





## Case Report

# Incomplete sagittal septum of the bladder with cystolithiasis

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### Abbreviations & Acronyms

BD = bladder duplication  
 CSSB = complete sagittal septum of the bladder  
 ISSB = incomplete sagittal septum of the bladder  
 PVR = postvoid residual urine  
 Qmax = maximum urine flow rate  
 VUR = vesicoureteral reflux

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### How to cite this article:

Fujimoto T, Goto T, Kanematsu A *et al.* Incomplete sagittal septum of the bladder with cystolithiasis. *IJU Case Rep.* 2022; 5: 264–267.

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Received 9 February 2022; accepted 27 March 2022.  
 Online publication 30 April 2022

**Introduction:** Incomplete sagittal septum of the urinary bladder is an extremely rare congenital anomaly and one of the variations in bladder duplication. Herein, we report a case of incomplete sagittal septum of the bladder with cystolithiasis.

**Case presentation:** A 20-year-old man was referred to our department for examination and treatment of symptomatic cystolithiasis and a suspected giant ureterocele on the left side. Cystoscopy and urography performed under general anesthesia revealed anatomical structures suggestive of the sagittal septum of the bladder. Subsequently, transurethral septostomy and cystolithotripsy were performed. The detrusor muscle was microscopically identified, leading to the diagnosis of an incomplete sagittal septum of the bladder.

**Conclusion:** Although extremely rare, an incomplete sagittal septum of the bladder may be difficult to differentiate from a ureterocele, and should be considered when a large cystic lesion is found in the bladder.

**Key words:** cystolithiasis, incomplete sagittal septum of the bladder, ureterocele.

## Keynote message

We report a rare case of an incomplete sagittal septum of the bladder with cystolithiasis that was successfully treated with transurethral surgery. Although extremely rare, an incomplete sagittal septum of the bladder may be difficult to differentiate from a ureterocele, and should be considered when a large cystic lesion is noted in the bladder.

## Introduction

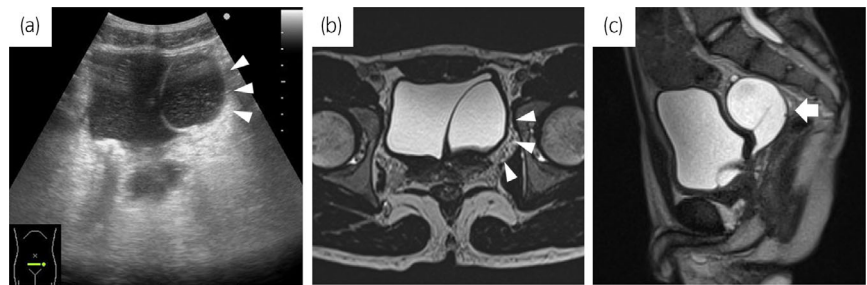
ISSB is a rare congenital urinary tract anomaly and one of the variations of BD.<sup>1</sup> There are a limited number of publications on BD, most of which are case reports or case series.<sup>2,3</sup>

Herein, we present a case of ISSB with cystolithiasis that was successfully treated using transurethral surgery.

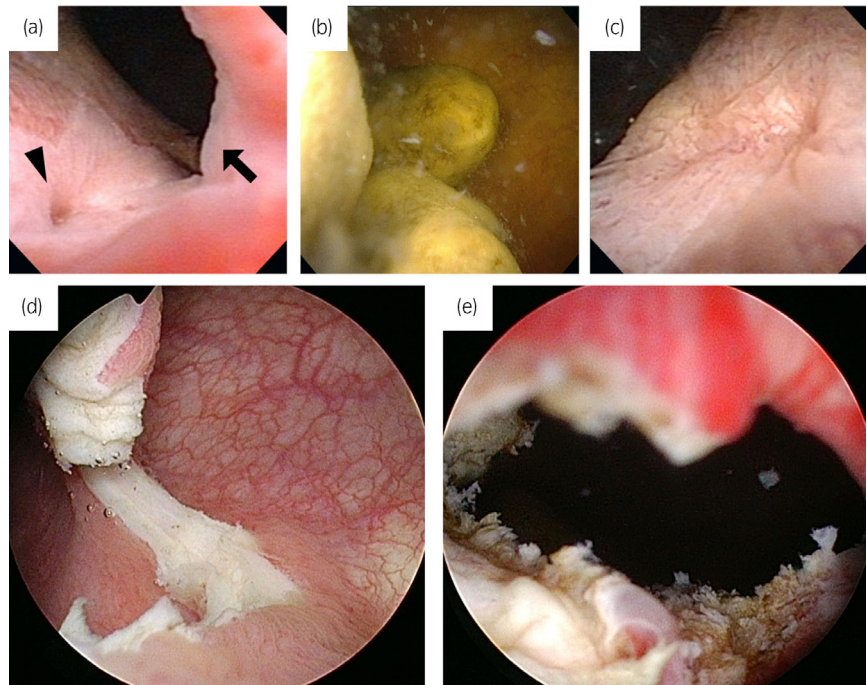
## Case presentation

A 20-year-old man was referred to our department for examination and treatment of symptomatic cystolithiasis and a suspected giant ureterocele on the left side. At birth, he was diagnosed with esophageal atresia, tracheomalacia, and cleft lip and cleft palate, for which he had undergone surgery. At the age of 18 months, he underwent liver transplantation, with his father as the donor, owing to a diagnosis of intrahepatic cholestasis.

The patient experienced intermittent lower abdominal pain. A urinalysis revealed pyuria. Ultrasonography and magnetic resonance imaging revealed a well-defined cystic lesion with a thickened wall on the left side of the bladder (Fig. 1a,b) and multiple calculi on the right side. The left-sided cystic lesion communicated with the left ureter and was suspected to be a giant ureterocele. There was also a midline cystic structure extending superiorly above the prostate,



**Fig. 1** The findings of (a) ultrasonography and (b, c) magnetic resonance imaging of T2-weighted axial and sagittal images at diagnosis. A well-defined cystic lesion (white arrowheads) with thickened wall is seen on the left side of the bladder. There is also a midline cystic structure extending superiorly above the prostate (white arrow), suggestive of a Müllerian duct cyst.



**Fig. 2** Cystoscopy findings showing that the bladder neck was divided into two parts: (a) wide left (black arrow) and narrow right (black arrowhead) outlets. (b) The right chamber of the bladder communicated to the narrow outlet in the bladder neck, which contained multiple calculi and cloudy urine. (c) A single normal ureteral orifice is present in the left chamber of the bladder, which was communicated to the wide outlet in the bladder neck. (d) The sagittal septum was resected with electrocautery from the bladder neck toward the posterior direction, which allowed the two chambers to communicate with each other and (e) the bladder outlet obstruction was relieved.

which was suggestive of a Müllerian duct cyst (Fig. 1c). Computed tomography revealed normal bilateral upper urinary tracts that were not suggestive of duplicated collecting systems. Cystoscopy revealed that the bladder neck was divided into two parts: wide left and narrow right outlets (Fig. 2a). The narrow outlet on the right side led to the right chamber of the bladder, which contained multiple calculi and cloudy urine (Fig. 2b). The cystoscope was naturally inserted through a wide left bladder neck into the left-sided space suspected of a giant left ureterocele in which a single normal ureteral orifice was identified (Fig. 2c). Uroflowmetry showed that the Qmax was preserved at 14.8 mL/s, and the excessive PVR volume derived from the right chamber was 287 mL.

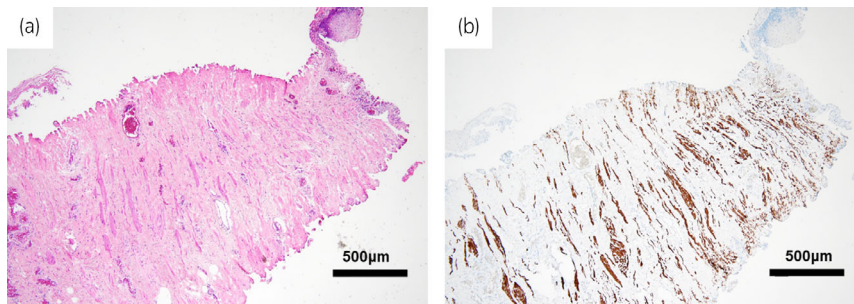
The findings from cystoscopy differed from those of a typical ureterocele, and transurethral surgery was performed under general anesthesia to determine the detailed anatomy of the urinary tract and treat cystolithiasis with bladder outlet obstruction. A single orifice was identified in each chamber and retrograde pyelography revealed a single bilateral upper urinary tract. Cystography revealed no VUR. The septum was incised and resected with bipolar electrocautery from the bladder neck toward the posterior, which allowed the two chambers to communicate with each other (Fig. 2d), and the

bladder outlet obstruction was relieved (Fig. 2e). Laser cystolithotripsy was performed at the end of the procedure to ensure that the patient was stone-free. Most components of cystolithiasis were calcium oxalate. Smooth muscle fibers were microscopically identified in the resected specimens of the septum (Fig. 3). In addition to anatomical findings, histopathological findings were consistent with the diagnosis of ISSB but not ureterocele (Fig. 4).<sup>1,4</sup> Postoperatively, the abdominal pain had disappeared, and dysuria had also improved with preservation of Qmax at 14.8 mL/s and a decrease in PVR volume at 37 mL on uroflowmetry. VUR was not observed in voiding cystourethrography.

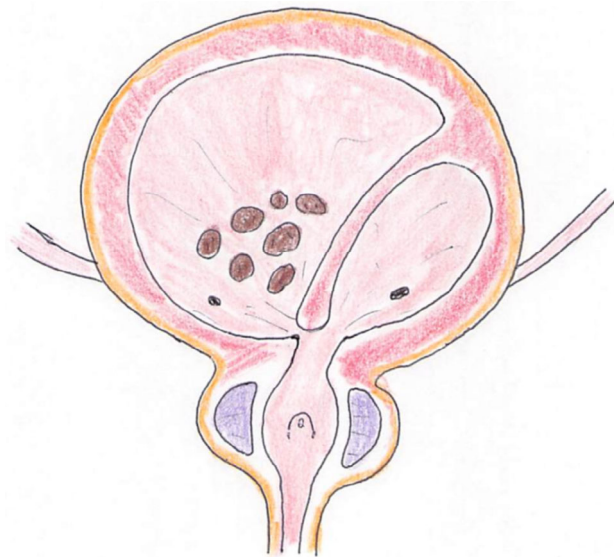
## Discussion

ISSB is one of the variations of BD, and only a few reports have been published. To the best of our knowledge, this is the first case report of ISSB concomitant with cystolithiasis.

BD is an extremely rare congenital anomaly of the urinary system that has been reported in less than 100 cases.<sup>3</sup> BD and related anomalies can be roughly divided into BD and septate bladder and further subclassified into various patterns according to the methods of separation: complete or



**Fig. 3** Histopathological findings of the resected septum. (a) Hematoxylin and eosin staining ( $\times 40$ ) and (b) immunohistochemical staining for Desmin ( $\times 40$ ). The smooth muscle is shown to be diffusely distributed from shallow to deep. Scale bar: (a) 100  $\mu\text{m}$  and (b) 100  $\mu\text{m}$ .



**Fig. 4** Image of the anatomical structure figured out from the operation. The bladder was separated into two chambers by a muscular sagittal septum, which communicated with each other at the bladder neck. The outlet of the right chamber was very narrow.

incomplete, sagittal or frontal, etc. BD consists of two bladders lying side-by-side and separated by a peritoneal fold, whereas the septate bladder consists of one bladder separated by the septum into two chambers.<sup>1</sup> BD is usually diagnosed in childhood and is associated with other congenital anomalies,<sup>3</sup> whereas in some cases, a patient with BD remains asymptomatic and therefore remains undiagnosed until adulthood.<sup>5</sup> Indications and methods of treatment for BD and related anomalies depend on the patient's symptoms and associated disorders; therefore, these need to be individualized for each case. Various surgical treatment options have been reported, including excision of the duplicated bladder and associated structures,<sup>2</sup> or transurethral septostomy.<sup>6</sup>

The present case was unique in that a ureterocele was suspected based on preoperative imaging findings, and it was difficult to differentiate ureterocele from ISSB. A ureterocele is defined as a cystic dilatation of the terminal ureter within the bladder, urethra, or both,<sup>4</sup> and it usually affects the upper moiety of a complete pyeloureteral duplication.<sup>7</sup> Moreover, hydronephrosis and VUR are often present in ureterocele cases.<sup>8</sup> Histologically, the wall of the ureterocele comprises a low amount of smooth muscle fibers and fibrous tissue.<sup>4</sup> In the present case, in addition to the anatomical findings, the

histological finding suggested the presence of sagittal septum of the bladder but not a giant ureterocele. Furthermore, the absence of upper urinary tract abnormalities, such as duplicated collecting systems or hydronephrosis, and no radiographic findings of VUR may have supported the diagnosis. In addition, whether this case involved a CSSB or ISSB was a problem. In the case of CSSB, the septum attached to the bladder wall along the entire periphery completely shuts the outlet from one chamber to the urethra, leaving only the other chamber in direct communication with the urethra. On the other hand, in the case of ISSB, the attachment of the septum to the bladder wall is not entirely circumferential, which allows the two chambers to communicate with one another and with the urethra.<sup>1</sup> The anatomical structures identified in the present case were consistent with those of ISSB.

In the present case, it was unique that although dysuria associated with ISSB may have been present for a prolonged period, it remained undiagnosed until adolescence owing to a lack of subjective symptoms, resulting in concomitant cystolithiasis. As for the clinically plausible mechanism of cystolithiasis formation, because excessive PVR and cystolithiasis were limited to the right chamber, the outlet obstruction of the right bladder chamber caused by the ISSB could have resulted in dysuria and cystolithiasis. Detailed cystoscopic observation under anesthesia allowed us to understand and diagnose the exact pathogenesis, leading to successful treatment with transurethral septostomy and cystolithotripsy.

## Conclusion

We encountered a rare case of an incomplete sagittal septum of the bladder with cystolithiasis that was successfully treated with transurethral surgery. Despite its rarity, it may be difficult to differentiate the incomplete sagittal septum of the bladder from other anomalies, such as ureterocele; therefore, incomplete sagittal septum of the bladder should be considered when a large cystic lesion is found in the bladder.

## Acknowledgments

We thank Editage ([www.editage.com](http://www.editage.com)) for English language editing.

## Author Contributions

Takeru Fujimoto: Conceptualization; data curation; writing – original draft. Takayuki Goto: Conceptualization; writing –

review and editing. Akihiro Kanematsu: Writing – review and editing. Hiroaki Nishimatsu: Resources; writing – review and editing. Masakazu Fujimoto: Data curation; writing – review and editing. Takashi Matsuoka: Writing – review and editing. Jin Kono: Writing – review and editing. Yuki Kita: Writing – review and editing. Kimihiko Masui: Writing – review and editing. Takeshi Sano: Writing – review and editing. Atsuro Sawada: Writing – review and editing. Shusuke Akamatsu: Writing – review and editing. Takashi Kobayashi: Conceptualization; supervision; writing – review and editing.

## Conflict of interest

The authors declare no conflict of interest.

## Approval of the research protocol by an Institutional Review Board

The study protocol for this research project was approved by our institutional review board (approval no. R1531).

## Informed consent

Informed consent was obtained from the patient for publication of this case report.

## Registry and registration no. of the study/trial

Not applicable.

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