

A CASE SERIES OF UNUSUAL PRESENTATION OF NEUROCYSTICERCOSIS IN CHILDREN

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Abstract

Background: Neurocysticercosis (NCC) is a common parasitic infection of central nervous system with diverse clinical and radiological presentations. Just like its presentation, there is no uniformity in management guidelines for NCC. This case series aims to discuss certain unusual and rare neuroimaging presentations of NCC, along with a detailed review of their sociodemographic, clinical, diagnostic and management characteristics. **Materials and Methods:** Ten children diagnosed with NCC with variable clinical and neuroimaging profile were selected for the study. Socio-demographic and clinico-radiological characteristics of all selected cases is described in detail. **Result:** Focal seizures (6 cases) was the most common presenting symptom followed by generalized seizures (3 cases). On neuroimaging, out of total 10 cases, 7 were reported as multiple ring enhancing lesions (MREL), 2 were single conglomerate ring enhancing lesions (SCREL) and one was single discrete ring enhancing lesion (SDREL). Six cases were found to be in colloid-vesicular and three in granulomatous stage. **Conclusion:** Focal seizures followed by generalized seizures are most common presenting symptoms observed in children with NCC. MRI (magnetic resonance imaging) is very valuable in diagnosis of NCC at variable location and stages in the brain.

INTRODUCTION

Neurocysticercosis (NCC) is a common parasitic infection of central nervous system caused by larval stage of *Taenia Solium*. It has a wide spectrum of clinical and radiological findings. Neuroimaging plays a pivotal role in its diagnosis and management. With advancement in neuroimaging techniques there has been a tremendous enhancement in the knowledge about topography, evolution through stages and host inflammatory reaction against cysticercosis. This has aided in better management, gauging response to therapy and long/short-term outcomes of the NCC.^[1]

In India the most encountered form of NCC is solitary cysticercal granuloma (SCG), visualized as single ring enhancing lesion (SREL) on neuroimaging. It may be single discrete Ring enhancing lesion (SDREL) or single conglomerated ring enhancing lesion (SCREL). NCC may also appear as multiple ring enhancing lesions (MREL).^[2-4]

This case series presents unusual aspect of NCC in rural India.

Objective

To describe the socio-demographic, clinical and radiological profile of 10 uncommon presentations of children aged between 5 to 13 years diagnosed with NCC at BPS GMC (W) Khanpur, Kalan from June 2022 to January 2023.

CASE REPORTS

All the cases were between 5 years to 13 years with 6 males and 4 females. All cases used toilets for defecation whereas 2 used open defecation. 60% (n=6) of the cases had pigs in and around their household. No slaughter house was reported in the vicinity of their residence. Source of water was hand pump for 80%(n=8) and only 20%(n=2) were using municipal water supply. Eating habits showed nearly 50% (n=5) had consumed non-vegetarian food in the past. The sociodemographic details of the 10 cases are given in [Table1].

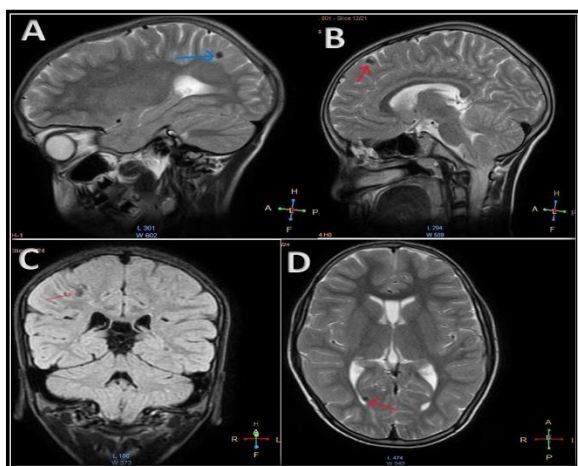


Figure 1: (case 1) T2 (A, B, D) and FLAIR (C) showing low signal intensity suggestive of calcified stage of NCC. T2 (D) showing calcified NCC in posterior horn of right lateral ventricle

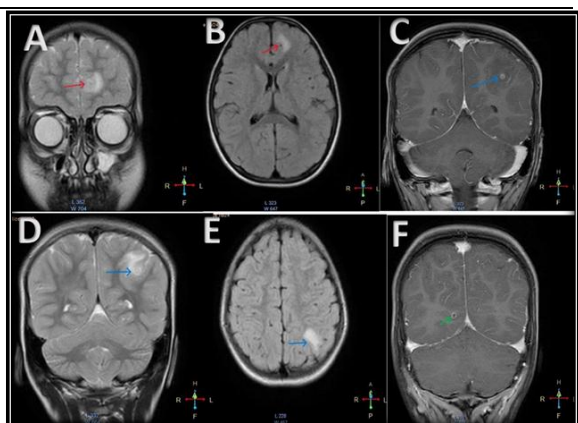


Figure 2: (case 2) T2(A, D), FLAIR (B, E) and CISS (Constructive interference in steady state) sequence (C, F) showing multiple cysts in colloid-vesicular stage with perilesional oedema.

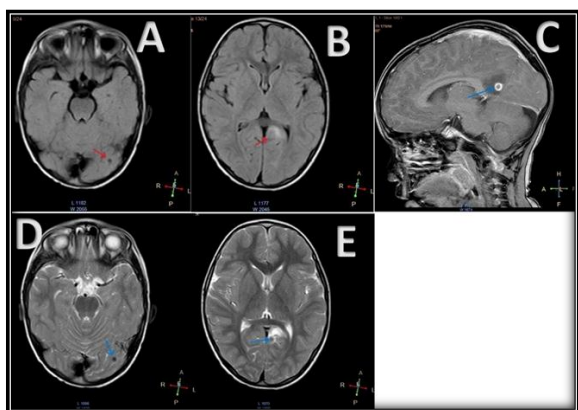


Figure 3: (case 3) Hypointense nodule in calcified nodular stage NCC seen in left occipital lobe on FLAIR (A) and T2 (D). Granular nodular stage NCC seen as hypointense lesion on T2(E), FLAIR(B) and CISS sequence (C) with some perilesional oedema.

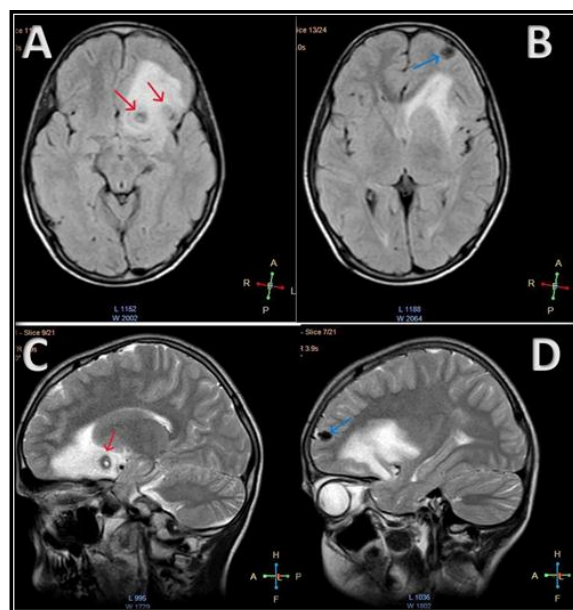


Figure 4: (case 4) Multiple colloid-vesicular stage of NCC seen on T2 (C) and FLAIR (A) with perilesional oedema. FLAIR (B) and T2 weighted image (D) showing calcified nodular stage of NCC.

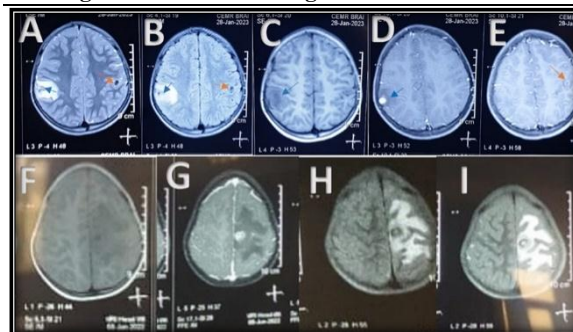


Figure 5: (case 5) [IMAGES FROM 2023] T2 (A), T1 (C) and FLAIR (B) showing low signal intensity suggestive of granular-nodular (blue arrow) and calcified (red arrow) stage of NCC with some perilesional oedema. Contrast enhancement (D, E) shows ring enhancing lesion (red arrow) and solid enhancing nodule (blue arrow) of NCC. [IMAGES FROM 2021] T1(F),T2(I),FLAIR (H) and post Contrast image (G) showing single conglomerated ring enhancing lesion of NCC in colloid-vesicular stage with extensive perilesional oedema.

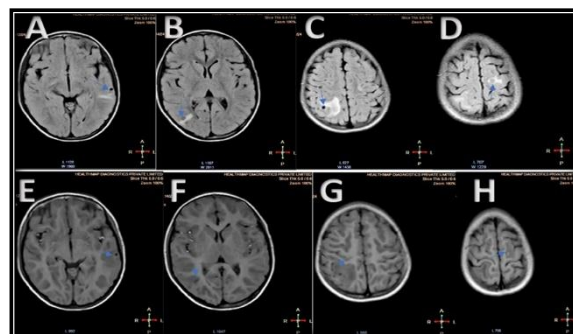


Figure 6: (case 6) Multiple NCC (blue arrow) seen in granular-nodular stage on FLAIR (A, B, C, D) and T1 (E, F, G, H) images with some perilesional oedema.

Single conglomerate ring enhancing lesions (SC-REL) were reported in case 2, 4,8.

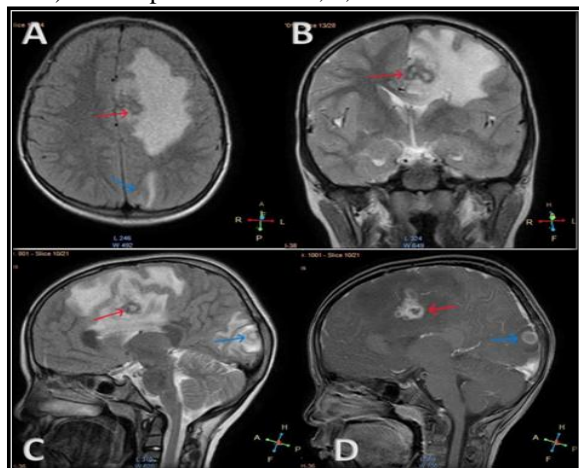


Figure 7: (case 7) FLAIR (A), T2 (B, C) AND CISS (Constructive interference in steady state) sequence (D) showing conglomerated lesion (red arrow) in colloid-vesicular stage cyst with extensive perilesional oedema with midline shift. A solitary cyst in colloid-vesicular stage is also seen in left occipital lobe (blue arrow).



Figure 8: (case 8) T2 weighted image showing conglomerated NCC in colloid-vesicular stage with perilesional oedema.

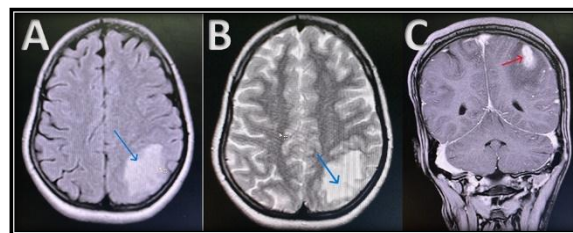


Figure 9: (case 9) FLAIR (A), T2 (B) AND CISS sequence (c) showing conglomerated NCC in colloid-vesicular stage with perilesional oedema.

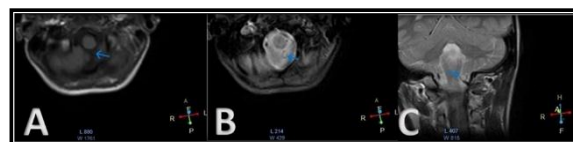


Figure 10: (case 10) Isolated single cyst in subarachnoid space in vesicular stage with minimal or no perilesional oedema on T1 (A), T2 gradient echo sequence (B). On T2 (C) cyst is hyper intense and scolex is isointense to parenchyma.

Table 1: Socio-demographic variables of cases.

Socio-demographic variables.	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8	Case 9	Case 10
Age at diagnosis	9yrs	7yrs	5yrs	8yrs	10yrs	6yrs	6yrs	8yrs	13yrs	6 yrs.
Gender	male	female	male	female	female	male	male	male	female	male
Religion	Hindu	Hindu	Hindu	Hindu	Hindu	Hindu	Hindu	Hindu	Hindu	Hindu
Level of education of care taker	primary school	illiterate	primary school	primary school	illiterate	illiterate	primary school	primary school	primary school	primary school
Source of water	hand pump	municipal supply	hand pump	hand pump	municipal supply	hand pump	hand pump	hand pump	hand pump	hand pump
Method of water storage	pots	pots	tank	pots	pots	water tank	tank	open bucket	pots	Pots
Method of water purification	none	none	none	none	none	none	none	none	none	none
Type of home	mud house	brick house	brick house	brick house	brick house	brick house	brick house	brick house	brick house	brick house
Type of latrine	open defecation	toilets	open defecation	toilets	toilets	toilets	toilets	toilets	toilets	toilets

Method of garbage disposal	none	disposed by sweeper	none	disposed by sweeper	disposed by sweeper	disposed by sweeper	disposed by sweeper	disposed by sweeper	disposed by sweeper	disposed by sweeper
Distance of garbage dumping from water source	>100 feet	>100 feet	>100 feet	>100 feet	>100 feet	>100 feet	>100 feet	>100 feet	>100 feet	>100 feet
Pigs in & around household	yes	yes	yes	yes	yes	no	none	no	no	Yes
Presence of slaughter house in vicinity	no	no	no	no	no	no	no	no	no	no
Peels fruits before eating.	no	no	no	no	no	no	no	no	no	No
Frequency of patient eating outside home.	once a month	once a month	once a month	more than once a week	once a month	once a month	once a month	once a month	occasionally	more than once a week
Washing salad before eating	yes	yes	yes	yes	yes	yes	yes	yes	yes	Yes
Vegetarian or non-vegetarian?	vegetarian	non vegetarian	non vegetarian	vegetarian	non vegetarian	vegetarian	non vegetarian	non vegetarian	vegetarian	Vegetarian
Which type of non veg food do you consume most often?	none	chicken	chicken	none	chicken	none	egg	chicken	none	none
Nonvegetarian food cooked at home or outside?	none	home	outside/home	none	home	none	home	home	none	None

In this case series, focal seizure was the most common (n=6; 60%) first symptom followed by generalised seizures (n=3,30%). Headache was reported to be the other most common associated symptom. Phenytoin was the most commonly (N=7; 70%) administered antiepileptic amongst these 10 patients. Half the number of cases took antiepileptic drugs for 24 months and two cases received for 12 months. Two cases (20%) on polytherapy received antiepileptics for 36 months. 90% (n=9) cases received dexamethasone and only one case received prednisolone. Duration of steroid use was variable ranging from 5 days to 14 days. Traditional practices/therapy was used by 2 (20%) patients and 1 (10%) patient was lost to follow-up. Clinical variables of the cases have been described in [Table 2].

Table 2: Clinical variables/profile of cases

Clinical variables	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8	Case 9	Case 10
First symptom	Generalized seizures	Focal seizures	Focal seizures	Focal seizures	Focal seizures	Generalized seizures	Focal seizures	Generalized seizures	Focal seizures	Headache
Other symptoms	None	Headache, loss of consciousness	None	Vomiting, headache	Headache, vomiting	Headache	Headache	Loss of consciousness	Headache	None
Visual disturbances and other neurological deficits	None	None	None	None	None	None	None	None	None	None
Recurrent NCC/reinfestation	No	No	No	No	Yes	No	No	No	No	No
First antiepileptic used	Phenytoin	Carbamazepine	Carbamazepine	Phenytoin	Levetiracetam	Phenytoin	Phenytoin	Phenytoin	Phenytoin	Phenytoin
Breakthrough seizure while on treatment with first antiepileptic drug.	Yes	No	Yes	No	No	No	No	No	No	No
Polytherapy if used then which AED used	Levetiracetam	.	Levetiracetam
Total duration of treatment with AED	36 months	24 months	36 months	24 months	Ongoing therapy; scheduled for follow up at 6 months	12 months	24 months	24 months	24 months	12 months

AED side effects	None	None	None	None	None	None	None	None	None	None
Antiparasitic drug used	Albendazole at standard dose of 15mg/kg/day	Albendazole at standard dose of 15mg/kg/day	Albendazole at standard dose of 15mg/kg/day	Albendazole at standard dose of 15mg/kg/day	Albendazole at standard dose of 15mg/kg/day + praziquantel (50mg/kg)	Albendazole at standard dose of 15mg/kg/day + praziquantel (50mg/kg)	Albendazole at standard dose of 15mg/kg/day + praziquantel (50mg/kg)	Albendazole at standard dose of 15mg/kg/day	Albendazole at standard dose of 15mg/kg/day	Albendazole at standard dose of 15mg/kg/day
Duration of antiparasitic treatment.	28days	28days	28days	28days	28days	28days	28days	28days	28days	28days
Steroid used	None	Dexamethasone	Dexamethasone	Prednisolone	Dexamethasone	Dexamethasone	Dexamethasone	Prednisolone	Dexamethasone	Prednisolone
Duration of steroid use.	None	5days	5days	14days	5days	5days	5days	14days	5days	14days
Compliance to treatment and follow up visits	Good	Good	Poor	Poor	Poor	Good	Good	Good	Good	Poor & lost to follow-up
Use of traditional medicine for treatment	Yes	No	Yes	No	No	No	No	No	No	No

Most (n=6) of cases were MREL and 3 were SCREL. At time of diagnosis six cases were in colloid-vesicular stage, three in granulo-nodular stage and one was calcified-nodular stage. Perilesional oedema was reported in 9 (90%) cases except in case 1. None of the cases reported hydrocephalous, leptomenigeal enhancement or midline shift. No evidence of cysticercosis was reported outside CNS (central nervous system). Two cases showed rather unusual location, case 1 showing intraventricular lesion and case 10 showing subarachnoid space lesion. Case 1 on MRI (magnetic resonance imaging) showed MREL of NCC at parieto-occipital intraventricular region of brain in calcified nodular stage with no perilesional oedema. Lesion showed resolution at 6 months on follow up MRI. Case 10 on neuroimaging reported SDREL in subarachnoid space in colloid-vesicular stage with perilesional oedema. Detailed investigative and radiological profile of cases is shown in [Table3].

Table 3: Radiological and investigative profile of cases

Radiological and investigational profile	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8	Case 9	Case 10
Number of lesions	Multiple	Multiple	Multiple	Multiple	Multiple	Multiple	Multiple	Multiple	Multiple	Single
Location of lesions	Parieto-occipital, intraventricular (posterior horn of lateral ventricle)	Frontal/parieto-occipital/temporal	Parieto-occipital.	Frontal/temporal	Frontal/parieto-occipital/temporal	Parietal	Parietal/occipital	Parietal	Parieto-occipital	Subarachnoid spaces
Type of lesion (SDREL/SCREL/MREL)	MREL	MREL	MREL	MREL	MREL	MREL	SCREL/SDREL	SCREL	SCREL	SDREL
Stage of lesion at diagnosis	Calcified nodular	Colloid vesicular	Calcified/granular-nodular	Calcified/colloid vesicular	Granular-nodular/calcified	Granular nodular	Colloid vesicular	Colloid vesicular	Colloid vesicular	Colloid vesicular
Perilesional oedema.	None	Present	Present	Present	Present	Present	Present	Present	Present	Present
Hydrocephalus	No	No	No	No	No	No	No	No	No	No
Midline shift	No	No	No	No	No	No	Yes	No	No	No
Leptomeningeal enhancement	No	No	No	No	No	No	No	No	No	No
Follow-up neuroimaging done after how many months of diagnosis	6 months	12 months	6 months	Not done	Planned at 6 month of therapy	6 months	6 months	6 months	12 months	Not done
Finding on follow up neuroimaging. (Resolution with or without calcification; non-resolution)	Resolution with calcification	Resolution without calcification	Resolution with calcification	Unknown	.	Resolution with calcification	Resolution without calcification	Resolution without calcification	Resolution without calcification	Unknown

with active lesion persisting)											
Persistence of lesion with surrounding oedema after completion of cysticidal treatment & requiring a second course of the same.	No	No	No	Unknown		No	No	No	No	No	Unknown
Intraretinal cysticercosis on orbital sonogram or fundoscopy	No	No	No	No	No	No	No	No	No	No	No
Evidence of cysticercosis outside CNS	No	No	No	No	No	No	No	No	No	No	No
Evidence of taeniasis on stool examination	No	No	No	No	No	No	No	No	No	No	No
Other test (MTX, CXR, HIV ELISA, MRS)	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done	MTX-non-reactive/ CXR-normal/ HIV ELISA-negative/ MRS - not done

(Mantoux=MTX, chest x ray=CXR, magnetic resonance spectroscopy=MRS)

Most (n=6) of cases were MREL and 3 were SCREL. At time of diagnosis six cases were in colloid-vesicular stage, three in granulo-nodular stage and one was calcified-nodular stage. Perilesional oedema was reported in 9 (90%) cases except in case 1. None of the cases reported hydrocephalous, leptomenigeal enhancement or midline shift. No evidence of cysticercosis was reported outside CNS (central nervous system). Two cases showed rather unusual location, case 1 showing intraventricular lesion and case 10 showing subarachnoid space lesion. Case 1 on MRI (magnetic resonance imaging) showed MREL of NCC at parieto-occipital intraventricular region of brain in calcified nodular stage with no perilesional oedema. Lesion showed resolution at 6 months on follow up MRI. Case 10 on neuroimaging reported SDREL in subarachnoid space in colloid-vesicular stage with perilesional oedema.

DISCUSSION

The 10 cases discussed here highlight several important findings about NCC in rural India, like its topographic distribution in brain especially unusual ones like in intraventricular and subarachnoid spaces. It also brings forth still prevalent traditional practices (jhara and tantra mantra) in rural north-west India. Recurrent infection is another important aspect of NCC that might not be as rare a phenomenon especially in endemic areas like north-west India as was thought to be previously.^[1] Cysts in the ventricular system is a serious manifestation of NCC

that has distinct features and management owing to obstruction of the ventricles by the cyst. According to a study by Nash et al ventricular involvement was mostly accompanied by multiple cysts at other locations in 74% cases. Headache was the most common symptom in these patients owing to hydrocephalus documented in 73.9% of the cases. However, in case of lateral ventricle cyst most of the patients did not have hydrocephalus and were asymptomatic with the cyst showing resolution irrespective of treatment.^[5] The cysts of NCC in the ventricular system, as reported by Jensen et al were most commonly found in the 4th ventricle (43% to 70%) followed by lateral ventricle (11% to 43%) and third ventricle (1% to 29%).^[6]

Case 1 in this series presented with generalized seizure. On MRI multiple calcified cysticercosis including an adherent cysticercosis in posterior horn of lateral ventricle with no hydrocephalus was seen. The adherent lesions are seen in intraventricular cysts due to local antigenic reaction causing ventriculitis and later cyst adhering to the ventricular wall.^[7] This patient was managed with anthelmintic and antiepileptic with no neurosurgical intervention done in the absence of hydrocephalus. Anthelmintic therapy was prolonged due to recurrent seizures.

In the largest study involving 500 children with NCC by Singhi P et al, it was reported that multiple lesions at presentation was seen in 20.4%. Children with multiple lesions also experienced more recurrent seizure (28.4% vs. 14.5%), more resolution with calcification (11.7% vs. 3.6%) on long term follow up, a lower resolution rate on 6 monthly follow up

(31.2% vs. 52.7%) when compared to solitary lesion.^[8] In this case series all cases had multiple lesions except case 10. Recurrent seizure was seen in 2 (case 1 and 5). Resolution with calcification was also reported in 3 cases while 4 cases had resolution without calcification.

Conglomerated ring enhancing lesion is a radiological feature commonly seen on MRI as 2 or more ring lesion grouped together. It is mostly missed or seen as SREL on CT owing to more thickness of slices on CT imaging. Garg et al using thin slice (2mm) on CT reported 58 SC-REL out of total 100 cases (58%). However lower percentage (28.8%) has been reported using MRI. A lower complete resolution rate was also seen for SCREL (13.8% VS 70%) when compared to SD-REL.^[9,10] SCREL were seen in cases 7,8,9. Of these all had complete resolution without calcification.

Recurrent NCC is defined as appearance of a new lesion after complete resolution of previous lesion or a change in morphology of the existing lesion. There have been isolated case reports of recurrent NCC. In a retrospective study of 278 patients, Kaur et al reported 15 (12.6%) recurrent NCC. 10 of these had documented complete resolution and then appearance of new lesions at different location.^[1] Case 5 in this series had recurrence of lesion after 2yrs. Case 5 had complete resolution of lesion without calcification at 6 month follow up after completing treatment (during first infestation of NCC in 2020) and remained seizure free for next 2 yrs. After 2 years of being symptom free, at an age of 10 years she had re-infestation (2023) and presented with focal seizure, headache & vomiting. On neuroimaging new lesions were seen at different locations shown in [Figure 5; A, B, C, D, E] with resolution of lesions from the first episode [Figure 5; F,G,H,I]. She is currently on levetiracetam monotherapy with no repeat seizures scheduled for a follow up at 6 months of treatment.

Subarachnoid NCC (SANCC) unlike parenchymal has certain peculiar characteristics. It is exceedingly rare in paediatric age group in Indian subcontinent and most cases are reported from South America.^[11] SANCC has a longer incubation period usually ranging from 1 to 2 decades. Symptoms like hydrocephalus, lacunar infarcts, nerve entrapments, long tract signs and symptoms are attributed to arachnoiditis accompanying SANCC. This warrants intensive long-term immunosuppression along with anthelmintic therapy. SANCC goes through stages of asymptomatic cystic proliferation, classic racemose (inactive) stage, stage of arachnoiditis and resolution (calcification and scarring).^[12] Case 10 in this series showed an isolated subarachnoid cyst. The patient was however asymptomatic and was lost to follow-up.

This case series highlights the diversity of presentation of neurocysticercosis and the need to formulate uniform guidelines for diagnosis and

management of neurocysticercosis in paediatric population.

CONCLUSION

NCC continues to be major burden on community in India. Focal and generalised seizures are the most common presenting complaints in children with NCC. Though solitary ring enhancing lesion is most common finding on neuroimaging in case of NCC but MREL and SCREL are not uncommon. MRI is a very valuable tool in diagnosing NCC at unusual locations and various stages of development. Apart from antiepileptic drugs, anthelmintic therapy (albendazole/praziquantal) with shorter duration of corticosteroids are quite effective against NCC in children.

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