



Case Report

A rare cystic lymphoplasmacyte-rich meningioma: A case report and review of the literature

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ABSTRACT

Background: Meningiomas are common central nervous system neoplasms, accounts for 30% of all primary intracranial neoplasms; the occurrence of meningiomas with cystic lesions is an exceptionally rare. Lymphoplasmacyte-rich meningioma (LPRM) is a rare pathological entity belong to the World Health Organization Grade I meningiomas. LPRM is characterized by abundant lymphoplasmacytic infiltrates which over-shadow the underlying meningeothelial component.

Case Description: A 42-year-old male was admitted to our hospital with a chronic headache for about 3 weeks prior to admission. His symptoms worsen, and subsequently, he experienced left extremities weakness about 1 week before admission. His brain magnetic resonance imaging revealed an irregular and heterogeneously enhancing solid lesion with intratumoral cystic changes at the temporal lobe. A gross total resection was performed; pathological examination revealed a cystic LPRM.

Conclusion: This rare variant of meningioma is a benign tumor entity featured with massive inflammatory cell infiltration and often less proportion of meningeothelial elements. Surgical resection remains the treatment of choice. This is the first report regarding cystic LPRM from Indonesia; we also summarized relevant literature up-to-date, May 2020, reported LPRM cases.

Keywords: Cystic lymphoplasmacyte-rich meningioma, Extremely rare meningioma variant case, Histopathology, Treatment, Up-date literature review

INTRODUCTION

Meningioma is a common neoplasm of the central nervous system, accounting for 30% of all primary intracranial neoplasms.^[5] According to the World Health Organization (WHO) 2016 classification of tumors, there are three grades and 15 subtypes of meningiomas.^[18] The incidence of meningioma with the cystic lesion is rarely encountered. According to many studies on cystic meningioma, this disease has an estimated frequency of 1.6–11.7%.^[8] During 35-years period, Fortuna *et al.* reported a frequency of 1.7%.^[7] Meningioma, with the features of lymphoplasmacyte-rich meningioma (LPRM), is also a very unusual. LPRM, characterized by exuberant lympho-plasmacytic inflammatory cell infiltration, is an exceptionally rare variant of meningioma.^[9] LPRM variant was first reported by Banerjee and Blackwood as a case of subfrontal tumor with features of meningioma and plasmacytoma in 1971.^[1] This tumor occurred

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most commonly over cerebral convexities. Other sites include sphenoid ridges, olfactory groove, parasellar region, petrous ridge, tentorium, and posterior fossa.^[3]

CASE REPORT

A 42-year-old male was admitted to our hospital with chronic headache since 3 weeks before admission. His headache progressively worsened and accompanied with weakness on his left side since 1 week before admission. His weakness was not associated with sensory deficits. He denied any history of trauma, seizures, vomiting, fever, or other associated symptoms. There was a history of alcohol-tobacco consumption >10 years. Neurological examination revealed a grade 4/5 motor strength on both left upper- and lower-limb, other examinations were within normal limits. A complete blood count, erythrocyte sedimentation rate (ESR), coagulation profile, liver-kidney chemistries, and urinalysis were within normal limits.

Brain magnetic resonance imaging (MRI) revealed an irregular and heterogeneously enhancing solid lesion at the right temporoparietal region, with intratumoral cystic changes, a broad attachment on the dura, and perifocal brain edema resulting in midline shift to the contralateral side [Figure 1a and b]. The enhancing portion exhibited hypointense to isointense signal on T1-weighted sequences and hypointense signal on T2-weighted sequences. There was no destruction of bone structure [Figure 1c]. A standard right-sided pterional craniotomy was performed. The dura mater was incised in a cruciate fashion, the tumor was exposed meticulously. The tumor was observed to adhere tightly with the dura mater along the sphenoid wing. They appeared grayish with ill-defined margins. There was also yellowish cystic fluid filling the surgical field. After debulking the tumor using an ultrasonic aspirator, the margins of the tumor and cerebral cortex were carefully dissected using

microsurgical technique. The surgical field was irrigated copiously, and any residual bleeding was cauterized. The temporal fascia was used to replace the dura mater and bone flap was fixed with titanium plates; a gross total resection of Simpson Grade 1 tumor removal was achieved.

The pathological examination revealed the proliferation of neoplastic epithelial cells with eosinophilic cytoplasm within solid nests, with surrounding inflammatory infiltrates, rich in lymphocytes, and plasma cells. The pathological examination also revealed the formation of lymphoid follicles and foci of fibrosis. The tumor tissue was positively stained for vimentin, but negative for glial fibrillary acidic protein. The inflammatory infiltrates were mostly stained with CD3 [Figure 2]. Our intraoperative finding consistent with WHO Grade I; the diagnosis of cystic LPRM was established. The postoperative period was uneventful. The patient muscle strength was gradually improved. His postoperative MRI 6 months after surgery showed no residual nor recurrence of the tumor [Figure 3].

DISCUSSION

LRPM, a rare WHO grade I subtype, usually occurs in young and middle-aged patients without sex predominance; in our case, it is even extremely rare since it occurred with a cystic lesion that has been only ten reported in the literature (became 11 with our case).^[6,10,11,15,16,20,25,28] LPRM can occur in a various locations,^[28] while, in our case, the lesion was located in the right temporal lobe behind the sphenoid wing extending to parietal lobe. Radiologically, these tumors usually show unclear borders with marked edema and also an invasion of adjacent brain tissue. These features suggest a high degree of malignancy, but histological examination revealed that these features were due to extensive inflammatory cell infiltration rather than tumor cell invasion.^[16,28] Lee *et al.* suggested that the perifocal brain edema is probably related

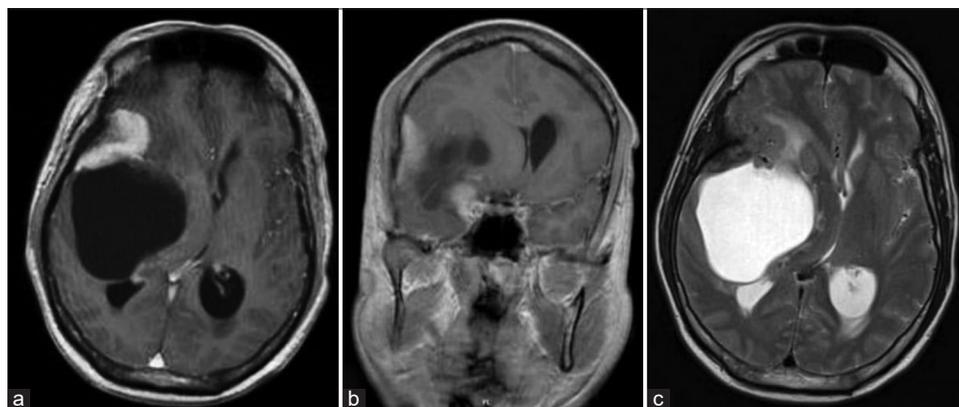


Figure 1: Axial (a) and coronal T1-contrast (b) image shows contrast enhancement dural-based tumor with cystic component size 7 × 5 cm resulting in midline shift of approximately 1.6 cm and third ventricular obliteration. Axial T2 image showing hypointense cystic component (c).

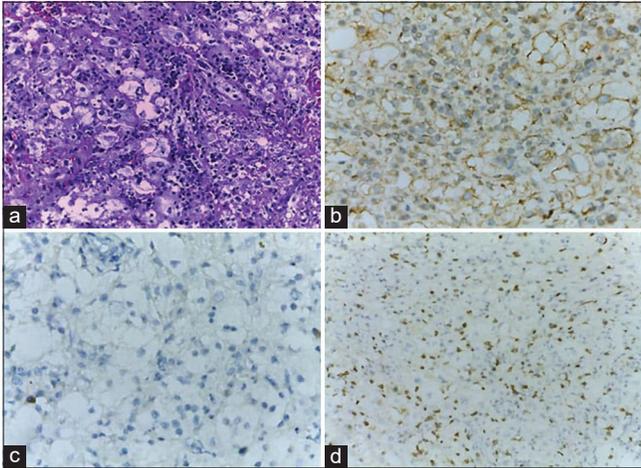


Figure 2: Microscopic examination revealed the proliferation of neoplastic meningotheial cells with pale eosinophilic cytoplasm forming solid nests, associated with a dense chronic inflammatory infiltrate rich in lymphocytes and some plasma cells (a) (H&E, $\times 20$). Both tumor cells and lymphocytes are positive with vimentin (b) ($\times 20$). Negative glial fibrillary acidic protein in tumor cells excludes the diagnosis of glioma with xanthomatous changes (c) ($\times 40$). CD3 staining in lymphocytes dispersed between tumor cells (d) ($\times 20$).

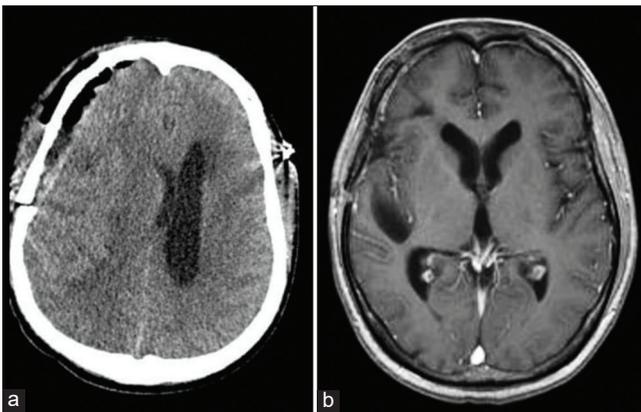


Figure 3: Immediate postoperative computed tomography scan showing total removal of tumor with some certain extent of midline shift (a). Magnetic resonance imaging axial T1-contrast scan 6 months after operation showing no recurrence (b).

to the amount of inflammatory infiltrates within the tumor, blood supply, and pathological type.^[14] Cystic components can also be encountered, as in our case.^[28] The imaging findings of LPRM are different from other types of common meningioma; the differential diagnosis may be extensive, ranging from neoplastic to reactive disorders.

Here, we reported – to the best of our knowledge – the first LPRM case from Indonesia and reviewed relevant up-date literatures summarized LPRM cases (using pathological examination methods); the summary of previously

documented LPRM cases [Table 1, except serial cases been reported previously].^[13,22,27,28] Among 21 patients [included our case, Table 1], the age of diagnosis ranged from 9 to 63 years old (39.2 ± 15.6 years old), and the male-to-female ratio was 10:11.^[3,4,6,9,11,12,15,16,19,20,23-26] The most common locations were convexity (10/21; frontal 2, parietal 2, occipital 2, frontotemporal 1, frontoparietal 2, and temporoparietal 1), cranial base (4/21; anterior 2, middle fossa 2), falx (1/21), and tentorium cerebelli (1/21). Occasionally, the lesions located at ventricle (4/21; lateral ventricle 1, 3rd ventricle 1, and trigone 2) and cerebellopontine angle (1/21). Multiple or diffuse lesions were found in three cases (Case #8, #9 and #17), which were more severe and tended to recur.

From all 21 patients, MRI and computed tomography scan images were available in all patients. Most of the lesions exhibited hypo-to-isointense signal on T1-weighted images and hypo-iso- to high-intense signal on T2-weighted images, usually with homogenous enhancement after gadolinium administration; a classical dural tail sign was observed in some case. Most cases were diagnosed as meningiomas but not LPRM before surgery. In addition, 71.4% (15/21; slightly-edema 1, moderate-edema 3, and severe-edema 11) of the lesions exhibited peritumoral edema with peritumoral or intratumoral cystic changes were observed in 52.4% (11/21) of the patients. Preoperative MRI diagnosis of meningioma was made in seven cases, an aggressive meningioma was in three cases, inflammatory granuloma in two cases, malignant brain tumor in two cases, metastatic tumor in two cases, lymphoma in one case, sinus thrombosis in one case, craniopharyngioma in one case, and glioma in one case. One case had an extensive left tentorial-petroclival extra-axial tumor that was irregular in shape and compressed the brain stem; subtotal resection was then performed followed by gamma knife surgery (GKS) at first to treat the residual tumor.^[23]

There are various of differential diagnoses for these types of cortical mass and biological abnormalities such as idiopathic hypertrophic pachymeningitis, giant lymph node hyperplasia, plasma cell granuloma, multiple myeloma, chordoid meningioma, solitary plasmacytoma, sinus histiocytosis, and lymphomatoid granulomatosis.^[4,17] The epithelial membrane antigen and vimentin-staining are reportedly useful in identifying the meningotheial origin of the tumor and differentiating it from other lesions.^[28] Various hypotheses have been proposed to explain the lymphoplasmacyte infiltration. The question arises as to whether the lesion is indeed primarily neoplastic or granulomatous change with a secondary meningeal reaction. Bruno *et al.* suggest that the LPRM can be considered intracranial inflammatory masses rather than neoplasms due to their biological behavior, immunoprofile, and clinical course.^[2] At present, it is not possible to determine whether the interspersed meningotheial cells are reactive/neoplastic, whether they are primary/secondary to

Table 1: Summary of all previously documented and present lymphoplasmacyte-rich meningioma (LPRM) cases.

No	Reference	Sex/ Age	Location	Edema	Peripheral blood abnormalities	Treatment	MIB-1 (%)	Outcome
1	Cambuzzi <i>et al.</i> , 2012 ^[3]	F/17	Right parietal	-	N/A	Total resection	1	Resolution of neurological symptoms
2	Liu <i>et al.</i> , 2012 ^[6]	M/52	Occipital sagittal sinus	+++	2 cases had been found	Total resection	N/A	In follow-up over 1-4 years, only one case of recurrence in the 1 st year after surgery was noted
3	Out of 7 cases: 4 cases were men, and 3 cases women, with an age range of 9-63 years with average of 38 years	F/39 M/30 ? ? ? ?	Left frontal Ventricle lateral Frontotemporal Cerebral falx Sphenoid ridge Cerebroptontine angle Right sphenoid wing	++ +++ +++ +++ +++ +++ -	to have hematopoietic abnormalities, with polyclonal gammopathy and high blood serum IgA and IgM	Total resection Total resection Total resection Total resection Total resection Total resection Total resection	N/A N/A N/A N/A N/A N/A N/A	One recurrent case above: no recurrence was found in the 2 years following the second surgery No recurrence after 9 month follow-up
9	Majumdar <i>et al.</i> , 2013 ^[19]	M/50	Right sphenoid wing	-	Mild elevated erythrocyte sedimentation rate	Total resection	N/A	No recurrence after 9 month follow-up
10	Wang <i>et al.</i> , 2013 ^[23]	F/60	Left tentorial-clivopetrosal	-	N/A	Subtotal resection	N/A	2 months → residual tumor (tentorial and cavernous sinus). One year → tumor in the cavernous sinus and suprasellar area
11	Wang <i>et al.</i> , 2014 ^[25]	F/37	Intraventricular trigone	+++	N/A	Total resection	N/A	No residual tumor or recurrence 3 months after surgery
12	Lee <i>et al.</i> , 2015 ^[15]	F/35	Left occipital	+++	Normal	Total resection	2	No residual tumor or recurrence 1 year after surgery
13	Cha <i>et al.</i> , 2016 ^[4]	M/55	Left tentorium cerebelli	++	Normal	Subtotal resection	20	Resolution of ataxia symptoms but experienced mild residual visual defects
14	Kaur <i>et al.</i> , 2016 ^[11]	M/21	Right frontoparietal	+++	N/A	Total resection	N/A	Resolution of neurological symptoms
15	Kurmi <i>et al.</i> , 2016 ^[12]	M/32	Right parietal involving scalp	+++	Normal	Total resection	<1	No residual tumor or recurrence 3 months after surgery
16	Kashyap, 2017 ^[9]	F/27	Right frontoparietal	++	Normal	Total resection	N/A	Resolution of neurological symptoms
17	Son, 2018 ^[20]	F/56	The odontoid process and clivus	-	Normal	Subtotal resection and biopsy	15	In follow-up (7 years, 3 years and 3 years) recurrence happen ×3 after surgery, respectively, was noted
18	Wang <i>et al.</i> , 2018 ^[24]	F/51	Left frontal scalp	-	N/A	Total resection	3	No residual tumor or recurrence 1 years after surgery
19	Yang <i>et al.</i> , 2018 ^[26]	F/47	Intracranial duramater and the right trigone area	+	N/A	Subtotal resection	N/A	The patient received radiotherapy and postsurgery MRI at 3 months demonstrated no progression of residual tumor
20	Ferreira <i>et al.</i> , 2020 ^[6]	M/21	The third ventricle	-	N/A	Total resection	0.1	No residual tumor or recurrence 6 months after surgery
21	Present case, 2020	M/42	Right temporoparietal	+++	Normal	Total resection	<1	No residual tumor or recurrence 6 months after surgery

F: Female, M: Male, -: Negative, -: Slightly, ++: Moderate, +++: Severe, N/A: Not available

Table 2: Comparison of clinical features of lymphoplasmacyte-rich meningioma cases until 2020.

Parameter	Zhu <i>et al.</i> , 1971–2012	Yongjun <i>et al.</i> , 2002–2013	Lal <i>et al.</i> , 2014	Tao <i>et al.</i> , 2009–2016	2013–2020*	Accumulative
No. of cases	62	9	16	56	21	164
Patients age ^f	40.7±18.3; 9–79	43±16.9; 99–63	55.3±14.7; 229–78	44.6±12.0; 159–66	39.2±15.6; 99–63	44.6±15.5; 99–79
Male/female ratio	0.97:1	1:0.79	0.45:1	1:1	0.91:1	0.9:1
Severe peritumoral brain edema	44.7% (21/47)	55.6% (5/9)	43.75% (7/16)	40% (22/55)	52.4% (11/21)	40.24% (66/164)
Gross total resection	61.3% (38/62)	77.8% (7/9)	75% (12/16)	80.4% (45/56)	81% (17/21)	72.56% (119/164)
Recurrence	11.3% (7/62)	11% (1/9)	25% (4/16)	8.9% (5/56)	14.3% (3/21)	12.20% (20/164)

*Published paper of LPRM other than serial case report by Zhu *et al.*, 1971–2012,^[28] Yongjun *et al.*, 2002–2013,^[27] Lal *et al.*, 2014,^[13] and Tao *et al.*, 2009–2016.^[22] ^fin years (mean±SD; range)

inflammation due to the various amount of inflammatory and meningotheial component among reposted cases.^[28]

The significance of blood abnormalities found in LPRM remains elusive. Stam *et al.* concluded that the plasma cell infiltrates were not tumoral in origin due to the abundant production of almost all classes of immunoglobulins.^[21] Preoperative laboratory tests disclosed hematopoietic abnormalities in two cases (2/21), with polyclonal gammopathy and high blood serum IgA and IgM [Table 1]. Most of the cases had normal peripheral blood (11/21; including ours) except one with mildly elevated ESR and there were seven cases not available (7/21).

Based on the Simpson grading criteria, 17 of 21 cases had total tumor resection (Simpson Grade I or II), while four cases had subtotal resection or biopsy (Simpson Grade III or IV) since a complete resection was not possible.^[4,20,23,26] After subtotal resection, in one case, treated with GSK after tumor biopsy, the tumor slightly reduced 7 months after GKS, significant tumor shrinkage was noted (postGKS: average 32% reduction by 3 years follow-up) without any adverse radiation effects.^[23] The prognosis of LPRM is favorable, according to previous reports with little recurrence.^[27,28] In this study, recurrence rate is 14.3% (3/21).

To date, 164 LPRM cases, including 11 cystic LPRM from previous case reports and retrospective case series, have been reported. Zhu *et al.*^[28] reviewed all published literature on LPRM from 1971 to 2012 and reported 62 LPRM cases; Yongjun *et al.*^[27] published case series on LPRM from 2002 to 2013 in the Second Hospital of Lanzhou University, Lanzhou, China and reported 9 LPRM cases; Lal *et al.*^[13] published case series on LPRM in 2014 and reported 16 LPRM cases; and Tao *et al.*^[22] published a large case series on LPRM from 2009 to 2016 in Tiantan Hospital, Beijing, China and reported 56 LPRM cases and this study reviewed published literature on LPRM from 2013 to 2020 and reported 21 LPRM cases, as summarized in [Table 2].

LPRM occurs at a higher rate in young and middle-aged patients without sex predominance, which differs from other

types of meningiomas.^[27] Consistent with previous reports, LPRM was commonly located in the convexity.^[13,22,27,28] From published literature, the characteristic of LPRM includes: (i) the mean of age is 44.6 ± 15.5 with a range from 9 to 79 years old, (ii) a slight female predominant, with the male and female ratio is 0.9:1, (iii) the imaging of severe peritumoral brain edema 40.24% (66/164), (iv) gross total resection achieved up to 72.56% (119/164), and (v) the recurrence rate is 12.20% (20/164).

CONCLUSION

Cystic LPRM is an extremely rare benign variant of intracranial meningioma that occurs at a higher rate in young and middle-aged patients without sex predominance with a low tendency of recurrence, mainly in the convexity, featured with a massive inflammatory cells infiltration and often a less proportion of meningotheial components. A definitive diagnosis was possible only through a histopathological examination, along with a good communication between the surgeon and the pathologist. Total surgical resection remains a primary goal of treatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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