

CASE REPORT

Lipoma of the Corpus callosum: Report of a Case and Review of the Literature

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Abstract

Intracranial lipomas account for less than 1% of all brain tumors. Corpus callosum lipoma (CCL) is a rare finding and up until now, documented cases are around 200 patients. These lipomas remain mostly asymptomatic, but when symptoms appear, they present through headache, seizure, dementia and in some cases, rather uncommon neurological disorders such as mirror writing.

Fourteen years old girl with history of 8 years headache and recent development of vertigo, extremities trembling and drop attack was referred to neurologist and brain imaging confirmed CCL.

Herein, we report another rare case of CCL, with common and uncommon symptoms of latter tumor in detail; in addition to reviewing the previous 80 years case reports aiming to achieve comprehensive perception from CCL.

Keywords: Lipoma; Corpus callosum; Brain tumors

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Intracranial lipomas, which originates from abnormal differentiate meninx primitive mesenchymal tissue, are rare brain tumors, accounting for less than 1% of all brain tumors (1) are rare brain tumors, accounting for less than 1% of all brain tumors. The most common site of these tumors is corpus callosum and there is no significant gender preference in its frequency. It is recommended to name these abnormal structures as a choristomas rather than neoplasms since they comprise of fat cells but are misplaced (2). Deletion of Xp is a recent suggested etiology accommodated with previous studies which also suggested the genetic basic of this condition leading to meningeal maldifferentiation and dysraphism. Morphological classification consists of anterior and posterior, and anterior lipomas are more associated with agenesis of corpus callosum (CC), which

are thicker. This lipoma remains asymptomatic in near half of the rare reported cases, which mostly presented as convulsion, headache, mental and behavioral disorders. Some patients develop rather uncommon neurological symptoms (3, 4). Many anomalies are reported alongside this tumor: corpus callosum agenesis as the most prevalent one, webbed toes, cleft lip, mongolism, funnel chest, facial asymmetry, high arched palate, ventricular septal defect, agenesis of the cerebellar vermis, pituitary tumor and acoustic neurofibroma which calls for further evaluation (3-9). Here, we reported another rare case of corpus callosum lipoma (CCL) in detail, in addition to reviewing the previous 80 years case reports aiming to achieve more comprehensive perception from CCL.

Case presentation

A 14 years old fair skin complexion girl from Iranian descent referred to cardiologist after a drop attack. Patient complained of a single episode of loss of consciousness, dullness and a headache after the aforementioned attack with history of lightheadedness before its occurrence. Cardiologist evaluated the patient for orthostatic

hypotension, arrhythmias, inner ear, vestibular, and cardiac structural disorders. Electrocardiogram showed normal sinus rhythm with normal axis, and echocardiography determined left ventricle ejection fraction of 60% without any valvular abnormalities. Patient's medical history revealed that the patient had been suffering from prolonged occasional headaches for 8 years, which is usually resolved spontaneously without response to painkillers and got visited by an ophthalmologist who ensured the patient of no sign of increased intracranial pressure. In the last 2 months, she developed vertigo and extremities trembling. Based on latter history, cardiologist ordered brain (computerized tomography) CT scan and referred the patients to a neurologist (figure 1). Neurological examination was normal with history of normal development and no significant family history. CT scan revealed midline abnormal structure. Neurologist ordered brain magnetic resonance imaging (MRI) as follow which confirmed the CCL (figure 2). For further assessment neurosurgeon consult was done, and annual brain MRI in addition to medication therapy with Carbamazepine 200 mg/day was decided

Table 1. Case review of the past 72 years.

Case	Year	Author	Age (years)	Gender	Convulsion	LOC	Headache	Behavioral disorder	Developmental/Mental retardation	Memory loss	Uncommon sign and symptoms	Craniofacial abnormalities
1	1946	Amyot(10)	39	F	+		+					
2	1949	Sutton(10)	25	F	+				+		Attacks of feeling of falling over since age 10	
3	1949	Sutton(10)	23	F	+						Sleep walking and attacks of leg weakness	
4	1950	Mullen(10)	30	M	+			+	+		Absence of triceps and Achilles reflexes, with wide-based gait and veering to left	
5	1951	Kinal	17	M	+			+	+			+
6	1951	Talairach(10)	16	F	+				+			
7	1951	Sheinmel(10)	20	M	+						Spina bifidia	
8	1951	Luten(10)	15	M		+		+			facial redness	
9	1951	Luten(10)	18	-					+		hyperactive left biceps and loss of right ankle jerk; restricted visual fields	+
10	1952	Cant(10)	5	M	+							+
11	1952	di Chiro (10)	31	M	+		+					
12	1953	Anderson (10)	24	M		+					Nocturnal attacks of crying and screaming	
13	1953	Huber (10)	13	M		+					Jacksonian attacks	
14	1953	Smith (10)	15	F	+		+	+				
15	1955	Nordin(10)	15	F	+							+
16	1960	Bonnal(11)	38	F	+							
17	1962	Ketz (11)	14	F	+		+				spastic hemiplegia	
18	1962	Bossi(11)	15	M	+				+			
19	1962	Bianchedi (11)	1	M					+			+

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Case	Year	Author	Age (years)	Gender	Convulsion	LOC	Headache	Behavioral disorder	Developmental/Mental retardation	Memory loss	Uncommon sign and symptoms	Craniofacial abnormalities
20	1965	Trillet(11)	19	F	+				+		vertigo, nystagmus. athetosis, microcephalus, syndactyly	
21	1966	Manganiello(11)	42	F			+	+			papilledema	
22	1967	Sabouraud (11)	27	M	+							
23	1967	Sabouraud (11)	31	M	+							
24	1972	Wolpert(12)	21	M	+		+					
25	1974	Cascino(11)	5	M	+		+					+
26	1975	Turpin(11)	8	M	+							+
27	1975	Turpin (11)	2 m/o	M							craniostosis	
28	1975	Turpin (11)	7 m/o	M					+			
29	1975	Turpin (11)	11	M	+				+			
30	1975	Turpin (11)	13	M	+			+				
31	1976	Wallac (2)	22	F	+	+			+		hemiplegic gait	
32	1976	Wallace (2)	8	M		+	+					
33	1977	Kushnet (13)	64	F	+							+
34	1978	Suemitsu(11)	13	M			+				gastrocnemius cramps while sleeping	+
35	1981	Nabawi (14)	43	F			+					
36	1981	Nabawi (14)	68	M							Light irritating the eye	
37	1981	Nabawi(14)	54	M			+					
38	1981	Nabawi(14)	55	F			+					
39	1981	Nabawi(14)	49	M	+		+			+	Intellectual and affective dullness. Spastic hemiparesis	
40	1982	Gerber (4)	35	M			+			+		
41	1982	Grasso (15)	35	M	+	+				+		
42	1982	Fujil(16)	2	M						+	Cerebral palsy	

Case	Year	Author	Age (years)	Gender	Convulsion	LOC	Headache	Behavioral disorder	Developmental/Mental retardation	Memory loss	Uncommon sign and symptoms	Craniofacial abnormalities
43	1990	Donati(17)	3m/o	M							hypotonia and bad control of the head	+
44	1993	Meguid (18)	6	M							Tetralogy of Fallot	+
45	1995	Silva (19)	18	F	+							
46	1997	Pinkofsky (20)	28	F					+	+	Schizophrenia	
47	2005	Alam(3)	22	M	+							
48	2010	Hidalgo (21)	19	M							Reading disabilities and mirror writing	
49	2014	Belkouch (22)	18	—	+		+					
50	2014	Navarrete-Dechent (23)	4 m/o	F								+
51	2015	AlJallaf (24)	38	M		+						
52	2016	Moreno (25)	1d/o	F								+
53	2017	Shah(26)	2	F								+
54	2018	Elhend(1)	18	M	+		+					
55	2018	Elhend(1)	24	M	+		+				Vertigo	

LOC: loss of consciousness, F: female, M: Male, m/o: month old

Table 2. Age at diagnosis of corpus callosum lipoma

Age at diagnosis	Number	Percent (%)
0-10	12	13.1
10-20	18	31.6
20-30	10	23.6
30-40	8	13.1
40-50	3	6.2
50-60	2	6.2
60-70	2	6.2
Total	55	100

Table 3. Prevalence of common symptoms amongst patients with corpus callosum lipoma

Symptoms	Number (n)	Percent (%)
Convulsion	31	56.3
LOC	7	12.7
Headache	17	30.9
Behavioral disorders	5	9
Developmental/mental retardation	11	20
Memory loss	5	9
Craniofacial abnormalities	14	25.4

LOC: loss of consciousness

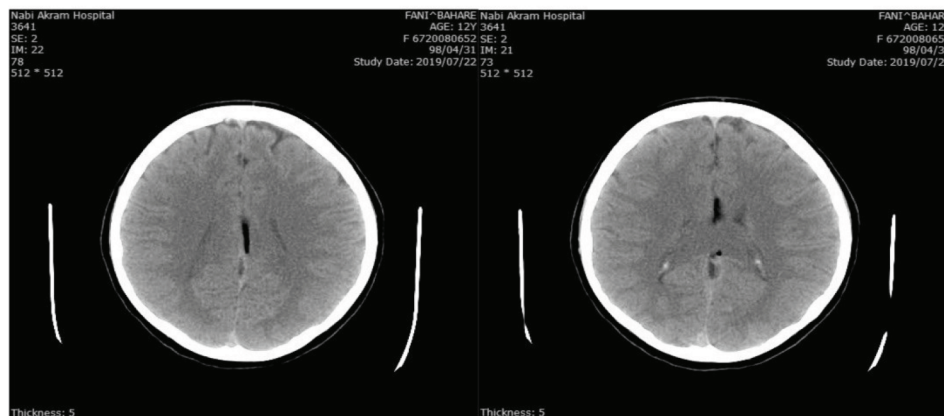


Figure 1. There is a tubular mass lesion extending superior to the corpus callosum with fat attenuation indicating curvilinear pericallosal lipoma.

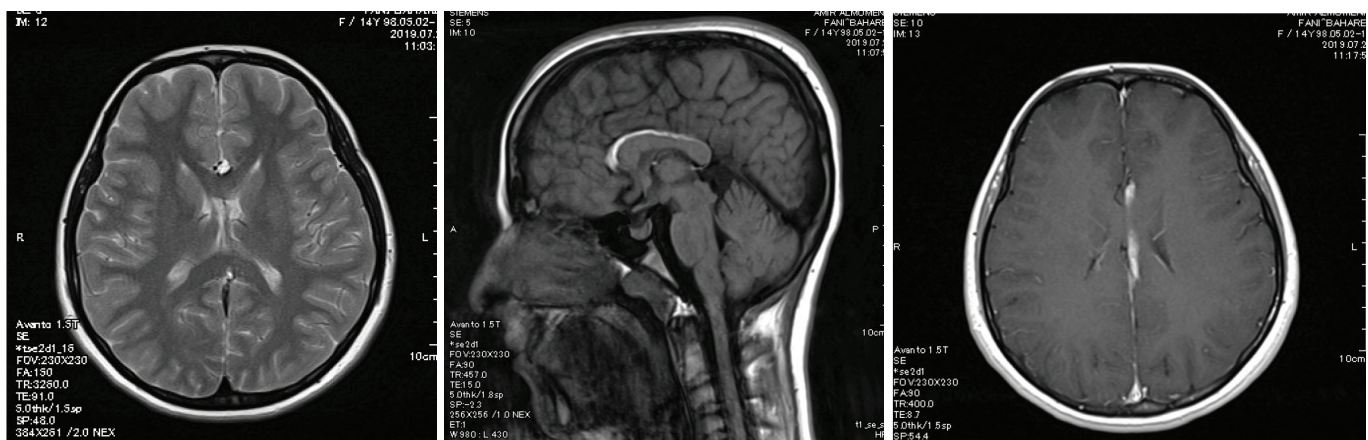


Figure 2. A T1 hypersignal tubular mass lesion superior to CC consistent with lipoma. Mild suspicious hypoplasia of splenium of CC was observed.

Discussion

Since the first report of CC lipoma by Rockitansky in 1856, many theories regarding its etiology/pathogenesis have been suggested such as pre-existing adipose tissue hypertrophy and a pseudo tumor from primitive meninges (27, 28). Nowadays, it is believed that pathogenesis is as a result of premature junction of neural ectoderm before closure of neural tube (1). Reviewing literatures of nearly 70 years fades the statement of CC lipoma are mainly asymptomatic, which could be due to increased numbers of latter case with wider range of symptoms and perhaps increased size (table 1) (10). The most reported symptoms are seizure, headache, behavioral disorders, dementia, hemiplegia and developmental disorders (4, 6). Reviewing almost 70 cases revealed convulsion was the most prevalent symptom with different presentations amongst more than half of the cases. It seems the etiology of convulsion is due infiltration of the neuronal tissue by dense gliosis and not the local pressure (28). In the presented case, the patient had a episode of post-ictal, described as LOC and dullness without remembering certain episode of time. Although her mother described it as a drop attack, we could not confirm convulsion. It is confirmed that surgery and resection of lipoma could not guaranty resolving the conclusion (29). Our patients most irritating and prolonged symptom was headache, which according to our literature review counts as the second most common symptom amongst CCL patients. Headache is often described in severe, intractable and generalized terms without responding to pain-killers amongst CC lipoma patients (3). In our case, even with 7 years history of headache, no neurologic evaluation was ordered until the drop attack. Other common symptoms

according to literature review were as follows: LOC, memory loss, developmental retardation and behavioral disorders. The aforementioned symptoms are unspecific, but usually grab the attention of physicians, by provoking image evaluation. However, in some cases there have been some report of behavioral and psychiatric disorders without considering pathologic etiologies which is alarming (30, 31). In comparison with reported clinical future in a sample size twice as ours by Suemitsu et al. convulsion rate was almost the same, but we recorded double rate of headaches. Trembling of extremities, as a very rare symptom which Suemitsu et al. recorded only 4 cases amongst near 80 cases, was also present in our case (11).

As for gender distribution, previous studies declared no difference in CC lipoma frequency between male and female cases while we recorded male to female 2:1 ratio. Baker et al. reported Xp22.31-Xp22.12 deletion in a female patient with CC lipoma, they declared no correlation between gender and chromosomal and occurrence of latter disorder since deletion of the mentioned chromosome would be lethal in males. However, controversies between our report and previous ones calls for further chromosomal study amongst reported CCL patients to reach a certain conclusion (6). Most patients were diagnosed with CCL in their second and third decades of life (3) while we recorded most decades of which diagnosis was made were the second and first. Most patients who were diagnosed with CCL during the first decade had frontonasal defect which led to its diagnosis. Symptom presentation decreases with age, which can almost ensure if patients have not developed symptoms by 30-40 years old, there is a great chance they never will (table 2).

We observed 14 craniofacial anomalies, which were mostly subcutaneous lipoma and midline deformities and a considerable attention was given to this issue. With our further evaluation, some case had less significant facial lipoma which was considered as a birth mark or ignored. Latter point in addition to the fact that in neurologic diseases skin disorders should be given more attention toward skin abnormal structure and neurological assessment. We managed our case with anti-convulsant, similar to previous reported CCL cases, and surgical approach was not considered after consultation with neurosurgeons. They declared that due to small size, location and insignificant and manageable symptoms of patient, yearly follow up MRI would be sufficing. Patient was asked to report if any symptoms or drug adverse events happened. Operative approach to CCL is almost contraindicated due too poor outcomes and mortality (8). In late 70s a literature review of operational outcomes amongst 22 CC lipoma patients revealed 63% mortality and only 14% improvement. In comparison with medical therapy, it is advised to avoid the surgical removal of CCL as much as possible as some even called it a contraindication (7, 30).

In conclusion CCLs reports are increasing as well as their symptoms which calls for serious attention as they are no longer very rare conditions. Convulsion, headaches and behavioral disorders are most prevalent symptoms in addition to noticeable records of frontonasal dysplasia. Management of CCL is recommended to be symptom therapy rather than surgical, unless highly indicated.

List of abbreviation

Computerized tomography: CT

Corpus callosum: CC

Corpus callosum lipoma: CCL

Magnetic resonance imaging: MRI

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Author's Contribution

All authors visited the patient at some point and were associated in final diagnosis. Dr.Ashkooh provided the writing of case presentation while Dr.Bozorgi contributed to provide a literature review and discussion with Dr.Safari guidance and collaboration

Conflict of Interest

The authors declare that they have no competing interests

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