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A Pericallosal Lipoma Case with Evidence of Surface Dyslexia

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Pericallosal lipomas are thought to occur when mesenchymal tissue that forms the meninges fails to be resorbed between the 8th and 10th weeks of development (Truwit & Barkovich, 1990). Curvilinear lipomas are a subtype of pericallosal lipomas that are found along the posterior distribution of the corpus callosum and are also associated with atypical development of the corpus callosum (Tart & Quisling, 1991). While inspecting de-identified data for a retrospective multi-site imaging study, which was approved by Medical University of South Carolina Institutional Review Board, we unexpectedly identified an 11.31 year-old male with a curvilinear lipoma associated with splenium hypoplasia and expansion of the occipital horns of the lateral ventricles (Figure 1A). Behavioral assessment of this lipoma case revealed below average real word reading (WJ Letter-Word Identification¹ = 72), reading comprehension (WJ Passage Comprehension¹ = 81), and working memory (CTOPP Memory for Digits¹ = 85) scores, which were discrepant relative to the child's average to above average phonological processing (WJ Word Attack¹ = 126; CTOPP Elision¹ = 115), rapid naming for numbers and letters (CTOPP RAN for numbers = 95, letters = 100), verbal comprehension (WISC Verbal IQ¹ = 125), and visuospatial function scores (WISC Performance IQ¹ = 138). Thus, atypical development of the splenium and visual system was associated with the child's orthographic abilities but not phonologic, oral language, and visuospatial abilities. This behavioral profile is consistent with developmental surface dyslexia, or an impaired ability to acquire lexical representations of written words (Castles & Coltheart, 1993)².

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¹Behavioral assessment included subtests from the Woodcock-Johnson (WJ), Comprehensive Test of Phonological Processing (CTOPP), and the Wechsler Intelligence Scales for Children (WISC).

To determine the extent to which the lipoma case's visual system anatomy was atypical relative to 256 age-matched children (age range = 10.32 – 12.25 years; 146 male; 11 research sites), the T1-weighted image from the lipoma case was denoised, segmented, and diffeomorphically transformed to the space of a study-specific template that was created from segmented images from the 256 children using SPM12 DARTEL (Ashburner, 2007). Jacobian determinant images, which reflect how much each brain image was spatially warped to fit to the study template (Eckert et al., 2017), were used to determine the extent to which the lipoma case's brain image was atypical relative to the mean and SD of the comparison sample. This comparison highlighted significant differences between the lipoma case and comparison sample (i.e., atypical size and shape), which were pronounced in the splenium, optic radiations, and occipital cortex regions (Figure 1B) ³. The dysgenesis profile for this lipoma case is anatomically similar to the lesion profiles for pure alexia cases that have included damage to the splenium and white matter adjacent to the occipital horn of the lateral ventricle (Déjerine, 1892; Damasio, 1983; Maeshima et al., 2011), despite the lipoma case's ability to read pseudowords. At least one other case with atypical occipital lobe development has been reported to exhibit a relatively specific deficit in reading irregular words (Samuelsson, 2000).

Atypical development of splenial fibers occur with atypical development of superior parietal, medial parietal and occipital regions, based on normative evidence from diffusion imaging tractography and post-mortem studies (de Lacoste, Kirkpatrick, & Ross, 1985; Putnam et al., 2010). Inferior temporal cortex development could also be affected, based on evidence that splenium fibers provide inferior temporal neurons information about the ipsilateral visual field in rhesus monkeys (Gross, Bender, & Mishkin, 1977). Thus, cortical regions that represent visual word forms may be affected by atypical development of the splenium in cases with curvilinear lipomas. While curvilinear lipomas are often incidental findings during MRI and autopsy, they can be observed with ultrasound (Atallah et al., 2018) and may occur with impaired reading development.

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²The gold-standard for identification of surface dyslexia is a pure test of irregular word reading (e.g. yacht), which was not administered here. However, the WJ Letter-Word Identification test contains a preponderance of irregular words, and also multisyllabic words that require lexical knowledge for correct stress placement. Therefore, impairment on this test in the context of above average performance on the WJ Word Attack tests is strongly indicative of a surface dyslexic profile.

³The Jacobian determinant images from the entire sample of 256 children were used to examine the extent to which morphologic variation related to differences between Word Identification and Word Attack scores, after controlling for sex, total brain volume, and research site. While small to medium effect sizes were observed for the relation between visual cortex morphology and the difference between real word reading and pseudo-word reading (e.g., maximum Cohen's $D = 0.55$), no significant effects were observed after correction for multiple comparisons.

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Highlights

- A pericallosal lipoma case was unexpectedly identified in a multi-site retrospective reading disability dataset.
- The case had atypical visual system morphology relative to an age-matched comparison sample.
- The case had relatively poor real word reading compared to pseudo-word reading ability.
- The results suggest that atypical callosal development in lipoma cases can occur with surface dyslexia.

Figure 1.

Atypical splenial and visual cortex morphology in a child with a curvilinear lipoma and evidence of surface dyslexia. **A.** The T1-weighted image from an 11.31 year-old male with a lipoma (yellow) and with partial agenesis of the corpus callosum. The splenium is absent and there is resultant colpocephaly (dilatation of the occipital horns of the lateral ventricles). **B.** A Z-score image demonstrates where the lipoma case's brain image required significant spatial warping to align with a study-specific template that was created from an age-matched comparison sample (red clusters: large structures that required compression; blue cluster: small structures that required expansion; threshold: $Z > 3$ and displayed on the MNI template. The Z-score image was created from the lipoma case's Jacobian determinant image and the mean and SD of the Jacobian determinant images from the comparison sample).