J Ped Surg Case Reports 3 (2015) 316–319

Contents lists available at ScienceDirect

Journal of Pediatric Surgery CASE REPORTS

journal homepage: www.jpscasereports.com

Supernumerary ovary presenting as a paraduodenal duplication cyst

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ARTICLE INFO

Article history:
Received 9 April 2015
Received in revised form
5 May 2015
Accepted 9 May 2015

Key words:
Supernumerary ovary
Accessory ovary
Duplication cyst

ABSTRACT

Supernumerary ovary is a rare gynecological anomaly and is generally excised due to its potential malignant transformation. We report a case of a patient who was referred for excision of a probable duodenal duplication cyst that was subsequently identified as a paraduodenal supernumerary ovary. Pediatric surgery was consulted on an adolescent patient due to a presumed congenital anomaly of the intestinal tract based on imaging studies.

Ectopic ovaries, whether accessory or supernumerary, are an extremely rare gynecological anomaly. Winckel et al. [1] reported the first case of supernumerary ovary in 1890. Since then about 44 cases have been reported in the literature. The exact incidence is unknown with Wharton et al. [2] reporting one accessory ovary case in 93,000 gynecological admissions and finding only one supernumerary ovary case in 29,000 autopsies performed. Patients are usually asymptomatic, but may occasionally present with chronic abdominopelvic pain, as was the case with the current case report. The pain presumably arises as a result of cystic or neoplastic transformation.

1. Case report

A 20 year old female presented to the emergency department (ED) with a two day history of epigastric and left upper quadrant abdominal pain. Patient described the pain as non-radiating constant pain that is associated with nausea and non-bloody diarrhea. There were no associated fevers and patient was otherwise hemodynamically stable. Patient's last menstrual period was 2 weeks prior to presenting to the ED. Lab work did not reveal any evidence of leukocytosis or pancreatitis. Abdominal exam was unremarkable apart from mild tenderness with no guarding in the epigastric region. An abdominal ultrasound was obtained in the ED, which revealed an avascular cystic structure in the region of the tail of the pancreas (Fig. 1). For better characterization of the cystic structure, an abdominal computed tomography was obtained (Fig. 2).

In view of the patient having persistent abdominal pain, associated with an abnormal mass, a decision was made to surgically excise the retroperitoneal cyst. A transverse left upper quadrant was made and the mass was found to be palpable through the transverse mesocolon. A Mattox maneuver was then performed to reflect the colon to the right. The cystic structure was then visualized in the retroperitoneal location adherent to the duodenum, but

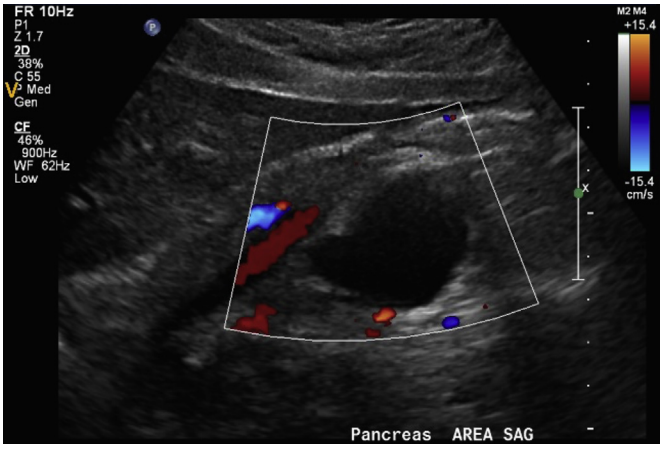

Fig. 1. Ultrasound image revealing a 3.4 × 2.8 × 2.6 cm avascular cystic structure in the pancreatic tail region.

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http://dx.doi.org/10.1016/j.epsc.2015.05.001

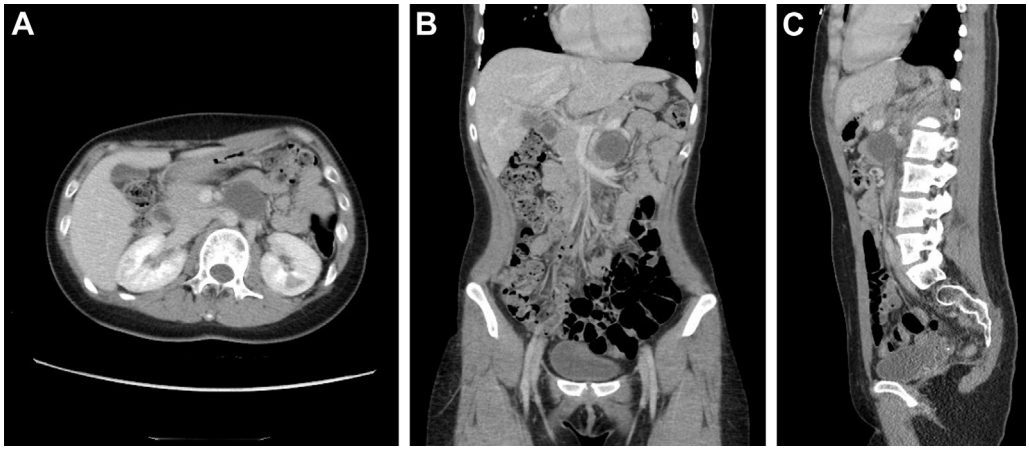

Fig. 2. CT axial (A), coronal (B) and sagittal (C) images revealed a 3.5 cm probable duplication cyst abutting the posterior wall of the third segment of the duodenum. No evidence of bowel obstruction or bowel wall thickening.

Table 1
Published cases of supernumerary ovary.

Case	Author(s)	Year	Age (years)	Location	Neoplasm/Histopathology
1	Winckel [1]	1890	77	Anterior uterine wall	N
2	Falk [17]	1891	37	Omentum	N
3a	Kriss [18]	1947	49	Infront of uterus to the right	Y/Granulosa cell carcinoma
3b	Kriss [18]	1947	34	Lateral to the right ovary	Y/Serous cystadenoma
4	Nicols and Postoloff et al. [19]	1951	36	Area of congenitally absent right kidney	N
5a	Wharton [2]	1956	37	Retroperintoneal right pelvic wall	N
5b	Wharton [2]	1957	39	Retroperitoneal sigmoid mesentery	N
6	Wharton [2]	1959	20	Left of aorta	N
7	Burnett [20]	1961	–	Retroperitoneal between left ureter and rectosigmoid	N
8	Pearl and Plotz [13]	1963	34	Inferolateral to left uterosacral ligament	N
9	Hogan et al. [21]	1967	21	Omentum	Y/Cystic teratoma
10	Williams et al. [22]	1971	26	Retroperitoneal in right iliac fossa	N
11	Printz et al. [14]	1973	23	Omentum	Y/Cystic teratoma
12	Arzapalo et al. [23]	1974	19	Left adnexal area	N
13	Huhn [24]	1975	–	Omentum	Y/Cystic teratoma
14	Abrego [25]	1975	38	Distal ileal mesentery	N
15	Kosasa et al. [26]	1976	31	Retroperitoneal left pelvic side wall	N
16	Roth and Ehrlich [27]	1977	48	Left retroperitoneal space	Y/Papillary Mucinous cystadenocarcinoma
17	Cruikshank and Van Drie [28]	1982	33	Pelvic mass attached to descending colon	N
18	Cruikshank and Van Drie [28]	1982	36	Left retroperitoneal, para-renal area	Y/Mucinous cystadenoma
19	Poma [29]	1982	50	Left infundibulopelvic ligament	N
20	Lee and Gore [30]	1984	30	Attached between left tubal fimbria and rectosigmoid	N
21	Hahn-Pedersen and Munkholm [31]	1984	20	Connected by fibrous ring to left ovary	N
22	Mercer et al. [32]	1987	36	Broad ligament, 2 cm caudad to right ovary	N
23	Mercer et al. [32]	1987	34	Omentum	Y/Cystic teratoma
24	Harlass et al. [33]	1987	37	Anterior descending colon	N
25	Navarro et al. [34]	1990	38	Left paracolic gutter	N
26	Alpern [35]	1990	40	Retroperitoneal right pelvic side wall	N
27	Cruikshank [36]	1991	37	Retroperitoneal left pelvic side wall	N
28	Barik et al. [37]	1991	–	–	Y/Adenocarcinoma
29	Besser and Posey [38]	1992	47	Omentum	Y/Cystic teratoma
30	McCullough [39]	1992	27	Serosal surface at junction of uterus and cervix	N
31	Badawy et al. [40]	1995	32	Left pelvis side wall over liopsoas muscle	N
32	Levy et al. [41]	1997	51	Right upper renal pole	N
33	Kini et al. [42]	1998	5	Left upper renal pole	N
34a	Kuga et al. [15]	1999	1/12	Omentum	N
34b	Kuga et al. [15]	1999	1/12	Omentum	N
35a	Kamiyama et al. [43]	2001	47	Omentum	Y/Fibroma
36b	Kamiyama et al. [43]	2001	28	Anterior wall of uterus	N
37	Litos et al. [44]	2003	32	Retroperitoneal medial aspect of descending colon anterolateral psoas muscle	N
38	Sonntag et al. [45]	2005	30	Retroperitoneal, caudal pole left kidney	Y/Mucinous adenocarinoma
39	Hartigan et al. [16]	2006	32	Right intrarenal upper pole	N
40	Imir et al. [46]	2006	30	Sigmoid colon	N
41	Zhigang and Wenlu [47]	2007	43	Intrarenal upper pole right	N
42	Matsubara et al. [48]	2009	31	Mesentery of rectum	N
43	Nomelini et al. [49]	2013	64	Retrouterine	Y/Serous papillary cystadenocarcinoma
44	Bae et al. [12]	2013	31	Retroperitoneal left pelvic side wall	Y/Serous papillary carcinoma

did not appear to be a duplication cyst. Pathology revealed benign ovarian parenchyma, with hemorrhagic corpus luteum and cystic follicles consistent with a supernumerary ovary.

2. Discussion

There are numerous examples of congenital anomalies presenting beyond the adolescent period such as malrotation [3], foregut duplication cysts [4], Morgagni hernia [5,6], choledochal cyst, and Hirschsprung's disease [7]. Adult surgeons infrequently encounter these congenital anomalies. This sometimes necessitates the expertise of pediatric surgeons, who are more accustomed to these anomalies compared with adult surgeons. Infantile hypertrophic pyloric stenosis (IHPS), which is the most common surgical condition causing emesis in infancy, is an example of a pediatric surgical condition that has been reported to be operated on by adult surgeons in peripheral hospitals [8,9]. However, it has been demonstrated that when IHPS is operated on in a specialized tertiary center, that the infection rate and hospital stay are significantly less [10].

Here we report a case of a patient found to have a supernumerary ovary resembling a paraduodenal duplication cyst. Supernumerary and accessory ovaries are an extremely rare gynecological anomaly [11]. Although rare, they need to be excised due to their malignant potential [12]. The term 'supernumerary ovary' is defined as a third ovary that is entirely separate from the normally placed ovaries. Whereas the term 'accessory ovary' is defined as excess ovarian tissue that is invariably situated near the normally placed ovary, which may or may not be connected to it [12]. Histological diagnosis of an accessory or supernumerary ovary requires demonstration of ovarian follicles. Imaging studies of the cystic structure around the third portion of the duodenum were interpreted in this 20 year old patient as most probably a duodenal duplication cyst. Pediatric surgery was consulted for excision for a presumed intestinal congenital malformation.

There have been two theories that have been postulated with regards to the development of the supernumerary ovary. Firstly, the "arrested gonocyte migration" theory, where the gonocyte's migration arrests as they migrate retroperitoneally through the dorsal mesentery [13]. Secondly, the "transplantation" theory of the germinal ridge following incorporation of the gonocyte [14]. From these proposed mechanisms, supernumerary ovaries tend to be located retroperitoneally as is the case with our current case report. Other reported locations include the pelvis, para-aortic, omentum, intrarenal, bladder and colonic mesentery [15,16]. Supernumerary ovaries are often incidentally found, and thus underreported, however they may also be painful, functional, multiple or associated with other congenital malformations of the genitourinary system [12]. Table 1 lists the previously reported case reports on supernumerary ovaries along with their location and histopathological description, whether benign or malignant.

3. Conclusion

Although this is a rare gynecological anomaly, we recommend complete excision of these cysts for accurate pathologic diagnosis and to eliminate risk of malignant transformation (See Table 1).

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