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Corpus Callosum Lipomas in Children

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ABSTRACT

AIM: To determine the clinical and radiographical aspects of six patients diagnosed with pericallosal lipomas (PCL).

MATERIAL and METHODS: A retrospective analysis of patients who presented to the neurosurgery outpatient clinics of Selcuk Faculty of Medicine between 2009 and 2019, revealed that six patients were diagnosed with PCL. The clinical and magnetic resonance imaging (MRI) data were obtained by reviewing patients' records.

RESULTS: A total of six patients (two girls and four boys), with a mean age of 53.8 months (38–72 months), were included in this study. They were followed up for a mean period of 36.5 months (32–41 months). PCL were detected on MRIs, which were obtained to investigate headache in two patients, epilepsy in one patient, frontal dermal sinus tract and left frontal epidermoid tumor in one patient, and subcutaneous lipoma associated with PCL in one patient. Five patients displayed tubulonodular lipomas and one patient displayed curvilinear lipomas. Agenesis or dysgenesis of the corpus callosum (CC) was observed in four (66%) patients. Two patients received surgical treatment for cosmetic skin problem.

CONCLUSION: Because of the benign course of PCL, i.e. no growth or very slow growth, and close proximity to the surrounding neurovascular structures, surgical removal should be considered only in symptomatic PCL. Furthermore, other malformations and anomalies may accompany PCL.

KEYWORDS: Pericallosal lipoma, Corpus callosum agenesis, Corpus callosum dysgenesis

INTRODUCTION

Intracranial lipomas are congenital malformations that are rare and accounting for 0.1%–0.5% of all intracranial tumors (3,13). They were first described by von Rokitansky for the first time in 1856 as a slow-growing, benign and hamartomatous condition (13,15). The origin of intracranial lipomas is unknown, and they are still considered to be a primary malformation is still accepted. Their origin may be related to the abnormal persistence and maldifferentiation of the primitive meninges during subarachnoid cisterns developments. This hypothesis is consistent with the fact that both the vessels and nerves go through the lipomas instead of getting displaced by them (9,14).

Pericallosal lipomas (PCLs) are known to be congenital, fat-containing brain malformations, which are closely associated with primitive meninges and corpus callosum (CC) development. PCLs usually remain undetected during the fetal stage. Furthermore, among all intracranial lipomas; PCLs are the most frequently occurring lipomas (14,16). PCLs are located within the interhemispheric fissure and they are defined by ultrasonography as hyperechoic, solid and well-demarcated masses (11,14).

Microscopically; they comprise fats, calcifications, and, on occasion, vascular structures. Imaging is crucial for the diagnosis and follow-up of PLC (4,13). Surgical treatment is

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controversial in such cases of asymptomatic lesions; moreover the risks of surgery can outweigh the potential benefits (13,18).

PCL has been described to have two morphological types: tubulonodular and curvilinear (9,16-18). The tubulonodular lipomas are reportedly more frequently associated with CC abnormalities. This study report the radiological and clinical findings of six patients diagnosed with PCL at our institution during 2009-2019.

MATERIAL and METHODS

A retrospective analysis of patients presenting at the neurosurgery outpatient clinics between 2009 and 2019 was performed. Six patients diagnosed with PCL were enrolled in this study. The patients' files, follow-up records, radiological reports, medical charts were reviewed and their magnetic resonance imaging (MRI) and clinical data obtained (Table I).

RESULTS

Table I demonstrates the clinical and MRI findings of the patients who were diagnosed with PCL. A total of six patients (two girls and four boys), with a mean age of 53.8 months (38–72 months), were included in this study. PCL were detected on MRIs, which were taken to investigate headache in two patients, epilepsy in one patient, frontal dermal sinus tract and left frontal epidermoid tumor (Figure 1A, B) in one patient, and subcutaneous lipoma associated with of PCL (Figure 2A, B) in one patient.

Evaluation of cranial MRIs showed that five patients exhibited tubulonodular lipomas and one patient exhibited curvilinear lipomas (Figure 3A-D). Agenesis or dysgenesis in the CC was observed in four (66%) patients. All four patients with CC malformation had tubulonodular type PCL. The mean follow-up period was 36,5 months (32-41 months).

Table I: Results of Retrospective Analysis of Six Pediatric Corpus Callosum Cases Followed Up and Treated

No	Age (months)	Sex	Clinical Presentation	Associated Malformation	Follow–up period (months)	Treatment	Clinical and Radiological Course	Morphology
1	72	F	Cosmetic skin problem	Dermal Sinus Tract and Epidermoid Tumor	36	Surgical treatment (for Dermal Sinus Tract and Epidermoid Tumor)	Stable	Tubulonoduler
2	38	M	Cosmetic skin problem	Corpus Callosum Dysgenesis and Extradural Lipoma Extension	32	Surgical treatment (for Extradural Lipoma Extension)	Stable	Tubulonoduler
3	44	M	Epilepsy	Corpus Callosum Agenesis	36	No	Stable	Tubulonoduler
4	53	M	No	No	41	No	Stable	Curvilineer
5	70	F	Headache	Corpus Callosum Dysgenesis	40	No	Stable	Tubulonoduler
6	46	M	Headache	Corpus Callosum Agenesis	34	No	Stable	Tubulonoduler

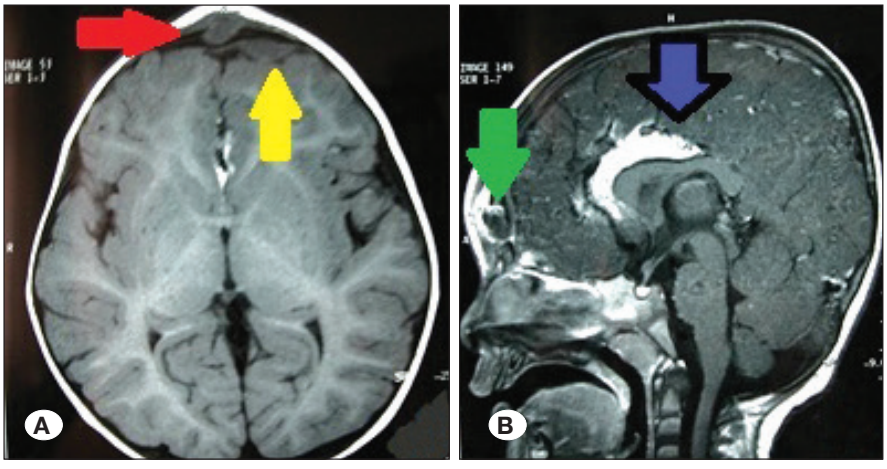

Figure 1: A) Red arrow indicates frontal dermal sinus tract and yellow arrow indicates epidermoid tumor on left frontal side in the axial MRI.
B) Green arrow indicates frontal dermal sinus tract and blue arrow indicates tubulonodular corpus callosum lipoma in the sagittal cross-sectional MRI (patient number 1).

Cosmetic skin problem in two patients received surgical treatment. In addition, surgery in the dermal sinus tract was performed in one patient. In another patient, subcutaneous lipoma associated with PCL was operated, whereas the area with intradural extension of PCL was not operated.

DISCUSSION

Intracranial lipomas are mostly asymptomatic and are usually detected during neuroradiological investigations for other conditions (6,10). They mostly develop on the midline and most commonly involve midline structures such as the CC and quadrigeminal cistern (6,10,17). These lipomas have also been reported to occur in the pontocerebellar angle, prepontine cistern, sylvian fissure, pontocerebellar angle, interhemispheric fissure, cervicomedullary junction, Galen vein system and cerebral convexity (1,2,18). However, along with accompanying anomalies, patients with intracerebral lipomas may admit with a history of psychomotor retardation, cerebral palsy or epilepsy (6,18).

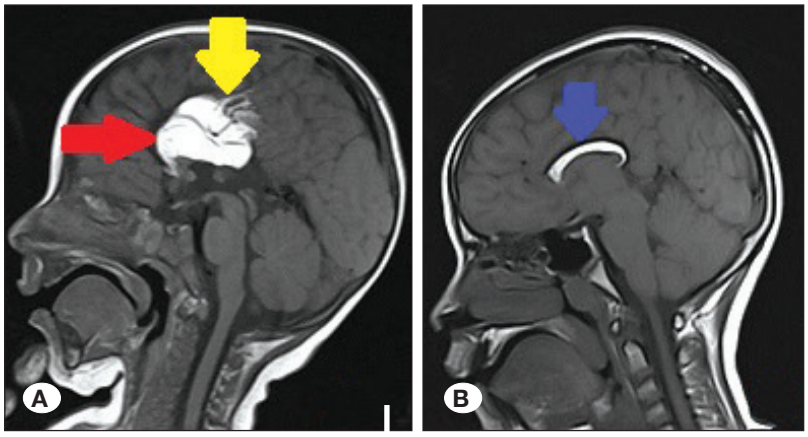

Figure 2: A) Red arrow indicates corpus callosum agenesis and yellow arrow indicates tubulonodular corpus callosum lipoma in the sagittal cross-sectional MRI (patient number 3). **B)** Blue arrow indicates corpus callosum curvilinear lipoma in the sagittal cross-sectional MRI (patient number 4).

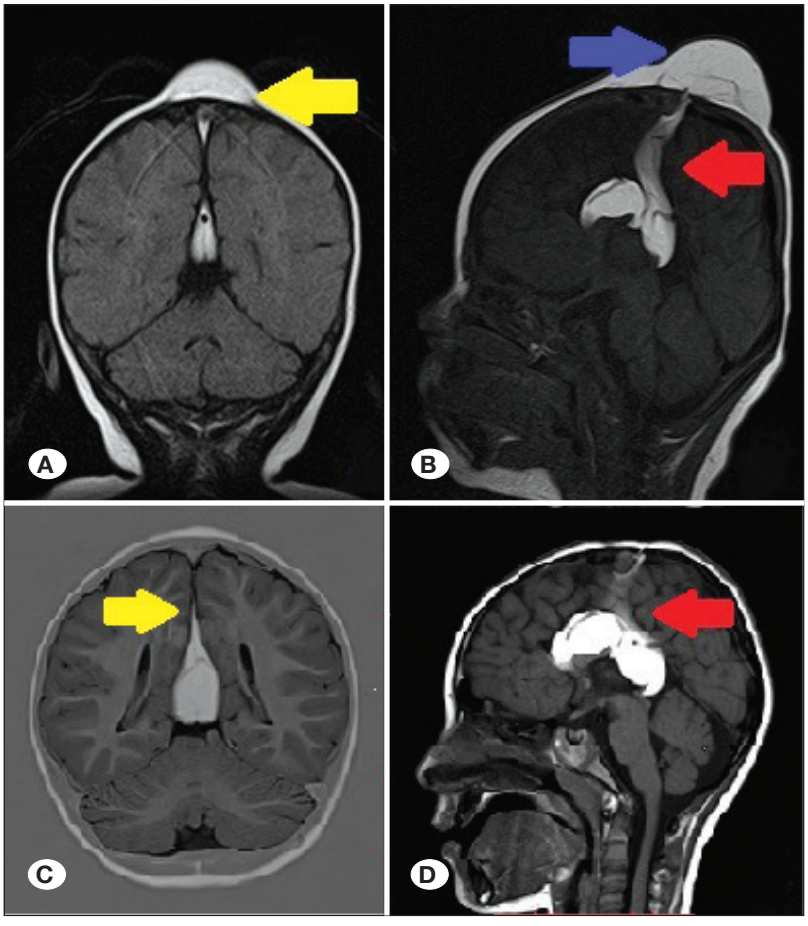

Figure 3: A) Yellow arrow indicates subcutaneous lipoma associated with pericallosal lipoma in the coronal cross-sectional MRI. **B)** Blue arrow indicates subcutaneous lipoma associated with pericallosal lipoma and red arrow indicates the association tract of subcutaneous lipoma and pericallosal lipoma in the sagittal cross-sectional MRI (patient number 2). **C)** Postoperative subcutaneous lipoma associated with pericallosal lipoma in the coronal cross-sectional MRI. **D)** Postoperative subcutaneous lipoma associated with pericallosal lipoma in the sagittal cross-sectional MRI (patient number 2).

CC agenesis has particularly been reported to have an association with psychomotor retardation, cerebral palsy or epilepsy (9,17). In this study, one patient with CC agenesis had epilepsy. We suggest that the frequent occurrence of epilepsy and psychomotor retardation in PCL is associated with the frequent occurrence of agenesis and dysgenesis of the CC in PCL.

Lipomas are assumed to develop as a result of abnormal persistence or maldifferentiation of primitive meninges, during subarachnoid space development between the 8th and 10th week of embryonic development (7,9). Similarly, poorly differentiated meningeal remnants are considered to give rise to lipomas around the CC (6,10,18).

CC dysgenesis and agenesis result from a neural tube closure defect (6). Reportedly, CC dysgenesis or agenesis exists in 50% of PCL cases (5,16). Another study reported that CC dysgenesis exists in 40% of PCL cases (6). In this study, 66% (four patients) of the patients with PCL have CC agenesis or dysgenesis.

Two subtypes, i.e., tubulonodular and curvilinear, of PCL have been identified based on their associated anomalies and morphology (10). The tubulonodular type is comprised of anteriorly present round- or cylinder-shaped lipomas that are generally >2 cm in diameter. They are closely related to CC dysgenesis, frontal encephaloceles and frontal lobe anomalies (9,12). In this study, four of five patients with tubulonodular type had CC agenesis or dysgenesis. One of these patients had frontal dermal sinus tract and left frontal epidermoid tumor and one patient had subcutaneous lipoma associated with PCL.

The curvilinear lipomas usually present as a thin, posteriorly situated mass that, curves around the splenium, and is <1 cm in size. These are usually associated with a normal CC and exhibit a low incidence of associated anomalies (12). One patient had curvilinear type of PCL. Consistent with previous studies, no CC anomalies were founding this patient.

Most patients with PCL are asymptomatic. The lesions are usually detected incidentally, and they have slow progression. Surgical removal is not recommended, as the complication rate is high and these lesions have a benign course (8,9,18).

In this study, asymptomatic PCLs were not operated. Two patients received surgical treatment. One patient received surgical treatment because of the existence of dermal sinus tract and epidermoid tumor. In one patient, subcutaneous lipoma associated with PCL was surgically removed and intradural field surgery was not performed due to the absence of symptoms. In other words, no surgical treatment for pure PCL was performed.

CONCLUSION

Because of the benign course of PCL, i.e. no growth or very slow growth, and close proximity to the surrounding neurovascular structures, the option of surgical removal should be considered only in symptomatic PCL. Furthermore, other malformations and anomalies may accompany PCL.

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