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CASE REPORT

Supratentorial neurenteric cyst mimicking hydatid cyst: A case report and literature review

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ABSTRACT

Neurenteric (NE) cysts are uncommon congenital cysts of endodermal origin. These cysts are commonly encountered in the posterior fossa surrounding the brain stem structures. We present a case of pathologically proven supratentorial NE cyst that mimicked a hydatid cyst in its clinical presentation and imaging appearance. Including this pathology in the differential diagnosis of supratentorial cystic lesions is important due to the differences in medical and surgical management.

Key words: Hydatid, neurenteric cyst, supratentorial

INTRODUCTION

Central nervous system (CNS) neurenteric (NE) cysts are uncommon congenital cysts that originate from the endoderm.[1] The majority of NE cysts is encountered in the spine and is reportedly associated with vertebral anomalies in approximately 50%.[1] Conversely, Intracranial NE cysts are rare and typically located in the posterior fossa anterior to the mid brain structures. Supratentorial NE cysts have been rarely reported and tend to be larger in size compared with their posterior fossa counterparts.[1] The present case emphasizes the importance of including NE cysts in the differential diagnosis of more common supratentorial cystic lesions as this may alter the management approach.

CASE REPORT

A 67-year-old Mediterranean male who had episodes of seizures with altered consciousness. He was subsequently placed on keppra and became seizure-free. Magnetic resonance imaging (MRI) of the brain with and without gadolinium revealed a large well-defined left frontal cystic mass measuring 4.8 cm × 4.0 cm × 4.8 cm. The mass displays cerebrospinal fluid (CSF) intensity with low signal on T1-weighted and fluid-attenuated inversion recovery (FLAIR) images [Figure 1] and high signal on T2-weighted (T2W) images [Figure 2]. The lesion contained sausage-shaped debris of high FLAIR signal in its dependent portion with an internal septation. On post contrast imaging, there is no discernible enhancement of the mass or any surrounding capsule. The diffusion tensor imaging (DTI) and diffusion weighted imaging exhibits presence of a high T2 signal wall surrounded by a low signal capsule. On the functional MRI, the left-sided primary motor cortex is seen along the posterior and lateral margin of the left frontal cyst [Figure 3]. The activated supplemental motor area is seen along the medial margin of the cyst. The DTI revealed that

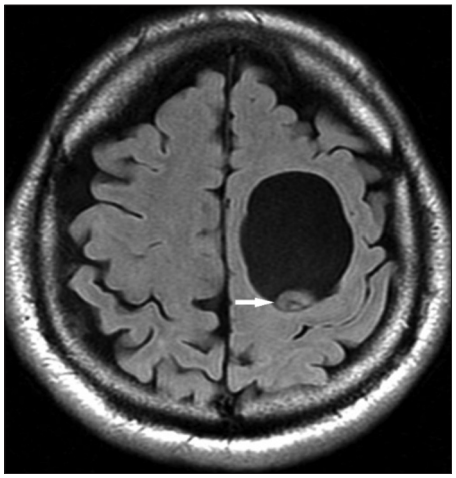

Figure 1: Axial fluid-attenuated inversion recovery image shows a well-defined left frontal low signal cystic lesion with high signal debris in the dependent portion. There is no surrounding vasogenic edema or significant mass effect

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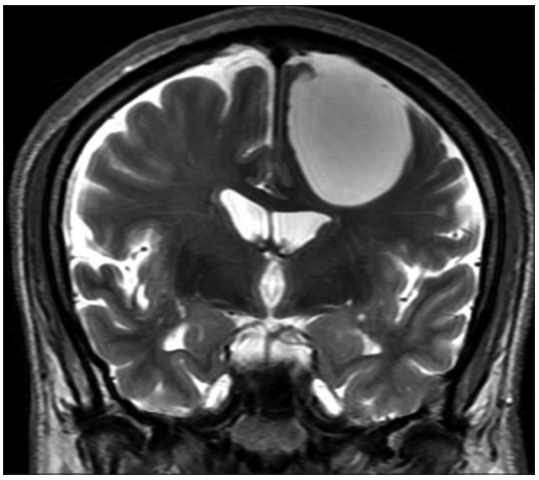

Figure 2: Coronal T2-weighted image shows the hyperintense left frontal cystic lesion significantly displacing the underlying parenchyma

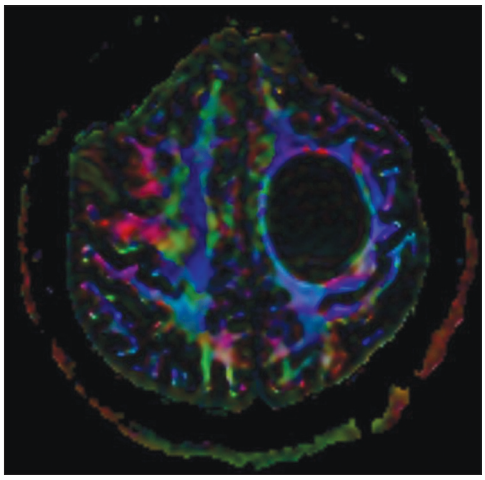

Figure 4: Axial tractography image at the level of the left frontal lesion shows cleavage of the white matter tracts by the lesion rather than infiltration or destruction

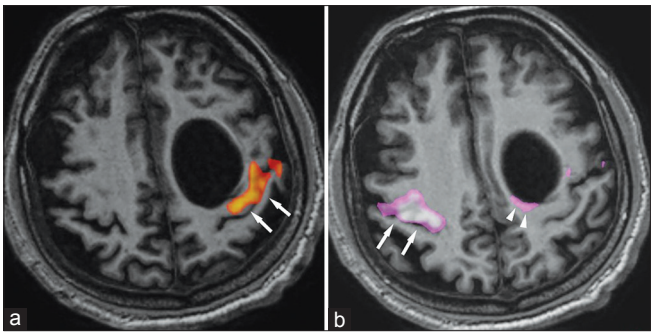

Figure 3: (a) Axial T1-weighted image overlay with a functional magnetic resonance imaging (fMRI) during the right finger tapping task demonstrates activation of the primary motor cortex (arrows) posterolateral to the cystic lesion. (b) fMRI overly image during left finger tapping task shows activation of the right primary motor cortex (arrows) and the left supplemental motor area along the medial margin of the cyst

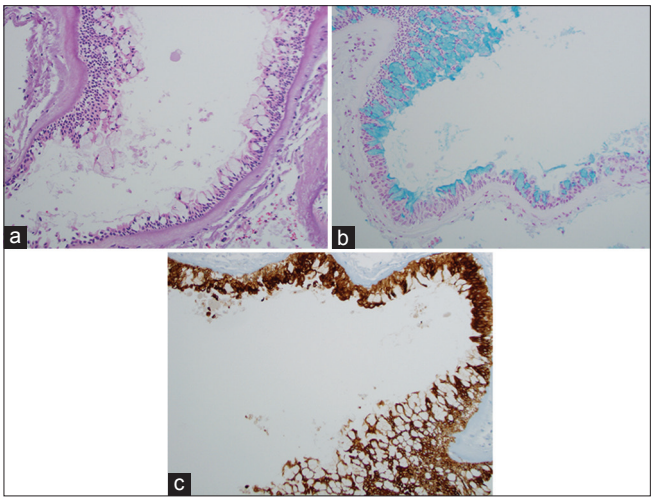

Figure 5: Histopathology of the cyst. (a) H and E stain shows a cystic lesion, with a fibrous wall, lined by pseudostratified columnar epithelium. Note that some cells harbor large intracytoplasmic mucinous contents. (b) Alcian blue staining highlights the mucin content of some cells in blue. (c) Immunohistochemical stain for pan-cytokeratin demonstrates the epithelial origin of the cyst

the white matter tracks of centrum semiovale are cleaved and displaced rather than infiltrated and destroyed [Figure 4]. The patient also underwent a computed tomography (CT) scan of the thorax and abdomen, as part of the patient's workup for suspected hydatid cyst disease, which revealed a mesenteric cystic lesion.

Based on the pre-operative imaging findings, the patient was started on albendazole for treatment of the presumed hydatid cyst.

Later, the patient underwent a stealth-guided left frontoparietal craniotomy for resection of cyst. The lesion was adherent to the cerebral pial surface and contained proteinaceous gelatinous yellowish discolored material. The lesion was irrigated several times with hypertonic saline to kill any hydatid ova. The patient's post-operative course was uncomplicated and albendazole was resumed. Histologically, the lesion showed a fibrous cyst wall lined by cuboidal to columnar, single to multilayered epithelium, which focally shows surface cilia and mucin production that is positive on alcian blue special histochemical stain. The cystic lining is immunoreactive for pan-cytokeratin and epithelial membrane antigen (EMA) and negative for glial fibrillary acidic protein (GFAP) and CK20 [Figure 5]. The histological features are consistent with an enterogenous/epithelial cyst. There is no evidence of hydatid cyst, scolex or other abnormal parasitic structure. Based on the final pathologic diagnosis, albendazole was discontinued.

DISCUSSION

CNS NE cysts are uncommon congenital cysts most commonly encountered in the spinal cord as intradural extramedullary cysts. They are generally, situated ventral to the spinal cord and are most commonly seen in the lower cervical and upper thoracic regions. These cysts

are believed to originate from the endoderm during early embryonic life.[1,2] Several theories have been proposed to explain the pathogenesis of neurenteric cysts. Failure of separation between the notochord and the foregut that leads to incorporation of primitive endodermal cells in the notochord was proposed. However, this mechanism does not explain the existence of supratentorial cysts as the most rostral extent of the endoderm is at the level of the clivus.[3] The "Seessel's pouch origin" hypothesis suggest a common origin for suprasellar neurenteric cysts, Rathke's cleft cysts and colloid cysts. This theory fails to explain the laterally positioned supratentorial cyst. On the other hand, Mittal *et al*. propose that anomalous endodermal cell migration occurs dorsally through the primitive neurenteric canal into the ectoderm and can travel cephalad and to more lateral positions. This progress explains the decreasing incidence of NE cysts from the spine toward the infra and supratentorial compartments, respectively.[2] Given the endodermal origin of NE cysts, elements of bronchial or gastrointestinal epithelium may be seen in these cystic lesions. Furthermore, NE cysts can also contain elements derived from mesoderm and ectoderm.[2] This complex histopathology lead to the use of several names that can be interchangeably used with NE cysts including endodermal, enteric, enterogenous, respiratory, gastroenterogenous and archenteric cysts and have also been labeled as gastrocytomas or intestinomas.[2,4]

In our review of literature, using the keywords, NE, enterogenous, endodermal and supratentorial cysts, we identified around 30 reported cases [Table 1]. The male to female ratio is 1.3:1 with age range between prenatal and 78 years. Clinically, supratentorial NE cysts present with symptoms of space occupying lesion depending on their location such as headache, visual changes, insomnia, seizures, mental and personality changes.

Supratentorial NE cysts are reported to be larger in size than those in the posterior fossa. Supratentorial locations include supra or parasellar,[9,14] septum pellucidum,[25] third ventricle,[7] anterior fossa[4,13,14,16,26] and along the optic nerve.[6] The majority of supratentorial NE cysts occur in extra axial location with no definite laterality predilection. However, there are few reports of intraparenchymal NE cysts in the frontal[3,5] and temporal lobes.[21] All the previously reported and pathologically proven cases of supratentorial NE cysts are solitary. Walls *et al*. reported a case of multiple intracranial enterogenous cysts in which they presumed that the supratentorial intraparenchymal cysts are of similar pathology to the infratentorial cystic lesions.[5] In general, NE cysts are well-defined, smooth and thin-walled, round or lobulated and rarely demonstrate marginal calcifications.

The signal characteristics of NE cysts vary depending on the protein content of the cysts. The majority of NE cysts is proteinaceous and exhibit iso to increased T1-weighted signal compared with the CSF. On T2W sequence, most of the cysts show increased signal and the remaining are hypointense. On the other hand, NE cysts can be hyperintense on FLAIR images and may show mildly impeded diffusion. Although rim of enhancement is rarely seen, Preece *et al*. described this finding in approximately 35% with no definitive correlation between chronic inflammatory changes/cyst rupture and rim enhancement.[1]

Complete surgical excision of supratentorial NE cysts is considered the treatment of choice. This has been achieved in most of the reported cases as the cyst could be easily dissected form the surrounding meninges and brain parenchyma. Incomplete resection or cyst fenestration may result in cyst recurrence[22] or seeding of the cyst contents into the arachnoid space.[27]

Histologically, NE cysts are classified by Wilkins and Odom's into three categories.[28] All types share the presence of single layer of pseudostratified columnar or cuboidal cells mimicking respiratory or gastrointestinal epithelium. Type B exhibits complex invaginations with glandular organization; mucinous or serous production; nerve ganglion, lymphoid, skeletal muscle, smooth muscle, fat, cartilage and/or bone elements. Cysts with ependymal or glial tissue are classified as Type C.[22]

The coexistence of supratentorial NE cyst and intraparenchymal subependymoma has been described.[22] Mucinous low-grade adenocarcinoma was reported to arise from an infratentorial NE cyst.[29] Furthermore, malignant transformation was seen in supratentorial NE cysts into invasive mucinous papillary cystadenocarcinoma[19] and well-differentiated papillary adenocarcinoma.[8]

In contrast to NE cysts, intracranial hydatid cysts are rare acquired cystic lesions secondary to Echinococcus granulosus infection encountered in 2% of cases.[30,31] The most common location of CNS hydatid cysts is in the parenchymal territory of the middle cerebral artery and less commonly in the subarachnoid space.[31] The lesions are usually well-defined, spherical, thin and smooth-walled with no peripheral calcifications or enhancement. The cyst contents follow the appearance of CSF on both MR and CT. MRI, particularly T2W, can be better in depicting the wall due to the increased contrast between the cyst contents and the hypointense wall. Hydatid cysts are rarely multiple secondary to hematogenous spread or when complicated by traumatic or intraoperative rupture.

Table 1: Summary of previous case reports describing supratentorial neurenteric cysts

Author	Year	Age	Gender	Symptoms	Laterality	Location	Location	Contents	Pathology	CT	T1W/T2W	Enhancement	Perilesional edema
Walls *et al.*[5]	1986	40	F	Ataxia	Bilateral	Intraparenchymal	Multiple infratentorial extraaxial and supratentorial intraparenchymal cysts		Enterogenous cyst in the infra tentorial. The supratentorial are presumed to be enterogenous	Hypodense		-	
Scaravilli *et al.*[6]	1992	36	M	Slowly progressive unilateral visual loss	Left	Extraaxial	Posterior intraorbital portion of the optic nerve	Milky	Columnar epithelium partly pseudostratified and ciliated with evidence of mucin secretion				
Büttner *et al.*[7]	1997	28	M	Progressive loss of vision in his right eye	Midline	Intraventricular	3rd ventricle						
Ho *et al.*[8]	1998	45	F	Sensory seizures	Right	Extraaxial	Parietal		(1) A solid, endophytic, well-differentiated papillary neoplasm; and (2) Single-layered, ciliated, simple cuboidal epithelium with a thin underlying fibrous capsule		Hyperintense/ hyperintense	Solid enhancing component with conspicuous papillary configuration	
Sampath *et al.*[9]	1999	27	M	HA, vomiting and left ptosis	Left	Extraaxial	Parasellar						
Cheng *et al.*[10]	2002	49	M	Memory difficulties	Right	Extraaxial	Frontal	Opaque, yellowish-white membrane, mucoid fluid	Ciliated columnar epithelium, necrotic debris		Hypointense/ hyperintense	-	-
Christov *et al.*[11]	2004	31	F	Mild left brachial and crural motor deficit, HA	Right	Extraaxial	Frontoparietal	Turbid liquid	Ciliated cubic cells		Hypointense/ hyperintense	-	-
Tan *et al.*[12]	2004	68	F	Seizures	Left	Extraaxial	Frontal	Opaque, creamy, pale, brown fluid	Ciliated columnar/ pseudostratified epithelium	Hypodense with calcifications		-	
Kachur *et al.*[3]	2004	35	F	Seizure-like episodes, right frontal HA	Right	Intraparenchymal	Frontal	Clear yellowish	Cuboidal epithelium		Hypointense	-	-

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Table 1: Contd...

Author	Year	Age	Gender	Symptoms	Laterality	Location	Location	Contents	Pathology	CT	T1W/T2W	Enhancement	Perilesional edema
Stubenvoll *et al.*[13]	2006	25	M	Seizure	Right	Extraaxial	Frontal	Comparable to CSF with slightly increased protein level	Single layer of ciliated epithelium		Hypointense		+
Preece *et al.*[1]	2006	72	F	Behavior changes leading to psychosis	Left	Extraaxial	Frontal		Low pseudostratified, ciliated, columnar to cuboidal	Hypodense	Hyperintense/ hyperintense	-	
		34	M	Seizures	Left	Extraaxial	Frontal		Simple columnar to cuboidal; mucin-producing cell rich	Hypodense	Isointense/ hyperintense	-	
		78	F	Seizures	Right	Extraaxial	Frontal	Fluid-fluid level	Pseudostratified ciliated columnar; mucin-producing cell poor	Isodense	ISO-hyper/ hyperintense	-	
		48	M	Tremor, numbness of right arm, facial droop	Left	Intraparenchymal	Intra/ paraventricular		Stratified columnar; mucin-producing cell poor	Hyperdense with calcifications	Hyperintense/ hyperintense	-	
		78	M	Seizures	Unknown	Extraaxial	Frontal		Ciliated columnar	Hypodense	Hyperintense/ hyperintense	-	
Neckrysh *et al.*[14]	2006	72	F	Progressive personality changes, gradual loss of smell	Midline	Extraaxial	Anterior cranial fossa	Yellowish semiliquid	Pseudostratified cubical and columnar epithelium		Hyperintense/ hyperintense	-	
Miyagi *et al.*[15]	2007	63	M	HA, dizziness	Right	Extraaxial	Parietal	Milky-white fluid	Single layer of ciliated columnar epithelial cells and pseudostratified columnar epithelium. Mucous-secreting goblet cells	Hypodense	Hypointense/ hyperintense	-	-
Takumi *et al.*[16]	2008	32	M	Status epilepticus	Left	Extraaxial	Frontal	Yellowish-opaque gelatinous liquid	Column-like and cubic cells		Hypointense/ hyperintense	-	
Marchionni *et al.*[17]	2008	20	F	HA, left hemiparesis, left deafness, unsteadiness, blurred vision	Left	Extraaxial	Temporal peri-insular and cerebellopontine region	Turbid and thick fluid/ lump of inspissated proteinaceous	Entergenous cyst		Hyperintense		
Basheer *et al.*[18]	2009	54	M	HA	Right	Extraaxial	Parietooccipital with extension into the posterior fossa	Yellowish oily fluid	Cyst lined by a ciliated stratified squamous epithelium	ISO to hypodense	Very hyperintense	-	

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Table 1: Contd...

Author	Year	Age	Gender	Symptoms	Laterality	Location	Location	Contents	Pathology	CT	T1W/T2W	Enhancement	Perilesional edema
Dunham *et al.*[19]	2009	58	F	HA, memory difficulty	Right	Intraparenchymal	Right parietal	Clear fluid	Benign respiratory/ GI like epithelium with focal areas of malignant features, pseudostartified cuboidal to columnar cells with obvious goblet cells	Peripheral calcification	ISO to CSF/ hyperintense	+	+
Reddy *et al.*[20]	2010	20	M	Suboccipital HA and painless, progressive vision loss	Left	Extraaxial	Posterior fossa with extension into the temporal region	Sterile, thick, yellowish fluid	Ciliated tall columnar epithelium, focal squamous metaplasia	Slightly hyperdense compared with CSF	Hyperintense/ isointense	Peripheral	-
Mittal *et al.*[2]	2010	76	F	Seizure	Right	Extraaxial	Frontoparietal	Opaque, viscous, greenish material corresponding to the hyperintense signal on MRI	Columnar epithelium with pseudostratification	Hypodense	Hypointense with focal hyperintensity/ hyperintense with focal hyperintensity	-	-
Jhawar *et al.*[21]	2011	41	M	Bitemporal HA, siezures	Right	Intraparenchymal	Temporal	Oily fluid with shimmering crystals of cholesterol/ white, cheesy solid deposits	Cystic lesion lined by pseudostratified columnar ciliated epithelium	Hyperdense with calcification	Hyperintense/ hypointense	-	
Little *et al.*[4]	2011	70	M	Gait disturbance	Midline	Extraaxial	Sella	Gelatinous fluid, single red solid nodule	Single layer of squamous epithelium and ciliated columnar epithelium lacking goblet cells	Hyperdense	Slightly hyperintense/ hyperintense	-	-
Natrella *et al.*[22]	2012	45	M	Seizure	Left	Extraaxial	Frontal		Type C cyst with respiratory-like pluristatified ciliated epithelium and Grade I WHO subependymoma		Hypointense with hyperintense solid component/ hyperintense with hypointense solid component	Non-enhancing cyst/sllight enhancement of the solid component	

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Table 1: Contd...

Author	Year	Age	Gender	Symptoms	Laterality	Location	Location	Contents	Pathology	CT	T1W/T2W	Enhancement	Perilesional edema
Krishna murthy *et al*.[23]	2010	32	M	Progressive left hemiparesis	Right	Extraaxial	Frontoparietal	Yellowish white milky fluid				-	
		44	F	Raised ICP, no deficits	Left	Extraaxial	Frontoparietal	Yellowish white milky fluid					
Pulido-Rivas *et al*.[24]	2012	Neonate	-	Increase head circumference	Unknown	Extraaxial	Parietooccipital	Thick mucous membranes					
		Neonate	-	Increase head circumference	Unknown	Extraaxial	Parieto-occipital	Thick mucous membranes			Hypointense/ hyperintense Hypointense/ hyperintense	-	

HA: Headache, MRI: Magnetic resonance imaging, CSF: Cerebrospinal fluid, ISO: International organization for standardization, GI: Gastrointestina, WHO: World health organization, ICP: Intracranial pressure

Although some intracranial cystic lesions may have distinct imaging features, the differentiation between NE cysts, hydatid cysts and other benign and malignant cystic lesions can be difficult on imaging due to the similarity in imaging features. In our case, the presence of debris within the cyst, the location and appearance of the lesion as well as the presence of mesenteric cyst initially suggested the diagnosis of hydatid cyst, which was the presumed pre-operative diagnosis. This case illustrates the similarity of imaging features between hydatid and supratentorial NE cysts that require a different management approach.

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