, even in asymptomatic subjects, should be aimed to resect spinal cord lipomas as much as possible [31]. There are subtypes of spinal cord lipomas depending on how they develop during embryogenesis: dorsal lipoma, transitional lipoma, terminal lipoma, and chaotic lipoma [31]. Details of each lipoma type are beyond the scope of the present study. Nevertheless, the one presented in the present case was dorsal lipoma, which develops if a dorsal defect develops in the dura and neural tube during normal ascent of spinal cord in embryogenesis phase. The surrounding mesenchyme invades the defect and forms a fibrofatty stalk that attaches to the sliding neural tube and entraps it [31]. It is a mistimed disjunction happening during primary neurulation [34]. This pathogenesis would also explain concomitant teratoma development in the present case. The ependymal lining of the neural tube might induce the invading mesenchyme to form fat, muscles, collagen, cartilage, and bone [31]. Another possible theory for pathogenesis of spinal lipoma will be explained in the forthcoming paragraphs.

Teratomas are neoplasms of multipotential cells of all three germ cell layers with power of autonomous growth [35]. Spinal teratoma, except sacrococcygeal one, is very rare (0.2–0.5% of all spinal cord tumors and 2% of all central nervous system teratomas) [35]. They mostly present at thoracolumbar segment of the spine. Adult-onset presentation is rare and mostly observed in men in their 4th and 5th decades of life [35]. The most common presentation is back pain, limb weakness, and bowel/bladder disturbance with gradual symptom onset. Back pain and lower limb weakness were the two most common presenting symptoms in the present series. They could present intramedullary, extramedullary, or extradurally [17, 36–39]. Intramedullary area was the most pronounced location of the spinal teratomas presented concomitant with SCM without any open spinal dysraphism.

Spinal teratomas have diverse imaging patterns on MRI due to heterogenous cell content. Generally, spinal teratomas appear as hyperintense lesions on T1-weighted MRI sequences because of their fat content. However, lesions with hypointense or isointense T1-weighted imaging properties have also been reported [35]. Concomitant congenital anomalies have been reported together with spinal teratomas [40]. Those dysraphic spinal lesions could be spina bifida, myelomeningocele, dermal sinus, tethered cord syndrome, and split cord malformation (9.6% of all spinal teratomas) [35, 41, 42]. Concomitant presentation of SCM with spinal teratoma without any open spinal dysraphism is rare, there have been thirty cases including the present case [7, 9–27]. Split cord malformations are more pronounced in female gender (specifically for SCM type I) [18]. Female and male distribution was even in the present case series. Spinal teratomas were observed at any spine level, yet with an important difference between pediatric and adult patients. They presented at thoracic or thoracolumbar region in pediatric patients, whereas at lumbar region in adult patients, same as SCM location in those patients. Lumbar location

of SCM with spinal teratoma might have caused delay in diagnosis of those adult cases.

Lellouch-Tubiana et al. [43] analyzed pathology specimens of 234 patients with former diagnosis of intraspinal lipoma. They found an interesting fact that those lipomas had contained foci of endodermal, mesodermal, and ectodermal origin, depicting a teratomatous component. This could explain concomitant presentation of teratoma within lipoma bed in the exemplary case. Spinal teratomas emerging concomitant with spinal dysraphic lesions have been presumed to have a dysembryogenic origin as other tumors (such as medulloblastoma) that occur in the midline [12]. This dysembryogenic theory is interrelated with pathogenesis of SCM: SCMs occur secondary to ontogenetic error happening during closure of primitive neuroenteric canal. An accessory neuroenteric canal emerges through trilaminar embryonic disc. It connects the amniotic cavity and the yolk sac. This neuroenteric canal ends up with split notochord and split neural plate. Endodermal and mesenchymal cells line up inside the neuroenteric canal. If endodermal cells survive up to the birth, then neuroenteric cysts would emerge. If mesenchymal cells survive, which have pluripotential capacity, then spinal teratomas would appear. If the neuroenteric canal has ectodermal connection, then congenital dermal sinus would be present at birth [44–46]. It should be kept in mind that spinal teratomas could present at remote side from the congenital spinal malformation and all spine should be scanned in such scenario [35]. Complete resection of spinal teratoma is warranted with attention given to possible intimate relation with spinal cord parenchyma [47]. Malignant transformation of intramedullary teratoma concomitant with SCM has rarely been reported in the literature that would necessitate complete resection of the tumor as much as possible under intraoperative electrophysiologic monitoring [9, 24].

Papillary thyroid carcinoma is one of the subtypes of differentiated thyroid carcinoma [48]. It composes the most common variant (> 80%) of all thyroid malignancies [49]. Surgery is the main treatment body with adjuvant therapies depending on histological and genetic properties of the tumor [48]. Metastasis of PTC is mostly local to the regional lymph nodes, with rare occurrence of metastasis to distal sites (5–7% of the cases) [50]. In the exemplary case, no primary or any other metastatic site for PTC was depicted on PET scan. The tumor was a teratocarcinoma rather than a metastatic one, similar to the case of Wang et al. [42], who presented carcinoid tumor in a lumbar teratoma associated with tethered cord syndrome in an adult. Presence of PTC within teratoma was formerly reported in an ovarian mass lesion of a 34-year-old woman [51]. They analyzed her thyroid gland and detected some nodules, which turned to be benign thyroid nodule following fine needle aspiration. This is the second case of PTC presenting in a teratoma, and the first case presented within a spinal teratoma. Thyroid gland and full body surveillance should be conducted whenever such or similar case is diagnosed, before acceptance of the lesion is a primary component of teratoma. Endocrine tumors arising within teratomas, such as in the present case, are presumed to originate from neuroendocrine cells of the respiratory or gastrointestinal epithelium that are parts of teratomatous neoplasms [42].

Conclusion

Concomitant presentation of SCM with spinal cord lipoma and teratoma including papillary thyroid carcinoma has not been reported until now, to the best of our knowledge. Such variable presentation of congenital spinal malformations is rare but not absent in the literature. Thoracolumbar region is the most common location for such entity in children, whereas lumbar region for the adults. Surgical resection should be done as much as possible under neuromonitorization. The resected material should be evaluated thoroughly not to miss any malign pathology. Surgical outcome is better when it is done at an early age.

Declarations

Conflict of interest The authors declare that they have no conflict of interest related with the current study.

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