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Case Report

Juvenile xanthogranuloma of central nervous system: Imaging of two cases report and literature review

Wangqiang Chen [a], Yue Cheng [b], Shengfa Zhou [a], Yu Chen [a], Xuming Chen [a], Shuang Xia [b,*]

[a] *Radiological Department, Second Affiliated Hospital of Wenzhou Medical University, Xueyuan West Road 109#, Lucheng District, Wenzhou, Zhejiang, China*
[b] *Radiological Department, Tianjin First Central Hospital, Fukang Road 24#, Nankai District, Tianjin, China*

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Abstract

The current paper reported imaging characterization in two cases with juvenile xanthogranuloma of central nervous system. The imaging appearances were detail discussed combined with the 25 cases from literature.

Keywords: Juvenile xanthogranuloma; Central nervous system; MRI

1. Introduction

Juvenile xanthogranuloma (JXG) is a kind of benign lesion and pathology is mainly with non-Langerhans cell histiocytosis infiltration. It is common seen in children younger than 10 years old [1]. The most common manifestation are dermal, with a predilection for the head and neck region [2]. Cutaneous JXG is usually benign and self-limiting. Systemic involvement occurs in about 4% of patients [3], including viscera, bone, and central nervous system (CNS). Intracranial JXG is uncommon, and often secondary to the systematic disease [1]. Primary isolated cerebral lesions are rare. Majority of intracranial JXG from literature are case reports, especially imaging appearances had not been systematical reviewed. Here we present MRI imaging of two cases of JXG with CNS involvement and review the MRI characterization of the JXG in the central nervous system.

2. Case reports

2.1. Patient 1

A 6-year-old boy who complaint of headache and vomit for 1 week visit hospital and head MRI was suggested. One and half year ago a lesion of left renal pelvis was found occasionally and biopsy revealed xanthogranuloma. Head MRI revealed multiple striped or specked space-occupying lesions in the choroid plexus of bilateral ventricle, cerebral falx and bilateral tentorium of cerebellum. The lesions showed isointensity on T1 weighted images (T1WI) and hypointensity on T2 weighted images (T2WI), with mild restricted diffusion. Obvious supratentorial hydrocephalous and periventricular interstitial edema were seen secondary to compression of the interventricular foramen (Fig. 1). The patient received lateral ventricle drainage to alleviate the symptoms. Head CT two years later demonstrated some calcification in the lesions.

2.2. Patient 2

A 7-year-old girl who had a history of hepatolenticular degeneration complaint of headache for 1 year and visited hospital. Brain MRI showed a solitary, well-demarcated mass in

* Corresponding author.
E-mail address: xiashuang77@163.com (S. Xia).
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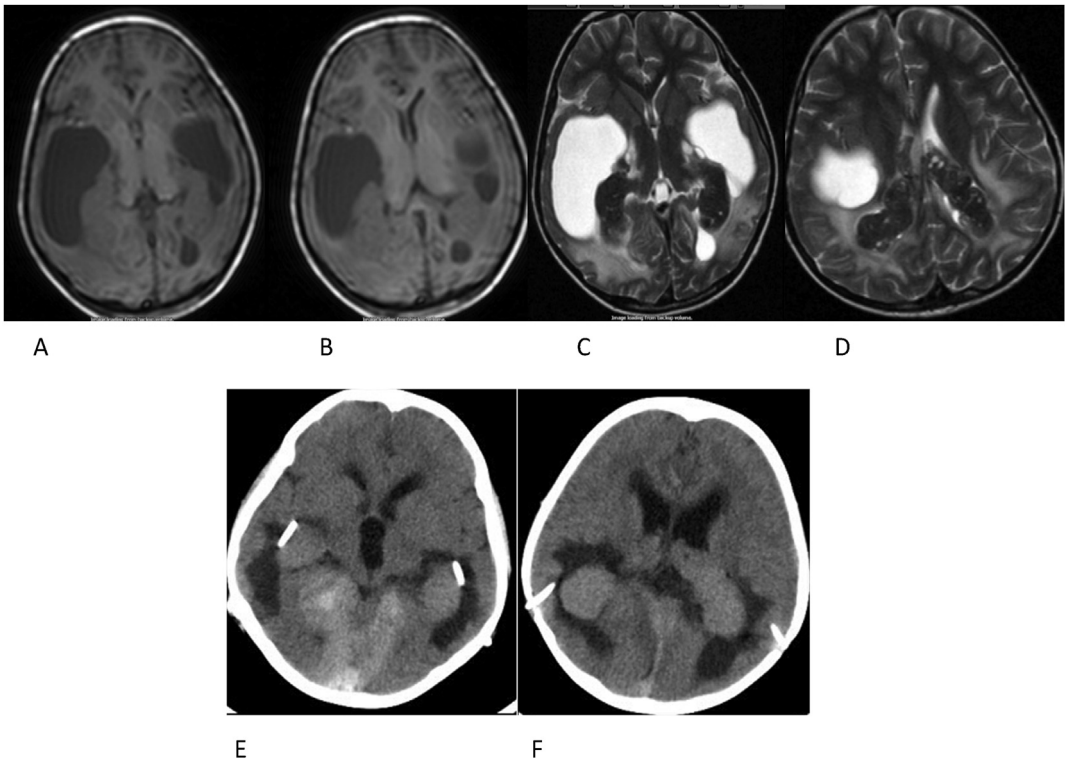

Fig. 1. A 6-year-old boy who complaint of headache and vomit for 1 week. Head MRI revealed multiple striped or specked space-occupying lesions in the choroid plexus of bilateral ventricle, cerebral falx and bilateral tentorium of cerebellum. The lesions showed iso-intensity on T1 weighted images (T1WI) and hypointensity on T2 weighted images (T2WI). Obvious supratentorial hydrocephalous and periventricular interstitial edema were seen secondary to compression of the interventricular foramen (A–D). Head CT two years later demonstrated some calcification in the lesions (E–F).

the right frontal lobe. The lesion was hypointense on TIWI, and heterogenous high signal on T2WI. Restrict diffusion and was seen on DWI perilesional edema had also been detected. After injecting contrast agent, the tumor showed strong homogenous contrast enhancement with a close connection with dural base (Fig. 2). Complete resection of the lesion had been performed. Pathologic examination confirmed the diagnosis of JXG.

3. Discussion

From 1980 to now, there are totally 43 cases of intracranial JXG had been reported [4]. Combined with 2 cases of our study together, 45 cases of intracranial JXG were reported and all of them are cases reported. Among 45 cases, 27 cases had routine MRI and 25 cases had contrast MRI examination. The detailed MRI appearances were summarized in Table 1. 16 cases had multiple intracranial lesions and some of them were extensive intracranial lesions [1,5,6]. There are three most common locations for the intracranial JXG, including parenchyma (10/27), ventricular-cortex (8/27) and dura (9/27). The most common imaging characterization is iso-hyper intensity on T1WI (18/20) and iso-hypo intensity on T2WI (14/18) (Table 2). Homogenous contrast enhancement has been seen 21 cases and heterogenous seen in 3 cases and only 1 case had no contrast enhancement.

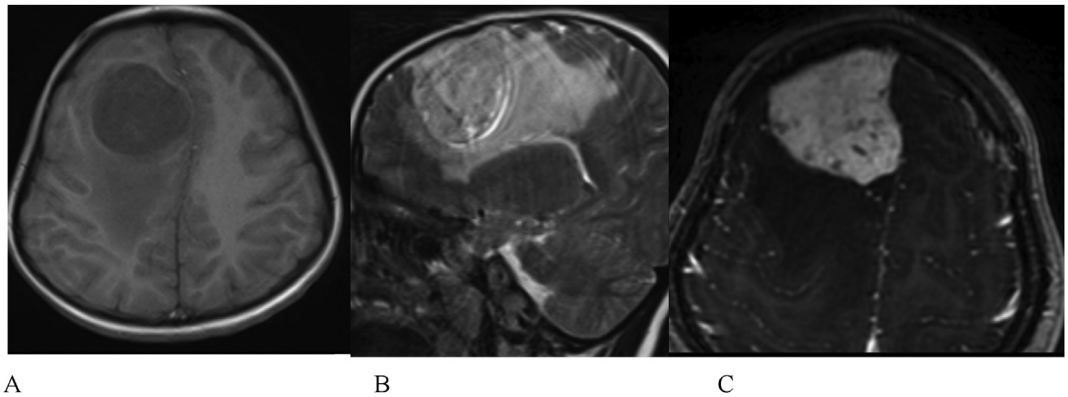

Fig. 2. A 7-year-old girl complaint of headache for 1 year. Brain MRI showed a solitary, well-demarcated mass in the right frontal lobe. The lesion was hypointense on TIWI, and heterogenous high signal on T2WI. After injecting contrast agent, the tumor showed strong homogenous contrast enhancement with a close connection with dural base.

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