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Case Report

Congenital angiolipoma of the chest wall in a child: A case report and review of literature

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ABSTRACT

Angiolipoma is the most common soft tissue tumor in human trunk and extremities, mostly affecting young male patients; however, in children, it occurs rarely and the most involved area has not been determined. A 9 year old girl was admitted to the pediatrics ward because of a newly arousal of pain associated with suddenly rapid increasing of the size of an old mass on the right side of her chest wall for almost one month without any history of trauma to the area; she had the lesion since her birthday. Otherwise, her physical examination and laboratory tests were unremarkable. Findings of different types of imaging of the mass were in favor of a benign chest wall lesion. A surgical excision was performed and histopathologic report of the lesion was consistent with typical features of a non-infiltrating angiolipoma. The patient recovered uneventfully with complete resolution of lesion. No recurrence of the tumor was observed in the patient in the following 30 months. This report seems to be the first documented case of congenital non-infiltrating angiolipoma of the chest wall in a child. Review of literature has shown that in children, angiolipoma is mostly a single, benign lesion commonly appears in the face or the spinal region, with complete recovery after a simple surgical excision and no recurrence.

Keywords: Angiolipoma, non-infiltrating, child, chest wall, review.

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INTRODUCTION

Angiolipoma is a rare type of soft tissue tumors presents as a vascular variant of lipoma and represents approximately 5 to 17% of all lipomas in humans (Jo and Fletcher, 2013; Choong, 2004; Bang et al., 2012). It is composed of mature adipocytes with prominent vascular supply including small vessels and capillaries which characteristically contain fibrin thrombi (Kransdorf et al., 1991; Gupta et al., 2016). Angiolipomas are slowly growing benign subcutaneous lesions, most commonly affecting young male patients in the second and third decades of life (Bang et al., 2012). It is the most frequent tumor in the trunk and the extremities of young people (Lee et al., 2011); however, its pathogenesis remains unclear. Generally, these masses can appear in the skin or deeply seated within underneath soft tissues (Murphey et al., 2004). In spite of primary clinical diagnosis of the mostly superficial mass lesions, the diagnosis of deep seated soft tissue tumors often requires performing imaging methods in the patient. Ultra-sonography is the choice method for diagnosing small and superficial lesions (Choong, 2004; Bang et al., 2012; Sheybani et al., 2016; Shin et al., 2016), whereas, Magnetic Resonance Imaging (MRI) is the best modality for diagnosing of the larger and deeper masses (Jo and Fletcher, 2013; Murphey et al., 2004; Sheybani et al., 2016; Navarro, 2009; Navarro et al., 2009). Besides, it is the most sensitive imaging procedure for excluding the invasion of the surrounding structures by the mass (Matsuoka et al., 1988). Ultimately, in most of cases, a definite diagnosis is made by histopathologic section of the lesion (Sheybani et al., 2016; Navarro, 2009). Despite, there is a broad spectrum of benign soft tissue tumors presented in children, including vascular and/or adipocytic lesions, just a few reports of angiolipoma in children mentioned in the literature; nevertheless, congenital variants have rarely reported in the articles. Angiolipoma may present as painless soft tissue masses or according to its size and location may compress

nearby structures. This tumor has been barely reported in the chest wall, and as we searched the literature, it sounds that it has not been reported as a congenital lesion in a child's chest wall. The author presents the case and reviews childhood angiolipomas reported in the literature.

CASE PRESENTATION

A 9 year old girl presented to our clinic with a mass on the right side of her chest wall. She complained of newly arousal of pain in the lesion with suddenly rapid increasing of its size since almost one month earlier. The pain was not permanent and had no referral to anywhere. Her parents mentioned that the mass has existed since her birthday as a greenish almost 2*2 centimeter size lump with gradually slow growth over time, but with no previous history of any pain or tenderness. Every history of recent trauma to the mass or surrounding area was denied. Other personal or family history of the case was unremarkable. On physical examination, there was a relatively firm well-circumscribed 5*7 centimeter smooth protuberance with tenderness associated with greenish color changes on some parts of overlying skin, located at the right anterior part of the chest wall at the mid-axillary line direction (Figure 1A and B).

No adhesion to the surrounding or underneath tissues as well as overlying skin was found. There was no evidence of bruising or bleeding near the mass or in other parts of the body. No evidence of hepatosplenomegaly or lymphadenopathy was detected on the physical examination. Other aspects of physical examination were normal. All the laboratory tests including Protrombin Time (PT) and Partial Thromboplastin Time (PTT) as well as International Normalizing Ratio (INR) were within normal ranges. The report of superficial ultra-sonography of soft tissue has revealed a hyper echoic heterogenous mass with 50*40*15 millimeter in dimension included cystic and tubular areas in favor of dilated vessels. Moreover, no invasion of the mass to pectorals muscles or underneath ribs was seen. Furthermore, no pathologic lymph node in axillary or supra clavicular region was detected. Color Doppler surveys showed no vascular markings. These findings recommended a benign chest wall lesion including vascular mass with low flow veno lymphatic malformation. A spiral computed tomography scan (CT-scan) of lungs and mediastinum with and without contrast in bone window has ordered and has shown an image of soft tissue mass in the right lateral side of the chest wall which was over pectorals and serratus anterior muscles with almost 47*30*18 millimeter in dimension. There was no invasion to deep internal organs as well as no obviously enhancement in post contrast images. These features were in support of the ultra-Sonographic findings and brought up a benign chest wall lesion. A Magnetic Resonance Imaging (MRI) with and without contrast was performed. In T1-weighted sections without contrast, low signal tubular structures were observed in subcutaneous fatty tissues with significant enhancement in contrast images (Figure 2A and B).

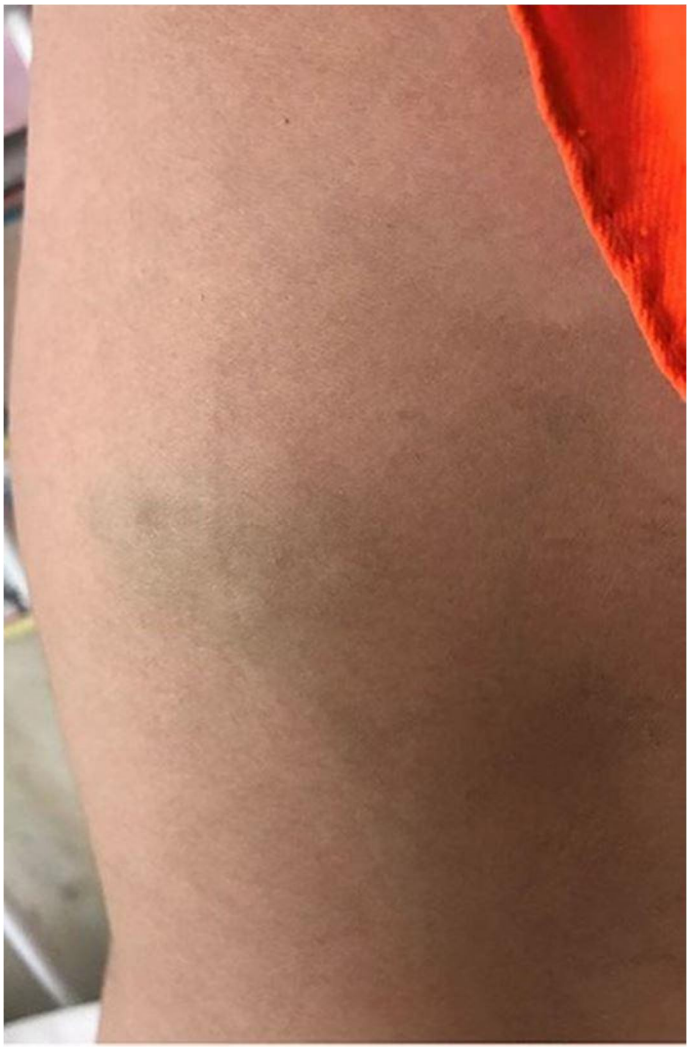

Figure 1A. The mass lesion from anterior view.

T2-weighted sections without contrast revealed fluid signals inside the high signal tubular structures with no significant enhancement in contrast images (Figure 2C and D).

These findings supported the diagnosis of dilated vessels too. Therefore, a decision was made in order to remove the mass by surgical excision. During the operation, it was revealed that the mass was placed under the sub-dermal fatty tissue, over pectoralis major muscle with some involvement of the muscle fascia, but no invasion to underneath muscles, nerves or vessels. It grossly appeared without a capsule or any vascularity. The mass was resected on bloc and has sent for biopsy. In histopathology report of the lesion, the specimen was well-circumscribed and unencapsulated, contained subcutaneous fatty tissue with 7.5*4.5*2.5 centimeter in dimension. Its external surface was unremarkable.

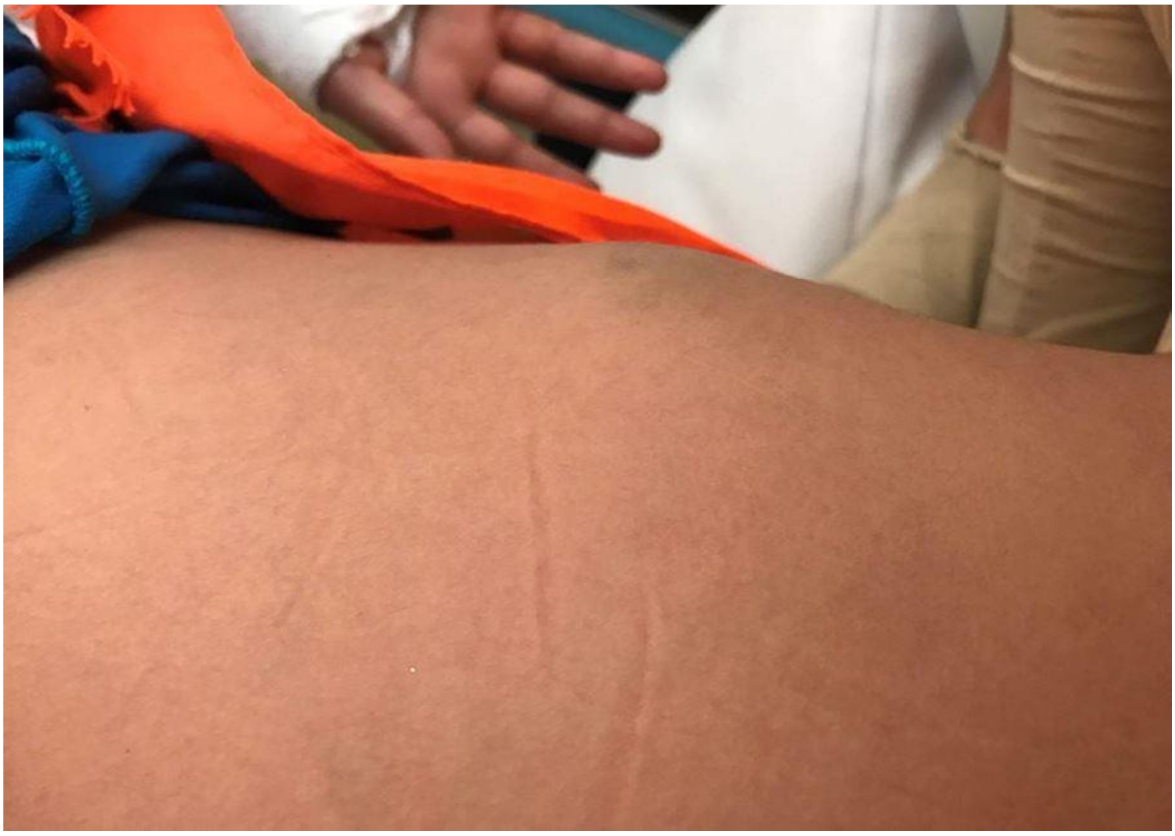

Figure 1B. The mass lesion in left lateral decubitus view.

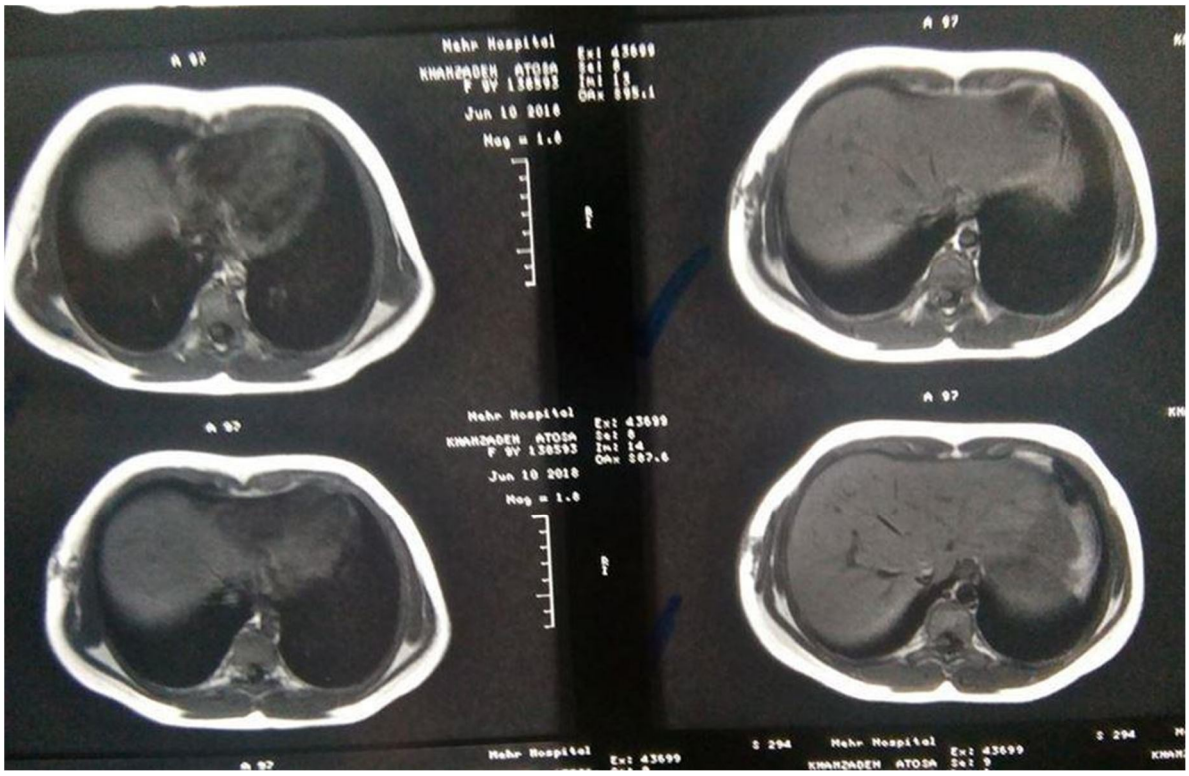

Figure 2A. MRI of the chest. T1- weighted, without contrast and without fat suppression.

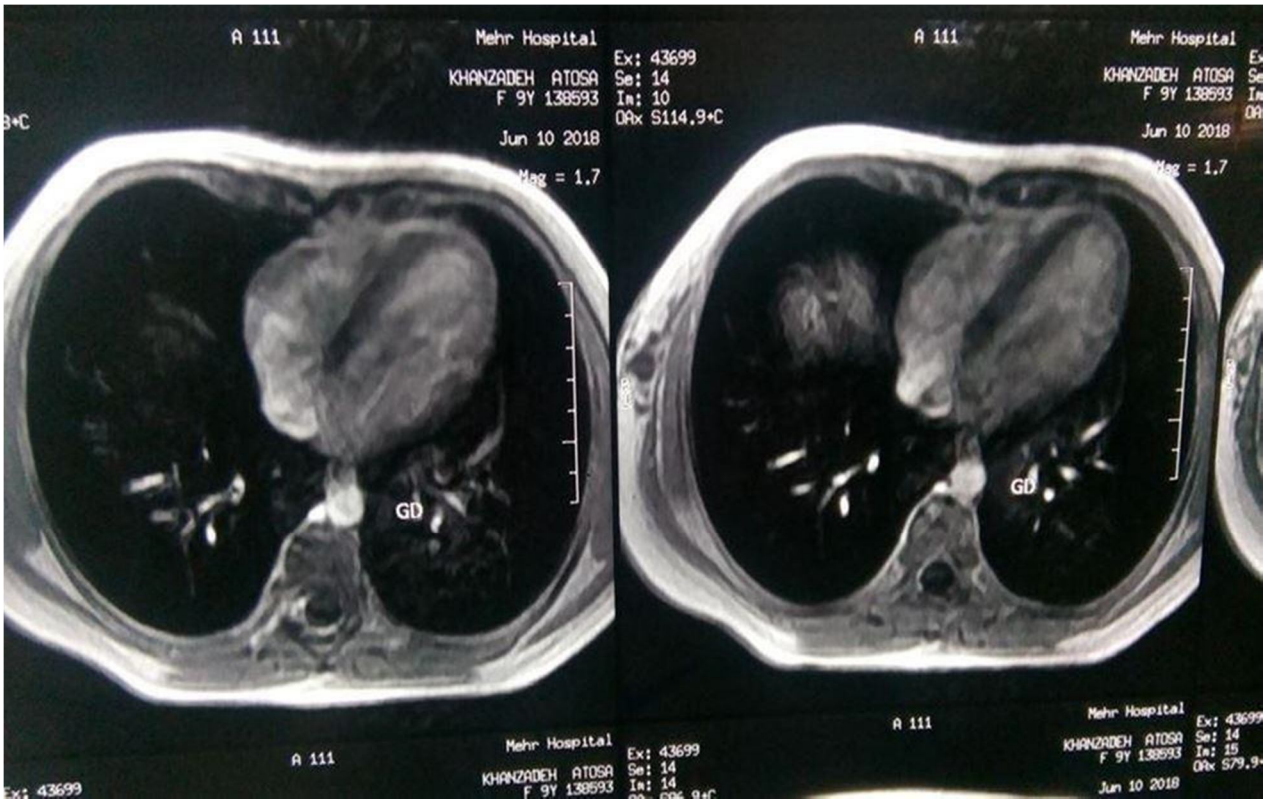

Figure 2B. MRI of the chest. T1- weighted, with contrast and without fat suppression.

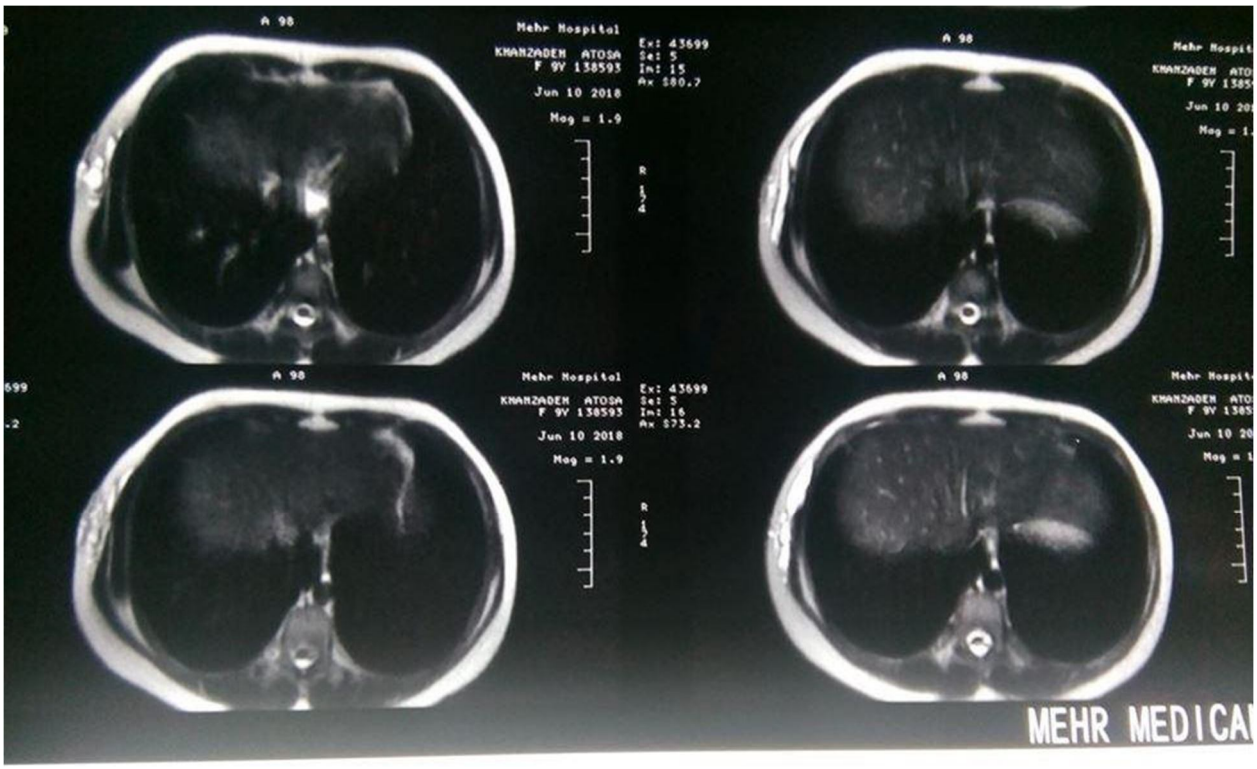

Figure 2C. MRI of the chest. T2-weighted, without contrast and without fat suppression.

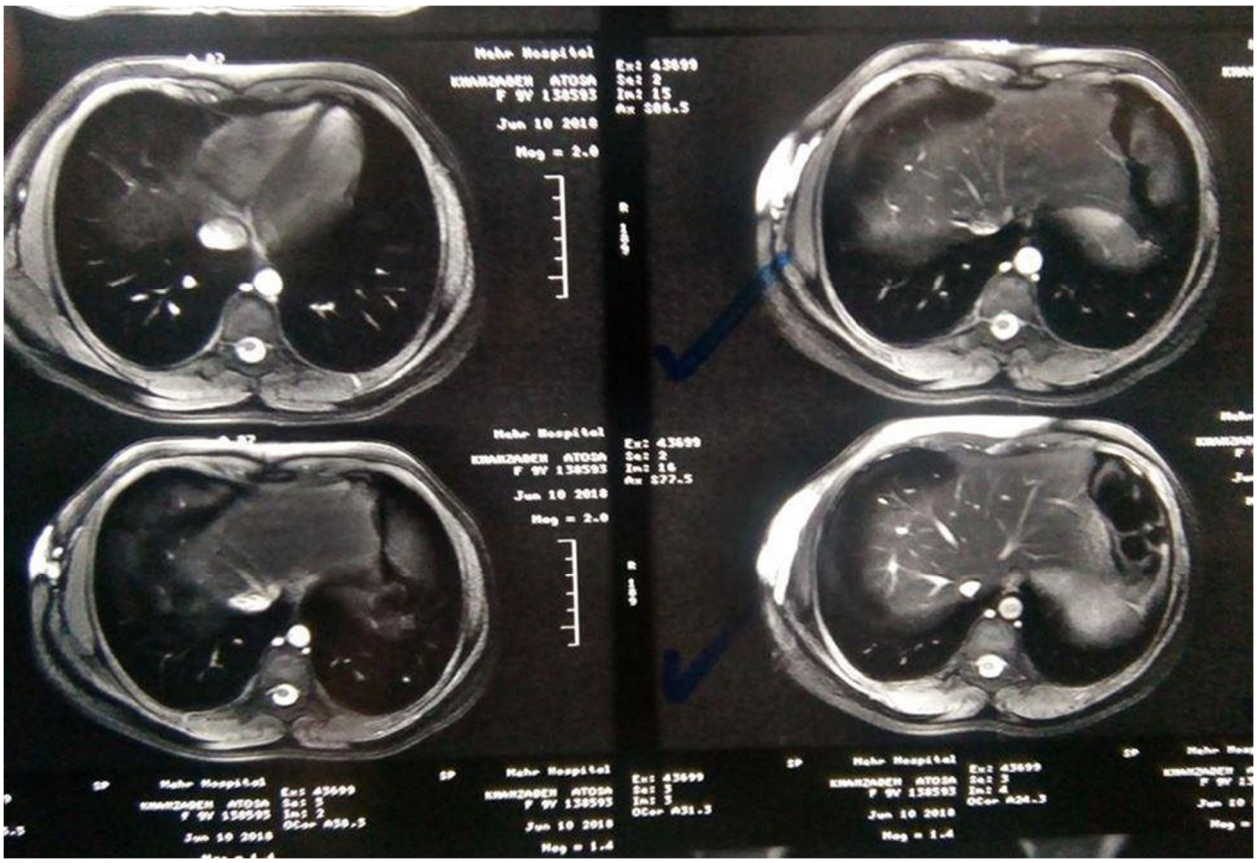

Figure 2D. MRI of the chest.T2-weighted, with contrast and with fat suppression.

The consistency of the mass lesion was rubbery and areas of cystic formation, brownish discoloration and myxoid changes were identified, which distance from nearest margin was 0.3 centimeter. The lesion was entirely composed of mature adipocytes arranged in irregular lobes interspersed throughout vascular spaces circumscribed with thin fibrose septa and areas of abundant small sized blood vessels that were more prominent at the peripheral localization. In addition, fibrin thrombi were noted within occasional vascular lumens (Figure 3A and B).

These findings are consistent with typical features of a non-infiltrating angiolipoma. After operation, the surgeon prescribed an elastic bandage over the area of removed lesion in order to prevent fluid accumulation. In a follow up visit, after one month, there was some mass formation on the lesion removal site with cyst consistency which was the result of no applying of the bandage by the patient. A MRI was ordered again and the images have revealed a cystic collection of fluid in favor of seroma. The cyst lesion was aspirated and the bandage was applied. At the next follow up visit performed two weeks later, the site of operation was completely closed, flatted and cleared. The patient recovered uneventfully with complete resolution of pain. No recurrence of tumor was observed in the following 30 months.

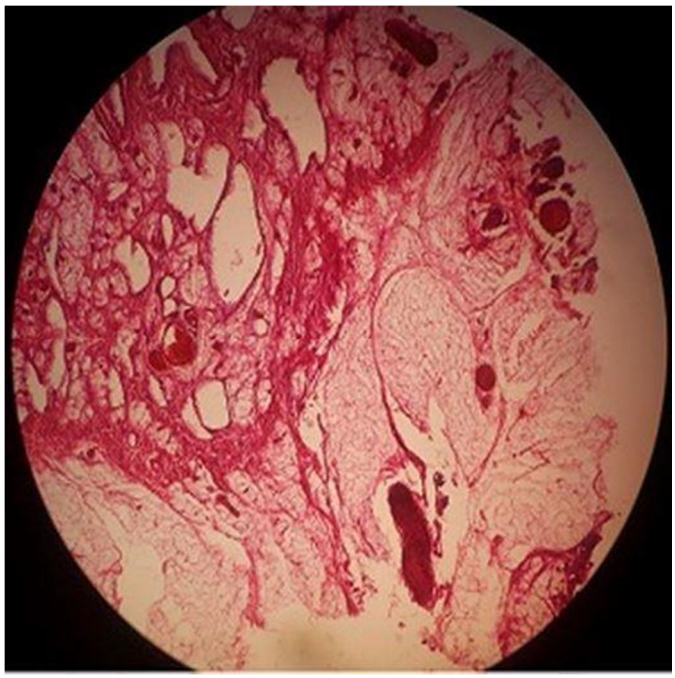

Figure 3A. Photo of biopsy section of the mass lesion. Based on the histomorphologic findings, fibrin thrombi were noted in some vessels and the sections showing adipose tumor containing proliferation of capillary vessels.

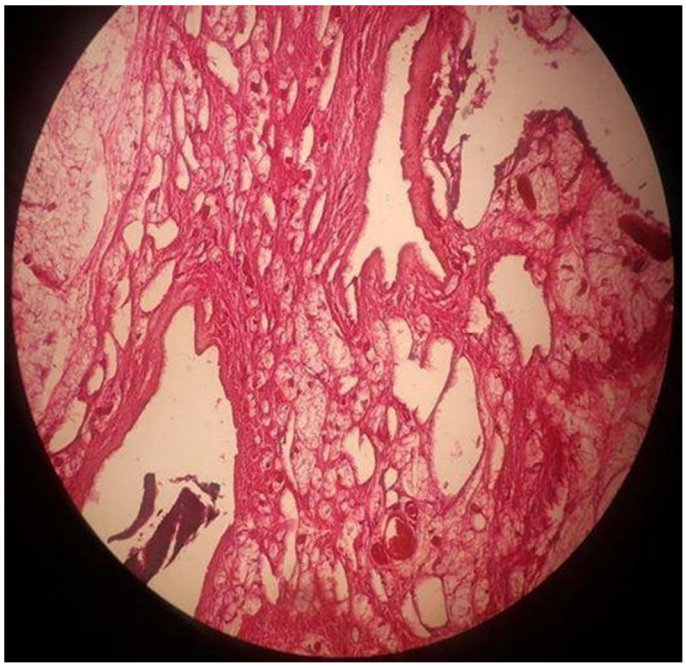

Figure 3B. Photo of biopsy section of the mass lesion. Mature adipocytes arranged in irregular lobes interspersed throughout vascular spaces as well as fibrin thrombi were noted in some vessels.

DISCUSSION

Angiolipomas are benign acquired subcutaneous mesenchimal tumors most commonly affected young males. This article introduces a case of congenital non-infiltrating angiolipoma of the chest wall in a 9 year old girl presented to our clinic with newly arousal of pain and tenderness in an old mass as well as rapidly enlargement of its size for almost one month with seemingly no history of trauma. The tumor has appeared in the chest wall since her birthday and had a slow growth over time with no history of pain or tenderness. As we searched the literature, there were reported a few cases (50 cases) of angiolipoma in childhood (Table 1).

Generally, twenty five cases (52%) appeared in males versus twenty three cases (48%) in females. The sex of two cases not determined because of no access to the two non-English articles' full text. Twenty six cases (53%) occurred before 11 years of age and twenty four cases (47%) occurred from 11 till 20 years of age in children. According to the location, the frequency of lesions has occurred more than one time was as follows: 18 cases in the facial region (37.5%), 16 cases in spinal regions (33%), 4 cases in knee, 2 cases in neck, 2 cases in chest wall, 2 cases in back or lower back, 2 cases in ankle, 2 cases in orbit and 2 cases in forearm. In each of the following location just 1 case has been reported: lower extremity, thigh, foot, back of the tongue, frontal bone, mandible, upper lip, nose, eyelid, hard palate, and parotid as well as pectoralis major muscle. In the face, cheek was the most common area for tumor presentation (5 cases, 30%). Therefore, it sounds that the pattern of tumor involvement in children is different from adults in whom most of the cases appeared on the trunk or

Table 1. Summary of Reported Angiolipomas in childhood in the literature.

Author	Age	Sex	Location	Histopathology type	Additional finding
Caucci (1959)	newborn	?	Forearm	?	Congenital
Cvetinović et al. (1988)	5 month	?	Neck	?	
Reilly et al. (1988)	6 month	Female	Parotid	Noninfiltrating	
Shahi et al. (2014)	9 month	Female	Buccal mucosa	Infiltrating	
Sandvik et al. (2015)	1 year	Male	Spinal	Noninfiltrating	
Takahashi et al. (1998)	1 year	Female	Tongue	Noninfiltrating	
Jaiswal et al. (2020)	1.5 year	Female	Spinal epidural	Noninfiltrating	
Maier (1962)	1.5 year	Female	Spinal epidural	Infiltrating	
Stimpson (1971)	2 year	Female	Pectoralis major muscle	Infiltrating	
Feinfield et al. (1988)	3 year	Male	Eyelid	Noninfiltrating	
Carruth and Meyer (2015)	3 year	Male	Orbit	Noninfiltrating	
Gonzalcz-Crussi et al. (1996)	3 year	Male	Knee	Infiltrating	
Weitzner and Moynihan (1978)	4 year	Male	Cheek	Noninfiltrating	
Gelabert-González et al. (2002)	4 year	Male	Thoracic spinal	Noninfiltrating	
Turgut (2004)	4 year	Male	Thoracic spinal	Noninfiltrating	
Arenaz-Búa et al., (2010)	5 year	Male	Cheek	Noninfiltrating	
Kasper and Cowan (1931)	6 year	Male	Spinal epidural	Noninfiltrating	
Chew et al. (1980)	6 year	Male	Knee	Infiltrating	
Gonzalcz-Crussi et al. (1996)	6.5 year	Female	Knee	Infiltrating	
Koopmann (1988)	7 year	Male	Nose	Noninfiltrating	

Table 1. Continues.

Aljerian et al. (2019)	7 year	Female	Orbit	Noninfiltrating	
Yeo et al. (2018)	7 year	Male	Foot	Infiltrating	
Regan et al. (1946)	8 year	Male	Lower extremity	Infiltrating	Multiple
Flaggert et al. (1986)	8 year	Female	Hard palate	Noninfiltrating	
Sah et al. (2012)	9 year	Female	Upper lip	Noninfiltrating	Congenital
Alvi et al. (1998)	10 year	Female	Cheek	Noninfiltrating	
Aniceto et al. (1990)	11 year	Male	Cheek	Noninfiltrating	
Chew et al. (1980)	11 year	Female	Ankle	Infiltrating	
Michilli et al. (1993)	12 year	Male	Spinal extradural	Noninfiltrating	
Akhaddar et al. (2000)	12 year	Female	Spinal epidural	Noninfiltrating	
Ali and El-Zuebi (1996)	13 year	Female	Cheek	Noninfiltrating	
Shetty and Prabhu (2009)	13 year	Female	Face	Noninfiltrating	
İlyas et al. (2016)	13 year	Male	Knee	Noninfiltrating	
Matsuoka et al. (1988)	14 year	Female	Neck	Infiltrating	
Raghavendra et al. (2007)	14 year	Male	Spinal	Noninfiltrating	
Fernández et al. (1994)	14 year	Female	Spinal epidural	Noninfiltrating	
Kumar et al. (2013)	15 year	Male	Forearm & back	Noninfiltrating	Multiple
Shetty and Prabhu (2009)	16 year	Male	Mandible	Noninfiltrating	
Gelabert-González and García-Allu (2009)	16 year	Male	Spinal extradural	Noninfiltrating	
Lo Re and Michelacci, 1969	16 year	Male	Spinal epidural	Noninfiltrating	Congenital
Petrella et al. (2005)	16 year	Male	Spinal epidural	Noninfiltrating	
Atilgan et al. (2014)	16 year	Female	Frontal bone	Infiltrating	
Pearson et al. (1970)	17 year	Female	Spinal extradural	Infiltrating	
Lacour et al. (2018)	17 year	Male	Thoracic spinal epidural	Noninfiltrating	
Labram et al. (1999)	17 year	Male	Spinal epidural	Noninfiltrating	
Vijay et al. (2015)	17 year	Female	Ankle	Infiltrating	
Komatsu et al. (2013)	18 year	Male	Chest wall	Infiltrating	
Deviri et al. (1987)	18 year	Female	Chest wall & lower back	Noninfiltrating	Multiple
Shetty and Prabhu (2009)	19 year	Female	Face	Noninfiltrating	
Chew et al. (1980)	19 year	Female	Thigh	Infiltrating	

extremities (Lee et al., 2011; Lin and Lin, 1974; Howard and Helwig, 1960). It sounds that this is the first report of a chest-wall non-infiltrating angiolipoma in a child presented since birthday. According to other studies, just three cases have appeared congenitally in childhood (Caucci, 1959; Sah et al., 2012; Lo Re and Michelacci, 1969). Therefore, almost all the reported cases in the literature were acquired (47 cases, 94%). Furthermore, despite multiple lesions are frequently appeared in adults (Lin and Lin, 1974), in children occurrence of multiple masses are rare; only 3 cases include an eight year old boy with two huge lesions in lower extremity (Regan et al., 1946), a fifteen year old boy with multiple painful skin lesions over forearm and back (Kumar et al., 2013) and an eighteen year old girl with two lesions of chest wall and lower back (Komatsu et al., 2013) had been reported. It is interesting that, as we could find, there were just four case reports of chest wall angiolipoma in adults mentioned in the literature; all of them were acquired; 75% of the cases appeared in young adult patients. The cases included a tumor in a 25 year old male (Hamano et al., 2013), a postero-lateral thoraco-abdominal wall mass in a 41 year old male (Biondetti et al., 1982), a tumor in a 42 year old female (Mayooran et al., 2016) and a tumor in right chest wall in a 68 year old female (Sakamoto et al, 2019). All of the cases except one case (Sakamoto et al, 2019) had definite diagnosis of infiltrating angiolipoma and half of them were described as asymptomatic (Mayooran et al., 2016; Sakamoto et al, 2019). Contrary to the type of reported cases in adults' chest wall, present case was of non-infiltrating type and has appeared as a congenital lesion.

According to the literature, in adulthood these lesions are seen more commonly in a younger age group; generally in pubescent patients and are rare before puberty. They present as painless or tender subcutaneous nodules and have no tendency to recurrence (Arenaz-Búa et al., 2010; Howard and Helwig, 1960); however, infiltrating angiolipomas usually appear in patients over 30 years of age, and there is a recurrence rate of 50%. Besides, infiltrating angiolipomas have not been found to undergo malignant transformation

(Arenaz-Búa et al., 2010; Lin and Lin, 1974; Howard and Helwig, 1960). In the review of literature of children with angiolipoma, just 15 cases (30%) were of infiltrative type and most of the reported cases (33 cases, 59%) like our case had non-infiltrating histopathology. It is noteworthy that similar to adults, the treatment of an angiolipoma in children is surgical excision (Arenaz-Búa et al., 2010; Lin and Lin, 1974; Howard and Helwig, 1960) which ranges from simple excision in non-infiltrating cases to wide surgical excision in infiltrating angiolipoma. Furthermore, the studies have shown that non-infiltrating cases have no tendency to recur (Arenaz-Búa et al., 2010; Lin and Lin, 1974; Howard and Helwig, 1960). Like to reported cases in the literature, a simple surgical excision of the lesion has performed on our case; identical to other reported children, she hadn't experienced any recurrence on follow up visits.

CONCLUSION

In conclusion, angiolipomas are rarely seen in the chest wall region of the children, especially as a congenital mass. The presented case showed the typical clinical, imaging and histological findings of a non-infiltrating angiolipoma. Furthermore, its behavior regards to natural history, response to surgical excision and recurrence is identical to other non-infiltrating tumors. The more striking features in this case are its existence since the birthday as a single lesion, the uncommon location as well as the unusual way of its presentation after a long period of its asymptomatic existence with slow growth. Furthermore, review of literature has shown that children with angiolipoma mostly presented with single acquired benign lesion which appears most commonly in the face or spinal region. The lesions usually have non-infiltrating histopathology and recovered uneventfully with simple surgical excision without occurring of any recurrence.

Patient consent for publication

Written informed consent for publication of the case report and any accompanying images, without any potential identifying information, was provided by the parents of the patient.

Conflicts of interest

The author declares no conflict of interest.

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