CALVARIAL INTRAOSSEOUS LIPOMAS: TO SURGERY OR NOT TO SURGERY?

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ABSTRACT Intraosseous lipomas (IOLs) are rare benign bone tumors,accounts for 0.1% of all bone tumors. The disease is usually asymptomatic. Hips, vertebrae, ribs may be involved. However, the IOL of the skull is less common.There is no consensus about the indications of treatment. Most of the time lesions are operated for cosmetic reasons. In the presence of neurological deficits, dura infiltration, and massive bone destruction, surgical treatment is recommended to confirm the diagnosis. It has been proposed that the excision of an asymptomatic lipoma is not necessary when the diagnosis is definite. The risk of malignant transformation in IOL is very low. We present a case with radiologic evidence highly suggestive of skull lipoma. The patient refused surgery.The patient was followed at 3-month intervals. There was no change in imaging and the patient was neurologically intact at the end of one year.

KEYWORDS Intraosseous lipoma, skull, computed tomography,magnetic resonance imaging

INTRODUCTION

Calvarial masses can be classified into three groups as 1-benign lesions, 2-tumour-like lesions and 3-malignant lesions[1]. Bone lipomas are benign tumors derived from a proliferation of mature lipocytes. Intraosseous lipomas(IOLs) are among the most uncommon tumors of bone. Their incidence has been reported as 0.1% of all bone tumors[2,3,4,5,6,13]. In literature, there are 249 articles as intraosseous lipoma and only 28 articles with intraosseous lipomas of the skull[6]. İntraosseous lipomas (IOL) of the skull are rare benign bone tumors often seen in the frontal, parietal, temporal and ethmoid bones[2,3]. Clinical and radiologic follow-ups at regular intervals are preferred in asymptomatic patients without cosmetic deformities. Surgery as the mode of treatment remains controversial; however, the consensus is that IOLs are slow-growing benign tumors[5]. We report an asymptomatic IOL patient with cerebral hemispheric compression involving the left parietal bone.

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CLINICAL AND RESEARCH CONSEQUENCES

In the literature review, 17 patients diagnosed with intraosseous lipoma of the skull were published by different authors[7]. We evaluated the patients' complaints, deformities in the head, radiological examinations performed since 1978, and histopathology as a result of biopsy or surgical excision (Table 1).İn the cases, the age of the disease was between 3-70 years and higher in women than in men. Most of the patients were asymptomatic and diagnosed as coincidental. Also, cosmetic changes were observed from childhood to adulthood or in the postmortem period. Deformities of the frontal and parietal bones were more common. In our case, A 56-year-old female patient presented to our outpatient clinic with complaints of pain in the neck and dizziness. Her systemic and neurological examinations were normal. She was diagnosed incidentally. At first, Yaşuda et al. [8]. (1978) reported a 12-year-old boy that cranial deformity increased from 1 year old to 10 years of age. Although CT and right carotid angiography showed no intracranial lesions, there was thinning of the external tabula with enlarged interdiploic distance in the right frontoparietal bone. Small ML[9]. (1979) presented a 61-year-old male case with the prominent in the right anterior part of the head since the age of 16 years, repeated supraorbital density increase was detected on his radiographs, sudden death from a heart attack in 1976 and post mortem tumor was reported to be an intraosseous lipoma. Milgram, MD[2]. (1987) divided intraosseous lipomas into three according to their pathological

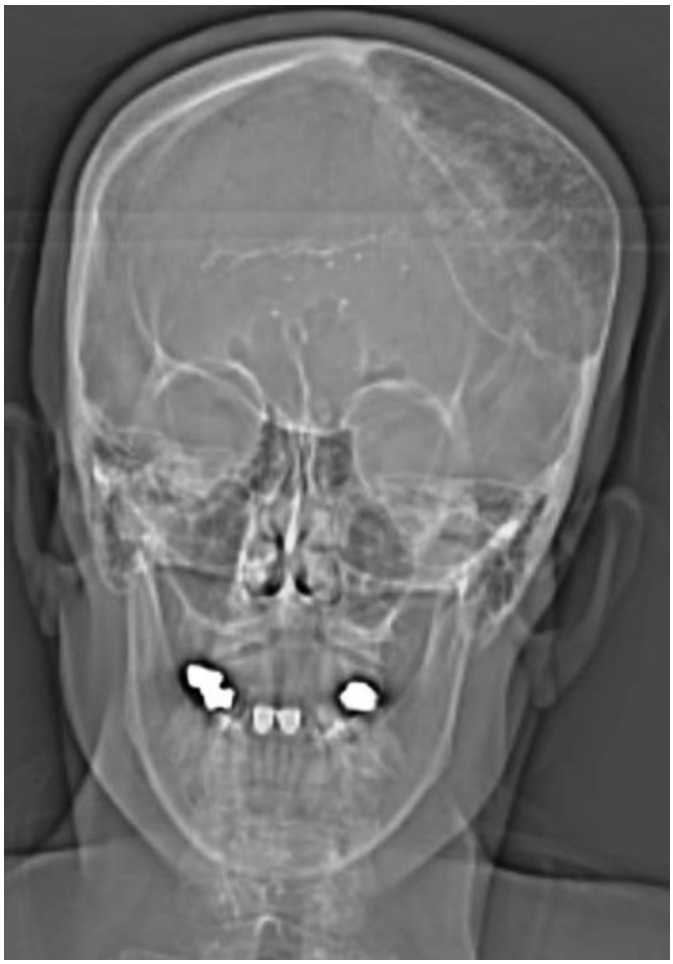

Figure 1: Plain skull x-ray showing dilation in the internal parietal tabula in the left parietal region.

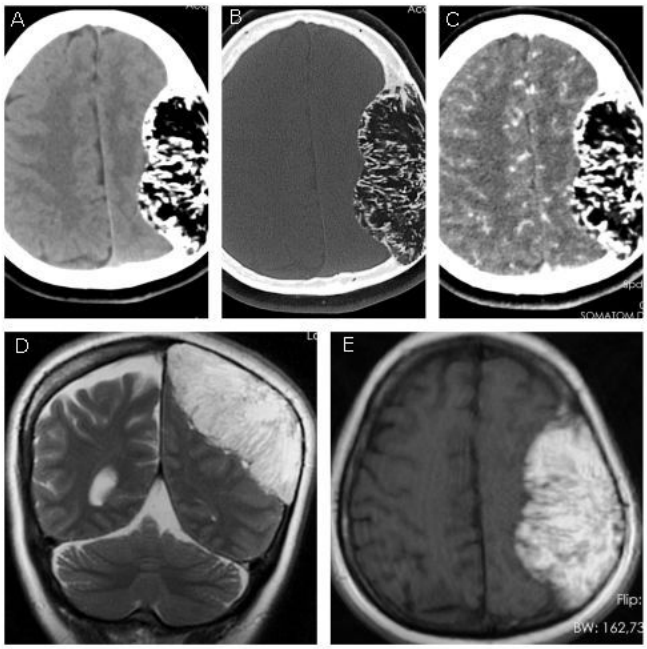

Figure 2: A,B,C,D,E Axial non-contrast CT images (soft tissue and bone window) show well-delineated thinned margin and typical hypodense (-80 HU) intraosseous lipoma in the left parietal bone (A, B). Enhancement is not seen in the lesion on axial contrastenhanced CT image (C). Coronal T2-weighted and axial T1-weighted MR images show typical hyperintensity of the lesion (D,E).

differences. I. solid tumors of living lipocytes, II. Transitional cases with viable lipocytes as well as partial fat necrosis and focal calcification III. late cases with cystic formation, calcification, and new bone formation. He analyzed 66 patients with solitary intraosseous lipoma according to their clinical, radiographic, gross and histopathological findings, reported a lytic lesion in parietal bone in two patients and mandibular lytic lesion in one patient, but did not publish two of these patients. Direct radiography and CT show in stage I, cortical expansion, in stage II, fat necrosis and increased bone density due to calcifying fat, in stage III, the bone is resorbed by osteoclasts, the original trabecular bone is destroyed in the lesion. On CT, there is the presence of radiodensity at both the periphery and center of the lesion is typical in stage III. Cortical expansion may be seen. Many of stage III lesions contain cystic regions.Once Tomabechi et al. (1992) reported intraosseous lipoma of the skull in three cases, then Gobinath et al.(1992) said that there were seven cases.Between 1978 and 2004, there were cases with hypodense and enlarged interdiploic distance at -50 / -150 HU lesion density confirming the intraosseous lipoma in the cases evaluated by CT to assist diagnosis[11]. CT and recently, MRI has been used to investigate the fat in the lesion since it is difficult to diagnose with direct radiographs. Most of the intraosseous lipomas are solitary lesions involving the most common calcaneus and metaphysis of the long bones (especially the femoral neck). These lesions have also been reported in other bones such as skulls, vertebrae, maxilla, pelvis, and ribs[5,10]. Arslan et al. [3]. (2000) reported that only 8 cases had intraosseous lipoma in the skull and in stage III lesions could be recognized with CT that ossification around the calcified necrotic fat, radiodensity and cortical enlargement around the lesion. Twelve of previous cases had a histopathologic diagnosis after biopsy or surgical excision, four patients had refused surgery, one patient had postmortem diagnosis after long-term follow-up, two patients had a recurrence, and two patients had an intracranial invasion.

The lesion in our case was an example of a stage I lesion, with no evidence of calcification on plain films or CT. Since our patient refused biopsy or surgical excision of the lesion, the diagnosis of lipoma was made only by conventional radiography and CT, MRI. With their specificity for adipose tissue, CT, and MR are the best radiologic methods for the diagnosis of lipoma, and they may prevent the need for biopsy. However, MR is less useful in detecting stage III lesions with considerable calcification. Resorption of the original trabecular bone, central and peripheral calcification and cystic regions in stage III lesions can be shown by CT[3].

in our case, plain skull films showed dilatation in the internal parietal tabula in the left parietal region (Figure 1). The cranial computed tomography (CT) revealed a left parietal lesion with -80 Hounsfield Unit, confirming the fatty nature of the tumor. There was an expansile lesion on the left cranium measured at 9x7x4.7 (APxCCxT) cm. No destruction or contrast enhancement was detected in the lesion. The CT angiography revealed normal intracranial vasculature. The lesion occupying the space compressed to the left cerebral hemisphere. There was some compression in the left lateral ventricle and a shift approximately 5 mm to the right. The pressure effect was observed in the left

Table 1 Demographic, radiological and surgical results of patients diagnosed in the literature.

	author year	age/ sex	complaints/ cosmetic deformity		radio-logical findings			surgery	patho-logy	outcome
				X-RAY	CT	MRI	angio			
1	Yasuda Y 1978	12/M	a hard tumor on the right frontoparietal bone	thickness of fron-toparie-tal bone	No intracra-nial invasion	-	No in-tracra-nial inva-sion	total excision	+	no recur-rence
2	1979	20/F	a hard tumor on left frontal bone at birth	thickness of left frontal bone	No intracra-nial invasion	-	No in-tracra-nial inva-sion	total excision	+	no recur-rence
3	Small ML 1979	61/M	The prominence of the right side of forehead	Supra-orbital density on frontal	-	-	-	Postmortem finding	-	exitus
4	Rosembloom SA 1985	32/F	asymptomatic	frontal lytic lesion	low density lesion between outer and inner tables. -40 HU	-	-	excision	+	no recur-rence
5	Milgram JW 1988	21/M	Incidental finding	left parietal lytic lesion	-	-	-	curettage	+	not de-scribed
6		64/M	Incidental finding	large lytic lesion in right parietal	-	-	-	curettage	+	
7		39/M	Incidental finding	Lytic lesion in mandibula	lytic lesion	-	-	curettage	+	not de-scribed
8	Tomabechi M 1992	36/M	intermittent headache	bulging into left frontal bone	thickness of left frontal bone	-	a mass lesion with no stain-ing	excision	+	no recur-rence
9	Kuzeyli K 1995	70/F	incidental finding following subarachnoid hemorrhage	-	left posterior parietal bone 5 mm hy-podense lesion, -50/150 HU	-	-	-	-	follow-up
10	Arslan G 2000	35 F	Incidental finding	Hipodense lesion in right frontal bone	hypodense lesion -62HU	-	-	the patient refused surgery	-	Follow-up

11	**Hayashi Y 2003**	32/F	a subcutaneous tumor in left parietal region, blepharoptosis	Inner thickening of +left frontoparietal bone	remarkable hyperostosis of the left frontoparietal convexity bone	-	fed by the left MMA	a left frontoparietal craniotomy after embolization	+	many meningioma cells had invaded the diploe of the calvarium
12	**Ovalı YG 2004**	48/F	headache	A radiolucent, oval lesion in left parietal bone	İn left parietal bone, a density of -44HU	The bright lesion on T1 and T2 weighted images	-	the patient refused biopsy, surgery		follow-up
13	**Nahles G 2004**	12/F	a hard tumor of the right frontoparietal region in born	-	a hypodense mass lesion with expanded diploic space	an intensified signal to the subcutaneous fat tissue	-	biopsy, excision+ cranioplasty	+	recurrence (+)
14	**Eyzaguirre E 2007**	50/F	Incidental finding	right parietal bone lytic lesion	-	-	-	excision	+	no recurrence
15	**Kaneko Y 2011**	62/F	deformation in the right fronta bone	right frontal bone	with expanded diploic space	to the subcutaneous	-	excision	+	no recurrence
16	**Taheri ST 2012**	43/F	left visual disturbance,headache, hyperprolactinemia	-	Sphenoclival lesion with -50 and -100 HU	A hiperintense lesion within the greater wing of sphenoid and the clivus	-	the patient refused surgery	-	follow-up
17	**Zhe H 2018**	3/M	left frontal deformity	left frontal bone	left frontal plate with oval low density the enlarged bone marrow cavity 78-500 HU	-	-	resection or curettage	+	recurrence (+)

18	**present case 2019**	56/F	Incidental finding	left parietal bone lytic lesion	extensile lesion on the left cranium lesion density -80 HU	a mass lesion originating from the medullary tabular of bone structures	intracranial invasion (+)	the patient refused surgery	-	follow-up

thalamic region (Figure 2. A, B, C).

In magnetic resonance imaging (MRI), a mass lesion originating from the medullary tabular of bone structures was observed at the left frontal-parietal level. The lesion was hyperintense in T1 and T2 weighted imaging. The mass effect of the lesion caused a 5 mm right shift, and lateral compression of the left lateral ventricle was observed. (Figure 2. D, E) The radiological findings were consistent with intraosseous lipoma of the skull. The patient refused surgery.

DISCUSSION

Intraosseous lipomas (IOLs) are rare benign bone tumors, and IOL of the skull is very rare [5,11]. It is localized in the skull in 4% of all IOLs [5]. It is seen higher in men than in women [4,6]. The disease is usually asymptomatic. Clinical symptoms are often local tenderness and pain .The diagnosis of intraosseous lipoma of the skull is difficult because of its non-specific radiological features. The pathological study is essential for a correct diagnosis. The etiology of IOL is not fully known[12]. It can be trauma, bone infarction, primary bone tumor, or diseases of unknown etiology and increased amounts of triglycerides and cholesterol in the body. Fat tissue proliferation and expansion of the medullary cavity may result in trabecular bone structure. Necrosis, central and peripheral calcification and cystic regions occur at the center of the tumor [2,3]. Plain skull films, MRI, CT, and CT angiography are performed for differential diagnosis. MRI helps to reveal the relationship between brain, cranial nerves, and dural sinuses and the location and type of the mass. The honeycomb appearance is suggestive for IOL. Hyperdensity is seen in T1 and T2-weighted sections in MRI [5,13]. The appearance of intratumoral adipose tissue signals provides essential evidence for the diagnosis of IOL. Differential diagnosis includes fibrous dysplasia, osteoblastoma, enchondroma, solitary bone cysts chondrosarcoma, chondroblastoma giant cell tumor, and intraosseous angiolipoma [2,4]. Intraosseous angiolipomas in the skull may mimic angiomas, lipomas, fibrous dysplasia and meningiomas [14]. They are composed of mature adipose tissue ad-mixed with arteries, veins, sinusoids, and capillaries. In our case, a mass showing hemispheric compression in the left parietal area was noted. No vascularization was detected in CT angiography, and the intra-mass density Hounsfield Unit result was -80, which was following the reported IOLs in the literature. IOLs are slowly growing benign tumors; there is no consensus about the indications of surgical treatment. Lesions are treated surgically for cosmetic reasons. In the presence of neurological deficits, dura infiltration, and massive bone destruction, surgical treatment is recommended to confirm the diagnosis. According to the results obtained from imaging in recent years, it is suggested that the excision of an asymptomatic lipoma is not necessary when the diagnosis is definite. The risk of malignant transformation in IOL is very low [2,3]. As seen in Table 1, the surgical treatment of patients diagnosed by radiological or histopathological findings was controversial due to cosmetic reasons, and some patients did not accept surgery. Surgery was refused by our patients. She was followed up with a 3-month periodic neurological examination and MRI. There was no change in imaging and neurological findings at the end of the one-year follow-up.

CONCLUSION

In recent years, contemporary imaging techniques demonstrate the presence of bone invasion and compression of intracranial structures in the early stage.İnfrequently, some patients with signs of invasion or intracranial compression prefer to follow with imaging techniques rather than biopsy or excision.

DISCLOSURE STATEMENTS

There were no financial support or relationships between the authors and any organization or professional bodies that could pose conflict of interests.

COMPETING INTERESTS

Written informed consent has been obtained from the patient for publication of this case report and any accompanying images.

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