

Jejunal metastasis of testicular seminoma presented with severe gastrointestinal bleeding diagnosed by means of capsule endoscopy

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Abstract. The authors describe the case of a 46-year-old male presented with repeated massive gastrointestinal haemorrhage as the first clinical symptom of jejunal metastasis of a malignant tumour. Orchiectomy of the enlarged testis was subsequently carried out and testicular seminoma was found on histology. Metastases of seminoma to the liver and lung were detected.

The source of gastrointestinal bleeding was identified by means of capsule endoscopy. Push enteroscopy was performed subsequently. At the present time, capsule endoscopy is a non-invasive method capable of visualizing a substantial part of small intestinal mucosa within a major part or even the whole small bowel. Push enteroscopy allows to see nearly all mucosal surface but only within the upper part of the small intestine, i.e. duodenum and the proximal part of the jejunum (up to 60 to 120 cm away from the Treitz ligament).

As the tumour of our case was generalised when diagnosed and could not be treated either endoscopically or surgically, a cisplatin based chemotherapy was started (bleomycin, etoposide, cisplatinum). Evident regression of the pulmonary and liver metastases was achieved according to control CT scan and there was no further gastrointestinal bleeding during the subsequent nine-month follow-up.

Keywords: jejunal metastasis, testicular seminoma, obscure-overt gastrointestinal bleeding, push enteroscopy, capsule endoscopy

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Souhrn. Autoři prezentují případ 46-letého muže vyšetřovaného pro závažné recidivující krvácení do trávicího traktu, jehož zdrojem byla solitární metastáza seminomu do proximálního jejunu. Byla provedena orchiektomie zvětšeného levého varlete a histologie potvrdila seminom. Současně byly zjištěny metastázy do plic a jater.

Zdroj krvácení do gastrointestinálního traktu byl diagnostikován pomocí kapslové endoskopie, nové diagnostické metody umožňující vyšetření tenkého střeva. Následně byla pacientovi provedena push enteroskopie. V současnosti představuje kapslová endoskopie nejméně invazivní vyšetřovací metodu, schopnou zobrazit podstatnou část slizničního povrchu větší části nebo dokonce celého tenkého střeva. Push enteroskopie naproti tomu umožňuje vyšetření téměř celého povrchu tenkého střeva, ale pouze v proximálním úseku (duodenum a proximální jejunum zhruba 60 - 120 cm za Treitzovu řasu).

Tumor popsaného případu byl v době diagnózy již generalizovaný a metastáza do jejunum nebyla řešitelná endoskopicky ani chirurgicky. Proto byla zahájena standardní chemoterapie BEP (bleomycin, etoposid, cisplatina). Po šesti cyklech bylo dosaženo regrese plicních a jaterních metastáz. Nedošlo k další recidivě krvácení do gastrointestinálního traktu v průběhu následujícího devítiměsíčního sledování.

Klíčová slova: metastázy do jejunum, seminom varlete, krvácení do trávicího traktu z nezjištěného zdroje, kapslová endoskopie, push enteroskopie.

Metastases of malignant tumours to the small bowel are quite rare. Tumours mostly metastasizing to the small intestine are melanoma, cancers of the stomach, colon, kidney, lung, uterus, thyroid gland, teratomas, and breast cancer (3,4,7,9,21,23,26,29). The first clinical manifestations of these metastases are usually obscure overt or occult gastrointestinal bleeding or ileus (4,20,23,25,26,29). Testicular seminoma is a less frequent tumour (incidence is about 1.9/100 000).

In approximately 75 % of patients, the seminomas are non-metastasizing when diagnosed. Testicular seminoma metastasizes mostly to draining lymph nodes in the retroperitoneum (15 %), mediastinum and supraclavicular fossa. Only rarely does seminoma spread haematogenously to involve lung parenchyma, bone, liver or brain (6).

Metastasis of testicular seminoma to the small intestine is quite exceptional (14,15,19,20,30). We describe a case of jejunal metastasis of seminoma presented with severe gastrointestinal bleeding as the first clinical symptom of a generalized tumour.

Case report

A 46-year-old male with a history of ischaemic heart disease and chronic obstructive lung disease was referred for recurrent melaena resulting in severe anaemia (haemoglobin 50 g/L). Repeated colonoscopies and gastroscopies done elsewhere failed to identify the source of bleeding (obscure-overt bleeding). A total number of 20 blood transfusion units had to be administered.

Physical examination revealed an enlarged left testis suspected of being a tumour. Ultrasono-

graphy confirmed a tumourous mass (3 to 2 cm) of the left testis. CT scan showed numerous metastases to the lung and liver. The serum concentration of human chorionic gonadotrophin (hCG-beta) was 148.841 u/L.

The serum levels of lactate dehydrogenase (LDH) and alfa-1 fetoprotein (AFP) were normal. Left orchidectomy was performed. Histological examination revealed seminoma (Fig. 1).

Because of recurrent severe gastrointestinal bleeding (presented as malaena) capsule endoscopy was performed (M2A, Given Imaging). It revealed a sessile polypoid tumour in the proximal jejunum infiltrating the intestinal wall with acute bleeding from a central ulceration (Fig. 2). A large amount of malaena was seen in the jejunum and ileum so that its mucosa could not be evaluated properly. As diagnosis of metastasizing testicular seminoma was already stated at the time of capsule endoscopy, the bleeding tumour was considered to be a metastasis of seminoma to the proximal jejunum. As the jejunal tumour was within reach of standard small bowel videoendoscopy, push enteroscopy was performed subsequently (Olympus SIF140). This examination showed the same picture of sessile polypoid tumour at the proximal jejunum close to the Treitz ligament with the adherent clot (2 cm in diameter) with no active bleeding (Fig. 3).

As the tumour in our case was generalised when diagnosed and could not be treated either endoscopically or surgically a cisplatin based chemotherapy was started (bleomycin, etoposide, cisplatinum). Severe anaemia (haemoglobin 54 g/L) required further blood transfusions

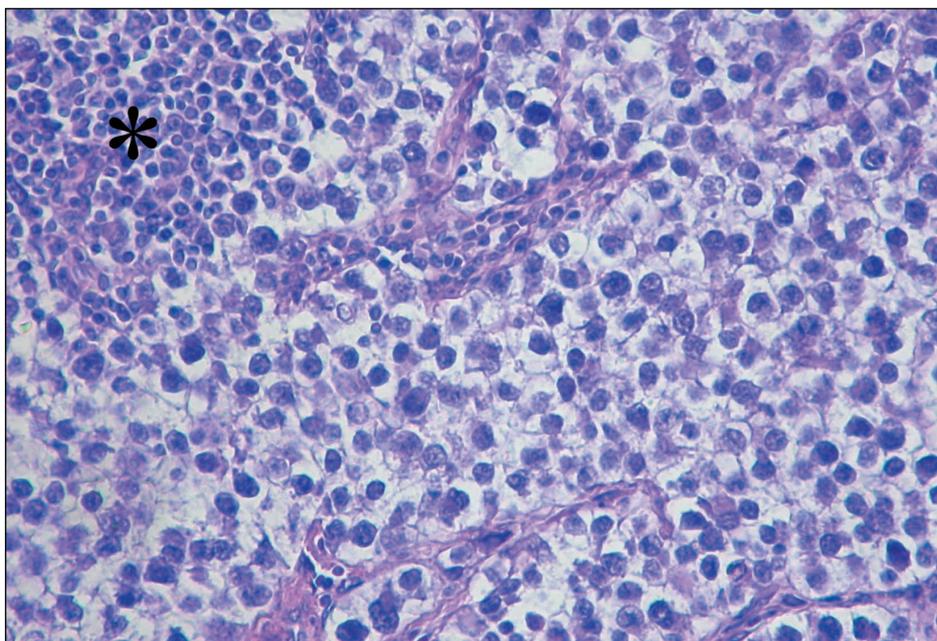


Fig. 1 / Obr. 1

Seminoma reveals a histologically solid growth pattern. It is composed of round polygonal cells with slightly anisomorphic nuclei and clear cytoplasm. Focally, interstitial lymphoid infiltration is present (asterisk) (haematoxylin-eosin).

Histologický obraz seminomu - solidně uspořádaný nádor tvořený okrouhlými či polygonálními buňkami s lehce anizomorfními jádry a světlou vakuolizovanou cytoplazmou. Fokálně je přítomna lymfoidní infiltrace v intersticiu (hvězdička) (Hematoxylin-eosin).

(a further 17 units). Evident regression of the pulmonary and liver metastases was achieved according to control CT scan after thirty-two weeks and there was no further gastrointestinal bleeding during the subsequent eight-month follow-up. However, a normal level of the hCG-beta had not been reached (control concentration is 37.2 u/L) and VIP chemotherapy was started (vinblastin, ifosfamide, cisplatinum). One month later the patient has been in partial clinical remission (remaining elevated level of the hCG-beta) with no signs of further gastrointestinal bleeding or metastases progression.

Discussion

The presented case of gastrointestinal bleeding from jejunal metastasis of testicular seminoma is quite exceptional for several reasons. Severe gastrointestinal bleeding was the first clinical symptom of a generalized tumour. The source of bleeding was recognised owing to capsule endoscopy, an advanced new technology that enables endoscopic investigation of the small intestine on a non-invasive basis. As standard push video-endoscopy was performed as well we could compare both these imaging methods.

Last but not least, the metastatic involvement of the jejunum by testicular seminoma is a very rare cause of severe overt gastrointestinal bleeding.

Germ cell tumors arising in the testes secondarily rarely involve the bowel, and such involvement is usually via extension from adjacent metastatic lymphadenopathy (15,19). To the authors' knowledge, sporadic reports published to date have described the metastasis of testicular seminoma to the small bowel (14,20,30). The predominant symptoms of metastatic involvement of the small bowel are mostly related to obscure-occult or obscure-overt bleeding or upper intestinal obstruction (4,20,23,25,26,29).

In our case the patient was indicated for capsule endoscopy after previous negative endoscopic (gastroscopy, colonoscopy) and X-ray (CT scan) examinations done elsewhere. We used a wireless capsule measuring 11 to 26 mm. The capsule contains a miniature video camera, a light source, batteries and a video transmitter. It moves through the small bowel, propelled by peristalsis and is naturally passed through. During the procedure, the capsule transmits data to a portable recorder, which is carried on a belt. Data can be analysed by special compu-

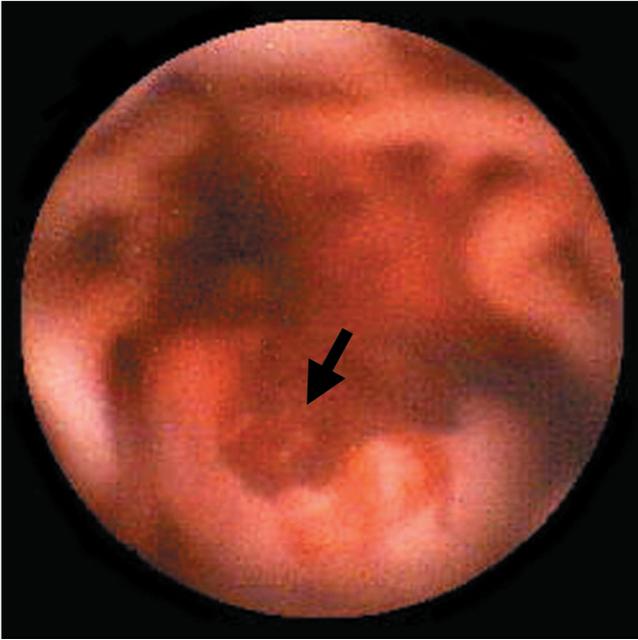


Fig. 2 / Obr. 2

Jejunal metastasis of seminoma (arrow) found by means of capsule endoscopy. Polypoid tumour of proximal jejunum with a central ulceration is bleeding spontaneously.

Kapslová endoskopie - metastáza seminomu v jejunu (šipka). Polypoidní tumor proximálního jejunu s centrální ulcerací a tekoucím krvácením.

ter software. It provides direct colour video images of the gastrointestinal mucosa at a rate of 2 images per second for approximately 8 hours. In our case the capsule endoscopy identified active bleeding in the proximal jejunum (Fig. 2), the source was a solitary polypoid metastasis. As the source of bleeding was within the reach of standard push video-enteroscope, we completed this investigation, too. Push enteroscopy confirmed the previous findings. However, push video-enteroscopy provides a higher quality of endoscopic imaging when compared to capsule endoscopy (see Figs. 2 and 3 for comparison). As the tumour of our case was generalised when diagnosed and could be treated neither endoscopically nor surgically, chemotherapy was started with relatively favourable effect.

The radical inguinal orchiectomy with high ligation of the spermatic cord is recommended for any patient with a suspected testicular tumor. It provides material for histologic diagnosis. The seminomas have been shown to have high sensitivity to both radiotherapy and chemotherapy. Adjuvant radiotherapy (pelvic, paraaortic) is the standard treatment for patients with early-stage seminoma after orchiectomy. Patients with more

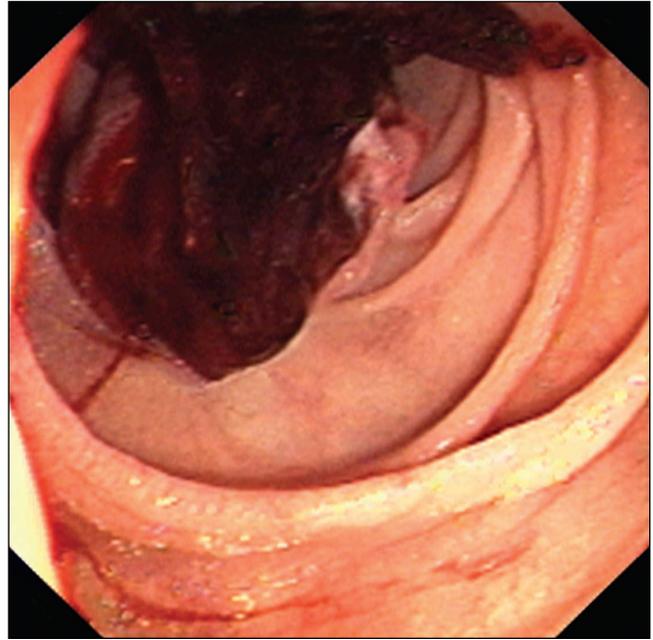


Fig. 3 / Obr. 3

Jejunal metastasis of seminoma. The same tumour (as seen in Fig. 2) at push-enteroscopy. The tumour is about 3 cm in diameter covered partly with an adherent blood clot.

Push enteroskopie - metastáza seminomu v jejunu. Polypoidní tumor, stejný jako na Obr. 2, zhruba 3 cm v průměru, částečně krytý Inoucí koagulem.

advanced disease (stage IIC and higher) have a high risk of systemic relapse, and the standard treatment for these patients after orchiectomy is combination chemotherapy (BEP) (6,24).

Measurements of AFP, hCG-beta, and LDH are important in management of patients with testis tumours. In 10 % of patients with seminomas, hCG-beta is elevated. Its elevation may correlate with metastatic disease but not with overall survival (10,12,18). If hCG-beta does not normalize after orchiectomy, some authors (24) suggest treating patients as if they have nonseminomatous germ cell tumours (with only 10 % incidence of pure seminomas producing hCG-beta, some patients dying of metastatic seminoma were found to have non-seminomatous elements at autopsy). The overall survival rates for patients with pure seminoma is generally greater than 90 %. Nearly 100 % of patients with stage I testicular seminoma are cured (6,24).

Capsule endoscopy is a new wireless endoscopic examination of the small bowel. To date, only limited data on its efficacy are available (16). Capsule is mostly indicated for both obscure overt and occult gastrointestinal bleeding (2) and its diagnostic yield is about 60 % (17). Capsule

endoscopy is a non-invasive method capable of visualizing a substantial part of small intestinal mucosa within a major part or even all of the small bowel (in 50 to 82 % of cases) (8,27,28). That is the most important advantage compared to other methods (push enteroscopy, enteroclysis) (16). However, it is important to realize that even capsule endoscopy can "miss a target" in an experimental setting (1). In Italian multicentre clinical study, capsule endoscopy failed to visualize small bowel tumour that was subsequently diagnosed by push enteroscopy (22). In our case, capsule endoscopy was successful, metastasis of seminoma in proximal jejunum was found.

The contraindications of wireless capsule endoscopy are known or suspected obstruction/stenosis of the gastrointestinal tract, intesti-

nal pseudo-obstruction, heart pacemakers and other electromechanical implants and swallowing disorders, early childhood and pregnancy.

In cases of the obscure/overt gastrointestinal bleeding, rare causes of bleeding, such as metastasis into the small bowel, should be also considered. Capsule endoscopy seems to be a promising method for the evaluation of the small bowel. The capsule endoscopy is a method of choice in diagnosing gastrointestinal bleeding in those cases, where the conventional diagnostic procedures (gastroscopy, colonoscopy,) were not successful. If the bleeding site is within the proximal small bowel, push enteroscopy will be indicated to endoscopic treatment and/or biopsy. A distal small bowel site will require surgical intervention (contingency with intra-operative enteroscopy).

References

- Appleyard M, Fireman Z, Glukhovskiy A, Jakob H, Shreiver R, Kadiramanathan S, Lavy A, Lewkowicz H, Scapa E, Shofti R, Swain P, Zaretsky A. A randomized trial comparing wireless capsule endoscopy with push enteroscopy for the detection of small-bowel lesions *Gastroenterology* 2002; 119: 1431 - 1438.
- ASGE Technology Status Evaluation Report. Wireless capsule endoscopy. *Gastrointest Endosc* 2002; 56: 621 - 624.
- Berger A, Cellier C, Daniel C, Kron C, Riquet M, Barbier JP, Cugnenc PH, Landi B. Small Bowel metastases from primary carcinoma of the lung, clinical findings and outcome. *Am J Gastroenterol* 1999; 94: 1884 - 1887.
- Bureš J, Široký M, Kopáčová M, Dědič K, Nožička J, Rejchrt S. Metastatic involvement of upper GI tract. *Endoscopy* 2000; 32, Suppl 1: E6.
- Cave D. Wireless video capsule endoscopy. *Perspect Gastroenterol* 2002; 5: 203 - 207.
- Davis JW, Schellhammer P. Testicular Seminoma. *Emedicine* 2002; <http://www.emedicine.com/med/topic2250.htm>
- Elsayed AM, Albahra M, Nzeako UC, Sobin LH. Malignant melanoma in the small intestine a study of 103 patients. *Am J Gastroenterol* 1996; 91: 1001 - 1003.
- Faigel DO, Fennerty M.B. "Cutting the cord" for capsule endoscopy. *Gastroenterology* 2002; 123: 1385 - 1388.
- Fassler I, Regent D, Gay G. Small intestinal metastasis of a malignant melanoma. *Acta Endosc* 1998; 28: 403.
- Fossa A, Fossa SD. Serum lactate dehydrogenase and human choriongonadotrophin in seminoma. *Br J Urol.* 1989; 63: 408 - 415.
- Gong F, Swain P, Mills T. Wireless endoscopy. *Gastrointest Endosc* 2000; 51: 725 - 729.
- Hori K, Uematsu K, Yasoshima H, Yamada A, Sakurai K, Ohya M. Testicular seminoma with human chorionic gonadotropin production. *Pathol Int* 1997; 47: 592 - 599.
- Iddan G, Meron G, Glukhovskiy A, Swain P. Wireless capsule endoscopy. *Nature* 2000; 405: 417.
- Johnson DE, Appelt G, Samuels ML, Luna M. Metastases from testicular carcinoma. Study of 78 autopsied cases. *Urology* 1976; 8: 234 - 236.
- Leslie JA, Stegemann L, Miller AR, Thompson IM. Metastatic seminoma presenting with pulmonary embolus, inferior vena caval thrombosis, and gastrointestinal bleeding. *Urology* 2003; 62: 144.
- Lewis B, Goldfarb N. Review article: the advent of capsule endoscopy - a not-so-futuristic approach to obscure gastrointestinal bleeding. *Aliment Pharmacol Ther* 2003; 17: 1085 - 1096.
- Lewis BS, Swain P. Capsule endoscopy in the evaluation of patients with suspected small intestinal bleeding: results of a pilot study. *Gastrointest Endosc* 2002; 56: 349 - 353.
- Mann K. Tumor markers in testicular cancer. *Urologe A* 1990; 29: 77 - 86.
- Miller TT, Mendelson DS, Wu LT, Halton KP. Seminoma of the testis presenting as an ulcerating mass of the duodenum. *Clin Imaging* 1992; 16: 201 - 203.
- Nakamura A, Ikeda Y, Morishita S, Sato Y, Matsumoto M, Inomoto T, Kariya T, Inoue T. Upper gastrointestinal bleeding arising from metastatic testicular tumor. *J Gastroenterol* 1997; 32: 650 - 653.
- O'Brien T, Matthews JB. Diagnosis and management of small bowel neoplasms. *UpToDate in Gastroenterology and Hepatology*. Wellesey, 2000, vol 8, No 3, <http://www.uptodate.com>
- Pennazio M, Santucci R, Rondonotti E, Abbiati C, Beccari G, Luchetti R, Dezi A, Capurso L, de Franchis R, Rossini F. Capsule endoscopy: diagnostic yield and comparison with push enteroscopy in patients with obscure gastrointestinal bleeding. The Italian multicentre experience. *Endoscopy* 2002; 34, suppl 2: A91.
- Rejchrt S, Dědič K, Živný P, Kopáčová M, Široký M, Bureš J. Metastatic involvement of small bowel. *Gastrointest Endosc* 2000; 51, No 4, Part 2: AB 299.
- Richie JP. Neoplasms of the testis. In: PC Walsh, AB Retik, ED Vaughan Jr, AJ Wein, eds. *Campbell's Urology*. 7th ed. Philadelphia: WB Saunders, 1998.
- Sebastian J, Zaragozo R, Vincente J, Gallego O, Trufero JM. Duodenal obstruction secondary to a metastasis from an adenocarcinoma of the cecum, a case report. *Am J Gastroenterol* 1997; 92: 1051 - 1052.
- Small Bowel Investigation & Atlas of Enteroscopy. J Bureš, S Rejchrt, eds. Praha: Grada Publishing, 2001: 480.

27. Smith MA, Mergener K, Schembre DB, Brandabur JJ, Kozarek RA. Complications and problems with capsule endoscopy. *Am J Gastroenterol* 2002; 97, No 9 Suppl: S301.
28. Swain P. Wireless capsule endoscopy. *Gut* 2003; 52, Suppl 4: 48 - 50.
29. Tacheci I, Ryška A, Čáp J, Široký M, Rejchrt S, Špaček J, Kopáčová M, Bureš J. Epiteloidní hemangiosarkom štítné žlázy - kazuistika (Epitheloid haemangiosarcoma of the thyroid gland - a case report). *Čas Lék čes* 2002; 141: 291 - 293.
30. Veen HF, Bruining HA. Gastro-intestinal bleeding and malignancy of the testis. *Acta Chir Scand* 1979; 145: 363 - 365.

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