RESEARCH PAPER

Cervicothoracic Lymphangioma with Periventricular Leukomalacia in a 4-Year-Old Child: MRI Findings and Clinical Correlation

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ABSTRACT

Lymphangiomas are benign congenital malformations of the lymphatic system that most commonly present in the head and neck region of children. They typically appear as painless, soft swellings that can enlarge or become symptomatic due to infection or hemorrhage. We report a case of a 4-year-old child with a history of delayed milestones and hypoxia at birth who presented with a left-sided neck swelling for evaluation. Magnetic resonance imaging (MRI) of the neck revealed a T1-hypointense and T2-hyperintense multiloculated lesion involving the left submandibular region, measuring approximately 3.8 × 1.8 cm, with multiple thin and thick septa. The lesion extended adjacent to the left pharynx and into the retropharyngeal space, demonstrating an enveloping phenomenon. A second smaller hyperintense lesion with septa was identified at the left thoracic inlet, measuring 2.1 × 1 cm. Additionally, the visualized brain showed periventricular hyperintensity in the posterior corona radiata bilaterally, consistent with periventricular leukomalacia (PVL) as a sequela of old hypoxic injury. Fine needle aspiration cytology (FNAC) suggested a benign cystic lesion, probably lymphangioma. This case illustrates the characteristic imaging features of cervicothoracic lymphangioma in a child with underlying hypoxic-ischemic encephalopathy and highlights the importance of MRI for accurate diagnosis and surgical planning.

Keywords: Lymphangioma; Cystic hygroma; Periventricular leukomalacia; Pediatric neck mass; MRI

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Introduction

Lymphangiomas are benign congenital malformations of the lymphatic system that result from failure of the developing lymphatic vessels to establish normal communication with the venous system.[1] They are most commonly diagnosed in children under two years of age, with approximately 50–60% presenting at birth and 80–90% by the end of the second year of life.[2] The head and neck region is the most frequent site of involvement, accounting for 70–80% of cases, particularly the posterior cervical triangle, submandibular region, and parotid area.[3]

Clinically, lymphangiomas typically present as soft, painless, compressible swellings that may enlarge rapidly during upper respiratory tract infections or following intralesional hemorrhage.[4] Large lesions can cause airway obstruction, feeding difficulties, or cosmetic deformity.[5] Imaging plays a crucial role in establishing the diagnosis, defining the extent of the lesion, and guiding surgical resection. Ultrasound is often the initial modality, but magnetic resonance imaging (MRI) is superior for demonstrating the multiloculated cystic nature, septal morphology, and relationship to adjacent vital structures.[6] On MRI, lymphangiomas appear as T1-hypointense and T2-hyperintense lesions with internal septa, often showing fluid-fluid levels if hemorrhagic.[7]

Periventricular leukomalacia (PVL) is a form of white matter injury that occurs in premature or term infants following hypoxic-ischemic events.[8] It is characterized by necrosis of periventricular white matter, particularly near the trigones of the lateral ventricles, and is a common cause of cerebral palsy and developmental delay.[9] The presence of both a cervicothoracic lymphangioma and PVL in a single patient is rare. We present a case of a 4-year-old child with delayed milestones and a history of hypoxia at birth who was found to have a multiloculated left neck mass extending to the thoracic inlet, along with MRI evidence of PVL. This report emphasizes the imaging features of lymphangioma and the importance of evaluating the brain in children with developmental delay.

Case Presentation

A 4-year-old child presented with a history of a left-sided neck swelling that had been noted by the parents for an unspecified duration. The child had a past medical history significant for delayed milestones and hypoxia at the time of birth. No details regarding gestational age, birth weight, or need for neonatal intensive care were available. On examination, there was a soft, non-tender, compressible swelling in the left submandibular region. No overlying skin changes or bruits were noted. No other swellings were palpable in the neck. The child was referred for magnetic resonance imaging (MRI) of the neck for further evaluation.

MRI of the neck was performed using T1-weighted, T2-weighted, and post-contrast sequences. The findings were as follows. A T1-hypointense and T2-hyperintense lesion was identified involving the left submandibular region, indenting the left submandibular gland and measuring approximately 3.8 × 1.8 cm (Figures 1 and 2). Multiple thin and thick internal septa were seen within the lesion. The lesion was located in the region of the second and third branchial clefts and did not extend into the posterior triangle. Notably, the lesion extended adjacent to the left pharynx and posteriorly into the retropharyngeal space, demonstrating a suspicious enveloping or wrapping phenomenon around adjacent structures (Figure 3). A second hyperintense lesion with internal septa was seen at the level of the left thoracic inlet, just above the great vessels, measuring approximately 2.1 × 1 cm (Figure 4). No significantly enlarged cervical lymph nodes were identified. The nasopharynx, oropharynx, laryngopharynx, epiglottis, valleculae, pyriform fossa, vocal cords, and larynx were normal. The carotid arteries and internal jugular veins showed normal flow voids. The thyroid gland was normal. The visualized skull base showed no abnormality.

Importantly, the MRI study included sections of the visualized brain. These images demonstrated periventricular hyperintensity in the posterior corona radiata bilaterally on T2-weighted and fluid-attenuated inversion recovery (FLAIR) sequences, consistent with periventricular leukomalacia (PVL) as a sequela of old hypoxic-ischemic injury (Figure 5). Based on the imaging features—multilocularity, T2 hyperintensity, internal septa, enveloping effect, retropharyngeal extension, and absence of significant solid component—a diagnosis of lymphangioma was favored. The differential diagnosis included a branchial cleft cyst, but the multilocular nature and extensive retropharyngeal involvement made lymphangioma more likely.

Fine needle aspiration cytology (FNAC) of the left neck swelling was subsequently performed. The cytological impression was a benign cystic lesion, probably lymphangioma of the left side of the neck. No surgical resection was performed at the time of this report. The child was referred to a pediatric surgeon for further management.

Discussion

This case illustrates the characteristic MRI findings of a cervicothoracic lymphangioma in a young child, along with an incidental finding of periventricular leukomalacia related to perinatal hypoxia. Lymphangiomas are part of the spectrum of vascular malformations and are classified histologically into three types: capillary lymphangiomas (lymphangioma circumscriptum), cavernous lymphangiomas, and cystic hygromas (macrocystic type).[1] The lesion in this case, with its large size, multilocular appearance, and extension into the retropharyngeal space, is most consistent with a macrocystic lymphangioma (cystic hygroma). These lesions are thought to arise from failure of the developing lymphatic sacs to connect with the jugular venous system during embryogenesis.[10]

The imaging diagnosis of lymphangioma is usually straightforward when characteristic features are present. On MRI, the lesion is typically hypointense on T1-weighted images and markedly hyperintense on T2-weighted images, reflecting its fluid content.[6] Internal septa of varying thickness are almost always seen, as in this case. Hemorrhage within the lesion may produce fluid-fluid levels or areas of increased T1 signal, but these were not prominent here. The enveloping or

infiltrative appearance—where the lesion seems to wrap around rather than displace adjacent structures—is a hallmark of lymphangiomas and helps distinguish them from other cystic neck masses such as branchial cleft cysts, which are usually unilocular and do not exhibit this behavior.[7]

The differential diagnosis of a multiloculated cystic neck mass in a child includes branchial cleft cyst (typically second branchial cleft, located anterior to the sternocleidomastoid muscle), ranula (sublingual or submandibular origin), dermoid cyst (more midline, may contain fat), and teratoma (often has solid components and calcification).[4] In this case, the location at the second and third branchial cleft region, the multilocularity, the retropharyngeal extension, and the presence of a second lesion at the thoracic inlet all favored lymphangioma. The FNAC result confirming a benign cystic lesion, probably lymphangioma, supported the imaging diagnosis.

An important additional finding in this case was periventricular leukomalacia (PVL) on the visualized brain sequences. PVL is a form of white matter injury that typically occurs in premature infants due to ischemia in the periventricular watershed zones.[8] The child had a history of hypoxia at birth, which explains the PVL. Delayed milestones are a common clinical consequence of PVL, often manifesting as spastic diplegia, cognitive impairment, or visual disturbances.[9] The fact that the child presented for evaluation of a neck swelling rather than the developmental delay underscores the importance of carefully reviewing all visualized structures on MRI studies. Radiologists interpreting neck MRI in children should always examine the included brain sections, as unsuspected intracranial abnormalities may be discovered.

Management of lymphangioma depends on the size, location, and symptoms. Small, asymptomatic lesions may be observed. Large or symptomatic lesions, especially those causing airway compromise or feeding difficulties, often require intervention.[10] Complete surgical excision is the treatment of choice but can be challenging due to the infiltrative nature of these lesions and the proximity of vital neurovascular structures. Incomplete resection is associated with recurrence rates as high as 10–15%.[5] Sclerotherapy with agents such as OK-432, bleomycin, or doxorubicin is an alternative for macrocystic lesions.[2] In this case, the child had a left submandibular lesion with retropharyngeal extension and a second thoracic inlet lesion, making complete surgical resection potentially difficult. A multidisciplinary approach involving pediatric surgery, interventional radiology, and otolaryngology would be ideal.

Limitations of this case report include the lack of contrast-enhanced MRI (which could have better delineated the septa and solid components), the absence of histopathological confirmation (only FNAC was performed), and the lack of follow-up data regarding treatment and outcome. Nonetheless, the imaging features are highly characteristic, and the case provides a valuable teaching point regarding the association of congenital lymphatic malformations with perinatal hypoxic injury, albeit likely coincidental.

Conclusion

Cervicothoracic lymphangioma in children presents with characteristic MRI findings of a multiloculated, T2-hyperintense lesion with internal septa and an enveloping growth pattern. The presence of a second lesion at the thoracic inlet in this case highlights the importance of imaging the entire extent of these malformations, as they can be multifocal. Additionally, the incidental discovery of periventricular leukomalacia on neck MRI underscores the need for radiologists to carefully evaluate all visualized structures, including the brain, when interpreting pediatric neck studies. Early and accurate diagnosis of lymphangioma is essential for appropriate management and to prevent complications such as airway compromise or infection.

References

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Figure Legends

Figure 1: Axial T2-weighted MRI shows a hyperintense lesion with internal septa involving the left submandibular region, enveloping adjacent structures. The lesion measures approximately 3.8 × 1.8 cm.

Figure 2: Coronal T2-weighted MRI demonstrates the hyperintense lesion in the left submandibular and sublingual region, with extension adjacent to the pharynx. Multiple thin and thick septa are visible.

Figure 3: Sagittal T2-weighted MRI shows the hyperintense lesion involving the region from the submandibular area extending posteriorly into the retropharyngeal space.

Figure 4: Axial T2-weighted MRI at the level of the thoracic inlet demonstrates a second hyperintense lesion with septa on the left side, measuring approximately 2.1 × 1 cm.

Figure 5: Axial T2-weighted or FLAIR image of the visualized brain shows periventricular hyperintensity in the posterior corona radiata bilaterally, consistent with periventricular leukomalacia (PVL) as a sequela of old hypoxic injury.

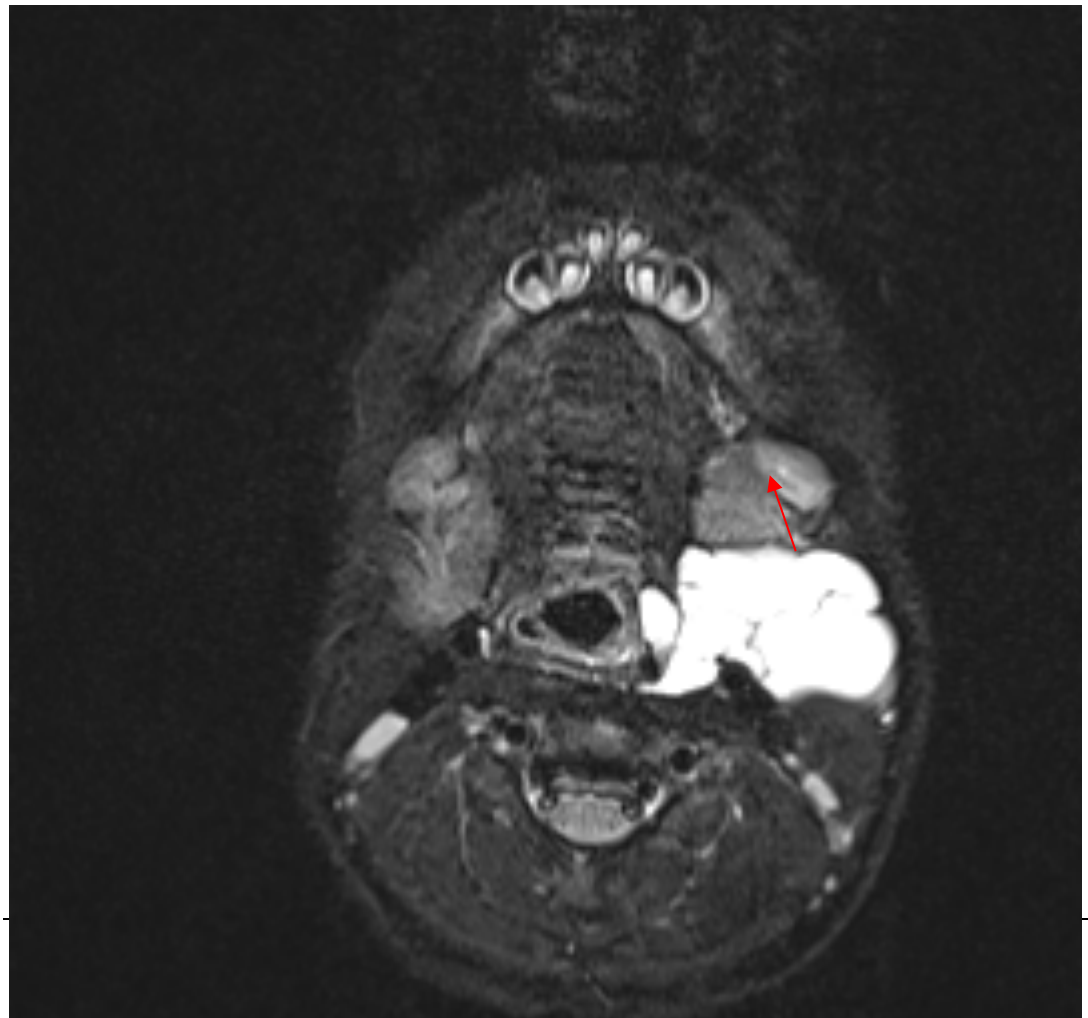

Figure 1

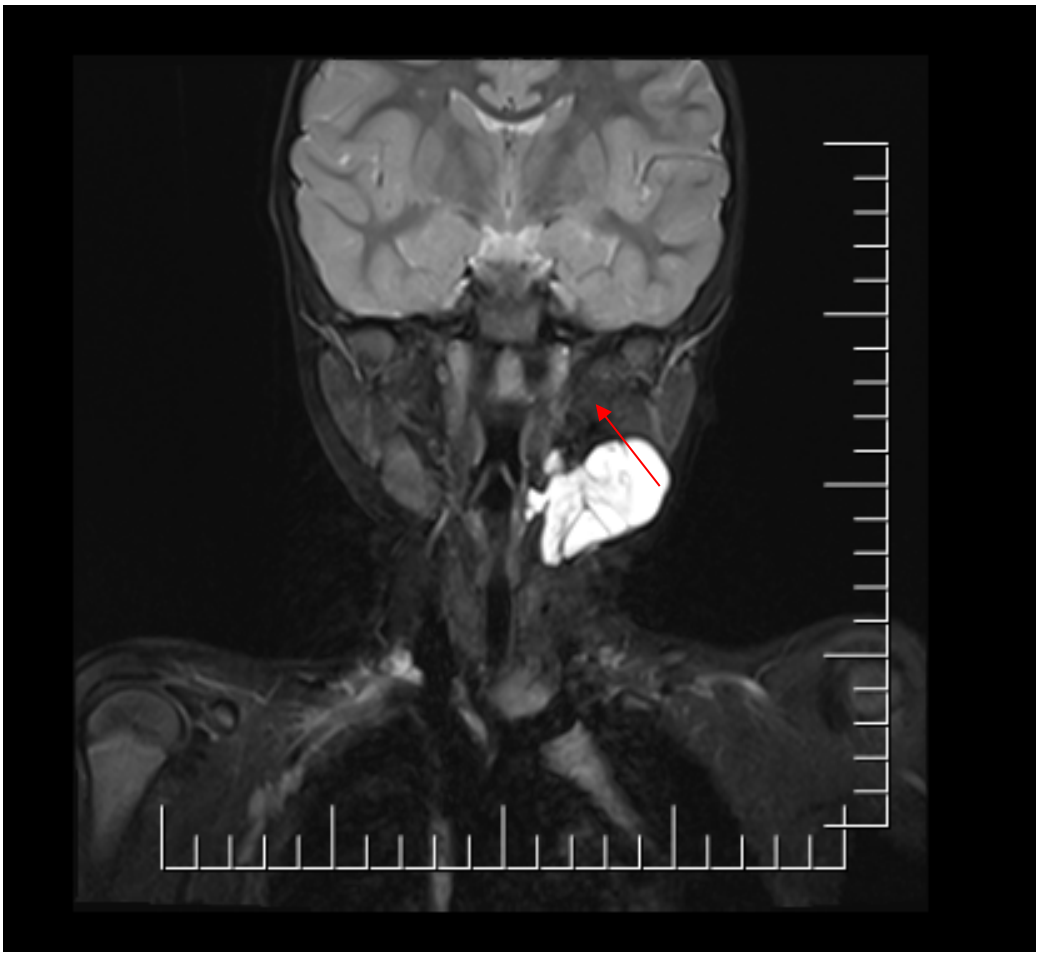

Figure 2

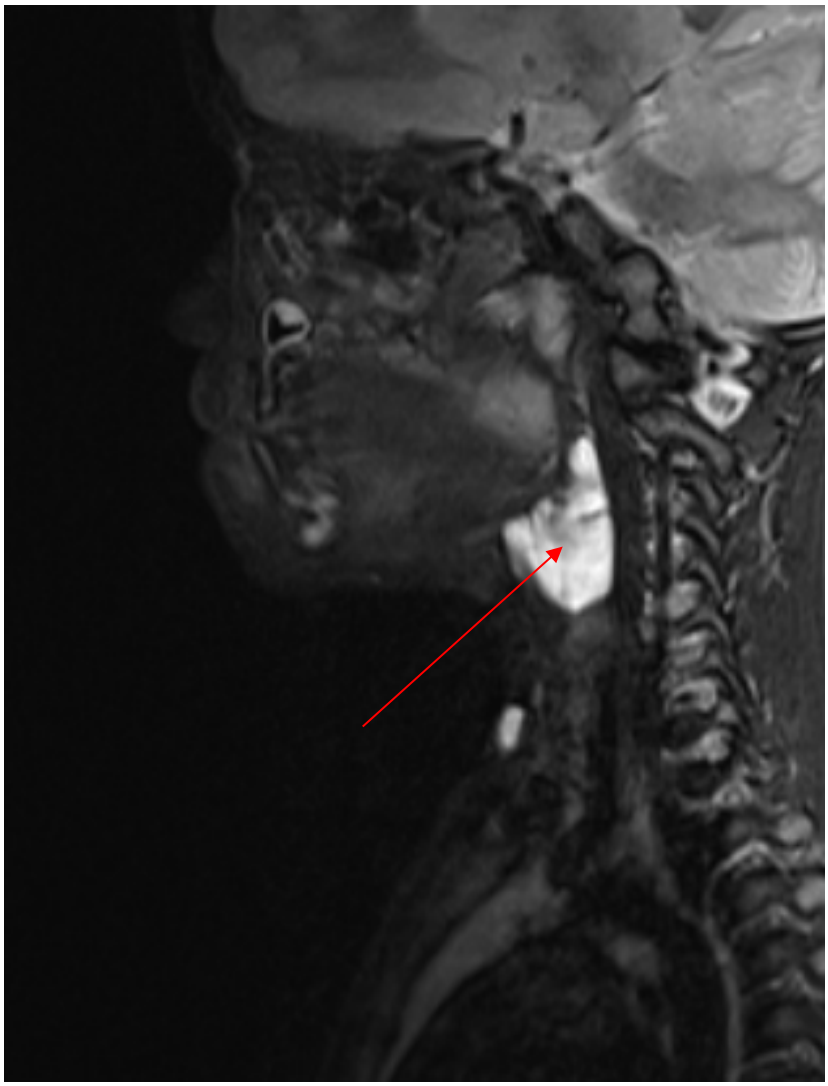

Figure 3

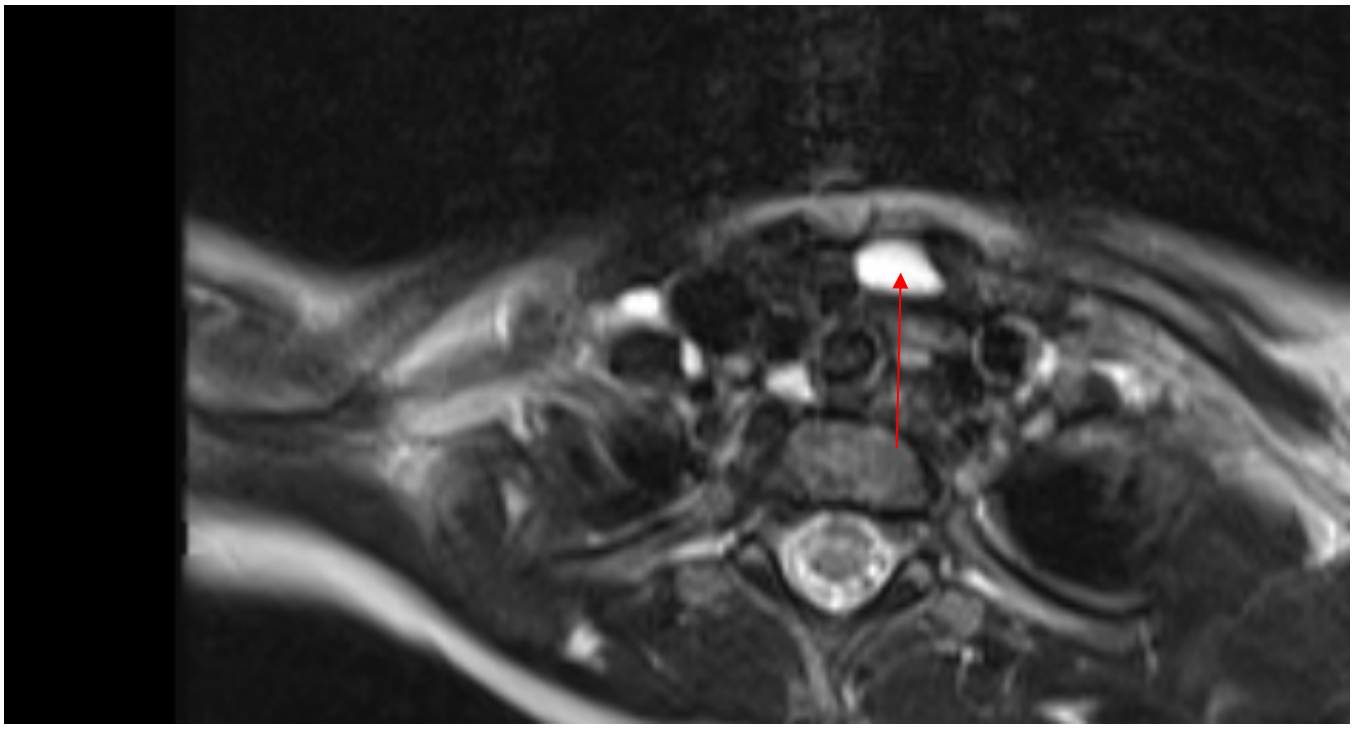

Figure 4

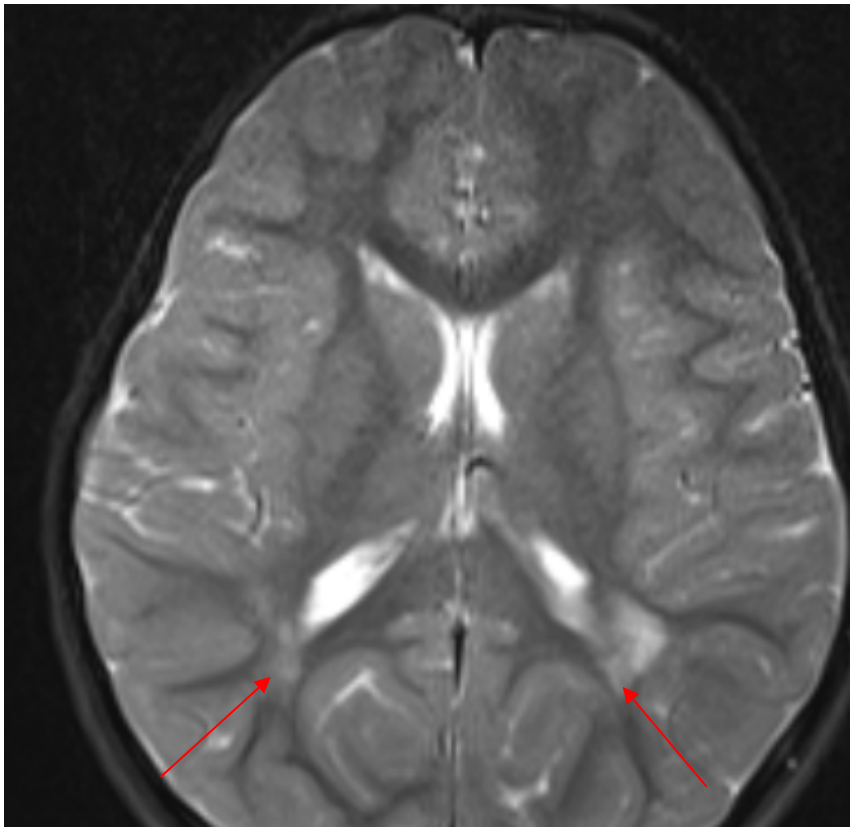

Figure 5

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