

· 临床研究 ·

胰腺内副脾误诊为神经内分泌肿瘤病例特点及误诊分析

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【摘要】目的 探讨胰腺内副脾患者的病例特点及误诊原因, 以提高对该病的认识, 减少误诊及不必要的手术。**方法** 回顾性分析 2012 年 1 月至 2022 年 8 月于中国人民解放军总医院第一医学中心肝胆外科及第二医学中心综合外科就诊、术前诊断为胰腺神经内分泌肿瘤、经术后病理确诊为胰腺内副脾的 10 例患者(共 12 个病灶)的病例资料, 分析其 CT、MRI、⁶⁸Ga 标记正电子发射计算机断层显像(⁶⁸Ga-dotatate PET/CT)图像与病理特点等。**结果** 10 例患者中 9 例为单发, 1 例有 3 个病灶; 12 个病灶均位于胰尾, 与周围组织边界清晰, 为富血供结节; 病灶最大径 0.30~3.00 (1.43±0.75) cm。MRI 表现: T1W1 均呈低信号, T2W1 8 例呈高信号(80%), DWI 7 例呈高信号(70%); 增强扫描动脉期 6 例呈均匀强化, 只有 1 例呈“花斑样”不均匀强化。3 例行⁶⁸Ga-dotatate PET/CT 检查的患者病灶均呈高代谢, 最大标准摄取值约为脾脏标准摄取值(SUV)的一半。**结论** 胰腺内副脾一般位于胰尾, 病灶<3 cm, 边界清晰; 在 MRI T1W1 序列上呈低信号, T2W1 及 DWI 多呈高信号, 增强扫描动脉期很少见到典型的“花斑样”不均匀强化, 静脉期及延迟期多为均匀强化, ⁶⁸Ga-dotatate PET/CT 检查时可能会出现高代谢。

【关键词】 胰腺; 副脾; 胰腺神经内分泌肿瘤; 误诊

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Characteristics of intrapancreatic accessory spleen misdiagnosed as neuroendocrine tumors and its misdiagnosis analysis

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【Abstract】Objective To investigate the characteristics of patients with intrapancreatic accessory spleen (IPAS) and the reasons for misdiagnosis to improve the understanding of the disease and reduce misdiagnosis and unnecessary surgery. **Methods** A retrospective analysis was made on 10 patients (12 lesions in total), who were admitted to the Department of Hepatobiliary Surgery of First Medical Center or Department of General Surgery of Second Medical Center of Chinese PLA General Hospital from January 2012 to August 2022, and were preoperatively diagnosed as pancreatic neuroendocrine tumor and postoperatively confirmed as intrapancreatic accessory spleen by pathology. Their CT, MRI, ⁶⁸Ga-dotatate PET/CT images, pathological characteristics and clinical data were investigated.

Results Out of 10 patients, nine had single lesion, and one had three lesions; 12 lesions were located in the tail of the pancreas, having clear borders with surrounding tissues, being nodules with rich blood supply, and having a maximum diameter of 0.30–3.00 (1.43±0.75) cm. On MRI, all lesions showed hypointensity on T1W1, eight hyperintensity on T2W1 (80%), seven hyperintensity on DWI (70%), six uniform enhancement in arterial phase, and one "patina-like" uneven enhancement. The lesions in the three patients who underwent ⁶⁸Ga-dotatate PET-CT all showed hypermetabolism, and the maximum standard uptake value (SUV_{max}) was about half of that of the spleen. **Conclusion** IPAS is generally located in the tail of the pancreas, with less than 3 cm and a clear boundary. The lesions show hypointensity on MRI T1W1 sequence, hyperintensity on T2W1 and hyperintensity on DWI sequence. The typical "patina-like" uneven enhancement is rarely seen in the arterial phase, venous phase and delayed phase display uniform enhancement mostly, and hypermetabolism may occur during ⁶⁸Ga-dotatate PET/CT examination.

【Key words】 pancreas; accessory spleen; pancreatic neuroendocrine tumor; misdiagnosis

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副脾指在脾脏以外的部位出现脾脏组织,是一种先天性异常表现,发生率约 10%,多出现在脾门(约 80%)及胰尾(约 20%)^[1,2]。副脾出现在胰腺即为胰腺内副脾(intrapaneatic accessory spleen, IPAS),是一种罕见的胰腺良性疾病,一般单发,发生率约 2%^[3]。IPAS 一般无明显症状,多于影像学检查时发现,但易被误诊为胰腺肿瘤,尤其是胰腺无功能性神经内分泌肿瘤(pancreatic neuroendocrine tumor, PNET),从而进行了原本不必要的手术。IPAS 临床罕见,相关报道多为个案报告。本文回顾性分析 10 例术前诊断为 PNET、术后经病理确诊为 IPAS 患者的影像、病理及临床资料,探讨 IPAS 病例特点及误诊原因,旨在提高对该病的认识,减少误诊及不必要的手术。

1 对象与方法

1.1 研究对象

回顾性分析 2012 年 1 月至 2022 年 8 月于中国人民解放军总医院第一医学中心肝胆外科及第二医学中心综合外科就诊、术前诊断为 PNET、术后经病理确诊为 IPAS 的 10 例患者的病例资料,通过电子病历系统采集年龄、性别、主诉、手术方式、影像学资料及病理结果等临床信息。

1.2 影像学资料分析

CT 及 MRI 由 2 位经验丰富的放射诊断科医师阅片,重点关注病灶位置、形态、数目、大小、钙化、囊变、密度、信号及强化特点等。正电子发射计算机断层显像(positron emission tomography/computed tomography, PET/CT)由 2 位经验丰富的核医学科医师阅片,重点关注病灶位置、形态、数目、大小、密度、是否高代谢及标准摄取值(standard uptake value, SUV)等。

2 结果

2.1 临床特点

共收集 10 例病例,其中男 7 例,女 3 例;年龄 14~72(50.50±16.62)岁。因体检发现 7 例,上腹部胀痛不适就诊 3 例。所有患者术前查糖蛋白抗原 19-9(carbohydrate antigen 19-9, CA19-9)、血清癌胚抗原(carcinoembryonic antigen, CEA)、糖蛋白抗原 15-3(carbohydrate antigen 15-3, CA15-3)、糖蛋白抗原 125(carbohydrate antigen 12-5, CA12-5)、甲胎蛋白(alpha fetoprotein, AFP)等肿瘤标记物均在正常范围。6 例行机器人胰尾切除术,1 例行机器人肝尾叶活检+胰尾切除术,3 例行腹腔镜胰尾切除术(表 1)。

表 1 10 例 IPAS 患者的临床特点

Table 1 Clinical characteristics of 10 IPAS patients

Case	Gender	Age	Chief complaint	Preoperative diagnosis	Surgical approach	Lesion location	Lesion number	Whether to protrude the surface	Largest meridian of lesion(cm)	Clear boundaries	Postoperative diagnosis
1	Male	50	Physical examination	PNET with cystic degeneration	Robotic tail pancreatectomy	Pancreatic tail	1	Yes	3.0	Yes	IPAS with cystic degeneration
2	Female	42	Physical examination	PNET	Laparoscopic pancreatectomy	Pancreatic tail	1	No	2.0	Yes	IPAS
3	Female	59	Abdominal distention	PNET	Robotic tail pancreatectomy	Pancreatic tail	1	No	1.8	Yes	IPAS
4	Male	14	Physical examination	PNET	Laparoscopic pancreatectomy	Pancreatic tail	1	No	1.2	Yes	IPAS
5	Male	47	Physical examination	PNET with liver metastases	Robotic liver biopsy and pancreatectomy	Pancreatic tail and liver	1	No	2.0	Yes	IPAS
6	Female	66	Abdominal pain	PNET	Robotic pancreatectomy	Pancreatic tail	1	No	1.0	Yes	IPAS
7	Male	72	Abdominal pain	PNET	Laparoscopic pancreatectomy	Pancreatic tail	1	No	1.9	Yes	IPAS
8	Male	40	Physical examination	PNET	Robotic tail pancreatectomy	Pancreatic tail	1	No	1.1	Yes	IPAS
9	Male	51	Physical examination	PNET	Robotic tail pancreatectomy	Pancreatic tail	3	No	1.0; 0.5; 0.3	Yes	IPAS
10	Male	64	Physical examination	PNET	Robotic tail pancreatectomy	Pancreatic tail	1	No	1.4	Yes	IPAS

IPAS: intrapaneatic accessory spleen; PNET: pancreatic neuroendocrine tumor.

2.2 影像学表现

2.2.1 MRI 特点 10例患者中,1例仅行MRI平扫,9例行MRI增强扫描。与病灶周围胰腺相比:在T1W1序列上10例均呈低信号;在T2W1序列上8例呈高信号,1例呈外周高信号局部低信号,1例呈等信号;在DWI序列上7例呈高信号,1例呈等信号,2例呈低信号。增强扫描动脉期5例呈明显均匀强化,强化程度高于周围胰腺;1例囊壁明显强

化,强化程度高于周围胰腺;1例呈均匀强化,强化程度等于周围胰腺;1例呈均匀强化,强化程度略低于周围胰腺;1例呈不均匀强化,强化程度低于周围胰腺。静脉期8例呈轻度均匀强化,其中5例强化程度与周围胰腺相当,2例强化程度高于周围胰腺,1例强化程度低于周围胰腺;1例囊壁呈轻度强化,强化程度高于周围胰腺。延迟期表现同静脉期(表2,表3)。

表2 10例IPAS患者的MRI平扫特点

Table 2 MRI features of 10 IPAS patients

Case	T1W1		T2W1		DWI	
	Compared to pancreas	Compared to spleen	Compared to pancreas	Compared to spleen	Compared to pancreas	Compared to spleen
1	Hypointensity	Hypointensity	Hyperintensity (Internal hypointensity)	Hyperintensity	Hyperintensity	Hypointensity
2	Hypointensity	Isointensity	Hyperintensity	Isointensity	Hyperintensity	Isointensity
3	Hypointensity	Isointensity	Hyperintensity	Isointensity	Hyperintensity	Isointensity
4	Hypointensity	Isointensity	Hyperintensity	Isointensity	Hyperintensity	Isointensity
5	Hypointensity	Isointensity	Hyperintensity	Isointensity	Hypointensity	Hypointensity
6	Hypointensity	Isointensity	Hyperintensity	Isointensity	Hyperintensity	Isointensity
7	Hypointensity	Isointensity	Hyperintensity	Isointensity	Hyperintensity	Isointensity
8	Hypointensity	Isointensity	Isointensity	Isointensity	Hypointensity	Hypointensity
9	Hypointensity	Isointensity	Hyperintensity	Isointensity	Hyperintensity	Hypointensity
10	Hypointensity	Isointensity	Hyperintensity	Isointensity	Isointensity	Isointensity

IPAS: intrapancreatic accessory spleen.

表3 10例IPAS患者的增强MRI特点

Table 3 Enhanced MRI features of 10 IPAS patients

Case	Arterial phase			Venous phase			Delay phase		
	Strengthening	Compared to pancreas	Compared to spleen	Strengthening	Compared to pancreas	Compared to spleen	Strengthening	Compared to pancreas	Compared to spleen
1	Cystic wall strengthening	Higher	Higher	Cystic wall strengthening	Higher	Equal	Cystic wall strengthening	Higher	Equal
2	Significant uniform strengthening	Higher	Higher	Uniform strengthening	Equal	Equal	Uniform strengthening	Equal	Equal
3	Significant uniform strengthening	Higher	Higher	Uniform strengthening	Equal	Equal	Uniform strengthening	Equal	Equal
4	Significant uniform strengthening	Higher	Higher	Uniform strengthening	Equal	Equal	Uniform strengthening	Equal	Equal
5	Un-uniform strengthening	Lower	Equal	Uniform strengthening	Higher	Equal	Uniform strengthening	Higher	Equal
6	Significant uniform strengthening	Higher	Higher	Uniform strengthening	Lower	Equal	Uniform strengthening	Lower	Equal
7	-	-	-	-	-	-	-	-	-
8	Uniform strengthening	Equal	Higher	Uniform strengthening	Equal	Equal	Uniform strengthening	Equal	Equal
9	Uniform strengthening	Lower	Higher	Uniform strengthening	Equal	Equal	Uniform strengthening	Equal	Equal
10	Significant uniform strengthening	Higher	Higher	Uniform strengthening	Higher	Equal	Uniform strengthening	Higher	Equal

IPAS: intrapancreatic accessory spleen.

与脾脏相比,在 T1WI 序列上 9 例呈等信号,1 例呈低信号;在 T2WI 序列上 9 例呈等信号,1 例呈高信号;在 DWI 序列上 6 例呈等信号,4 例呈低信号。增强扫描动脉期 1 例呈现与脾脏类似的“花斑样”强化,8 例未见“花斑样”强化。静脉期 8 例强化与脾脏类似,1 例囊壁强化与脾脏类似。延迟期表现同静脉期。

2.2.2 CT 特点 10 例患者中,7 例仅行 CT 平扫,2 例行 CT 增强扫描。与病灶周围胰腺相比:7 例平扫患者中,3 例未发现明确病灶,3 例呈稍低密度,1 例呈稍高密度。增强扫描动脉期 1 例呈明显均匀强化,强化程度高于周围胰腺,1 例呈均匀强化,强化程度与周围胰腺大致相同;静脉期 2 例均呈轻度均匀强化,强化程度高于周围胰腺;延迟期表现同静脉期。

与脾脏相比:7 例平扫患者中,3 例未发现明确病灶,4 例呈等密度。增强扫描动脉期 1 例呈现与脾脏类似的“花斑样”强化,1 例未见“花斑样”强化。静脉期及延迟期强化方式均与脾脏类似。

2.2.3 ^{68}Ga -dotatate PET/CT 特点 10 例患者中有 3 例行 ^{68}Ga -PET/CT(^{68}Ga -dotatate PET/CT),3 例患者均呈高代谢,1 例最大标准摄取值(maximum standard uptake value, SUV_{max}):11.4,脾脏 SUV :21.7;1 例 SUV_{max} :9.5,脾脏 SUV :20.2;另外 1 例为外院资料,无法对 SUV 进行测量。

2.3 病理特点

10 例患者共发现 12 个病灶,9 例为单发,1 例有 3 个病灶。位置:12 个病灶均位于胰尾,11 个病灶位于胰腺实质内,1 个病灶突出胰腺表面。大小:病灶最大径 0.30~3.00(1.43±0.75)cm。颜色、质地:11 个病灶切面呈灰红色、质中,1 个病灶切面呈囊性、灰黄色、质中。边界:12 个病灶均与周围组织边界清晰。

3 讨论

IPAS 是一种先天性胰腺良性变异,无需特殊治疗,由于胚胎期某部分背侧胃系膜内脾芽融合失败或少量脾组织脱离主脾发育而成^[4-6]。IPAS 多无特征性症状,多于影像学检查时无意发现,当其体积较大时可能会出现压迫周围组织而产生的腹胀、恶心、反酸等症状^[7]。IPAS 于胰腺尾部多见,一般单发,通常大小多<3.0cm,边界清楚。本组病例中,因体检偶然发现 7 例,上腹部胀痛不适就诊 3 例;10 例患者病灶均在胰腺尾部,9 例为单发,1 例有 3 个病灶;12 个病灶中最大径为 3.0cm,均与周围组织边界清晰。本组病例的临床特点基本与上述相关

报道一致。

由于病灶通常较小及胰尾部易受肠道气体干扰,普通超声检查不易发现。IPAS 在超声内镜表现为圆形或卵圆形病灶,边界清楚,回声均匀,回声较周围胰腺略低,与脾脏相仿^[8]。超声内镜联合细针穿刺活检,依据病理结果可确诊 IPAS,当高度怀疑 IPAS 而其他影像学资料又无法确诊时,为避免不必要的外科手术,可在术前考虑行超声内镜联合细针穿刺活检。IPAS 在 CT 上通常与脾脏密度相似,一般高于或等于胰腺密度,动脉期不均匀强化,门脉期均匀强化;但当病灶较小时可出现密度低于胰腺、动脉期均匀强化等不典型表现^[9,10]。本组行上腹部 CT 平扫的 7 例患者中,3 例未发现明确病灶,4 例与脾脏相比呈等密度,3 例密度低于胰腺,1 例密度高于胰腺。2 例增强 CT 患者中,动脉期 1 例呈现与脾脏类似的“花斑样”强化。本组病例中某些患者在密度及强化方式方面未出现典型 IPAS 表现,分析原因可能与病灶较小、红髓及白髓比例与正常脾脏不同有关。典型的 IPAS 在 MRI 各序列上与脾脏表现类似:T1WI 低于胰腺,T2WI 高于胰腺,DWI 为高信号,动脉期为“花斑样”不均匀强化,门脉期为均匀强化,强化程度一般高于胰腺实质^[11,12]。本组病例中,10 例患者在 T1WI 序列上均为低信号,8 例在 T2WI 序列上呈高信号,7 例在 DWI 序列上呈高信号;只有 1 例在动脉期呈现典型的“花斑样”不均匀强化,8 例静脉期呈均匀强化。本组病例与上述典型的 IPAS 在 T2WI、DWI 及强化各期表现存在一定的差异,可能与本组病例病灶相对较小、红髓及白髓比例与正常脾脏不同有关。

^{68}Ga -dotatate PET/CT 是诊断 PNET 的特异性检查,其灵敏度和特异度均高达 80%~100%^[13],是术前诊断 PNET 的金标准,但由于脾脏存在对 ^{68}Ga -dotatate 的生理性摄取,当脾脏异位至胰腺时可出现 ^{68}Ga -dotatate 高代谢,而误诊为 PNET^[14]。本组病例中共有 3 例患者行 ^{68}Ga -dotatate PET/CT 检查,3 例患者均呈高代谢,测量病灶平均 SUV_{max} 为 10.5,约占脾脏 SUV 值(20.2)的一半。由于病灶较小, SUV 值未呈现与脾脏一致的明显高代谢而误诊为 PNET。脾脏内网状内皮细胞可拦截和破坏受损红细胞,副脾亦具有该特点,因此可通过 $^{99\text{m}}\text{Tc}$ 热变性红细胞显像特异性鉴别 IPAS 与 PNET,但如果病灶太小,会存在漏诊的可能^[15]。该组病例术前并未意识到存在 IPAS 的可能,未行此检查。

本组病例中,病例 1 在 MRI DWI 及增强各期上未呈现典型 IPAS 表现,加之病灶中间囊变,而误诊为 PNET 伴囊变。病例 5 在增强 MRI T1WI、

T2WI、动脉期、门脉期及延迟期表现均与典型IPAS表现一致,但由于该患者同时发现肝脏占位,因PNET具有易发生转移的特点,该病例仍误诊为PNET伴肝转移(肝脏术后病理证实为炎性病变)。病例4、9、10虽在MRI某些序列与典型IPAS表现一致,但动脉期均未出现“花斑样”不均匀强化,加之⁶⁸Ga-dotatate PET/CT示病灶为高代谢,且因病灶较小,代谢程度较正常脾脏低,而误诊为PNET。病例2、3、6、7、8在CT或MRI动脉期均未出现典型“花斑样”不均匀强化的IPAS表现。

综上,IPAS一般位于胰尾,病灶<3 cm,边界清晰,多为富血供结节。大多数IPAS在MRI T1W1序列上呈低信号,在T2W1序列上呈高信号,在DWI序列上呈高信号,增强扫描动脉期很少见到典型的“花斑样”不均匀强化,静脉期及延迟期多为均匀强化。由于脾脏对⁶⁸Ga标记生长抑素受体存在生理性摄取,因此IPAS患者行⁶⁸Ga-dotatate PET/CT检查时可能会出现高代谢。因此,对于胰尾部<3 cm边界清楚的富血供结节,如在CT、MRI上与脾脏相仿,即使⁶⁸Ga-dotatate PET/CT阳性,一定不要想当然的认为是PNET,仍要考虑存在IPAS的可能,可进一步行^{99m}Tc热变性红细胞显像检查,必要时可考虑超声或CT引导下穿刺活检,一旦确诊IPAS,即可定期随诊,尽量减少不必要的手术。

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