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CASE REPORT

# Growth rate of a giant Tarlov (perineural) cyst with intrapelvic extension

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## Abstract

**Background and importance** Giant Tarlov cysts (GTCs) are perineural cysts and their presacral intrapelvic extension are extremely rare entities. We present a case of GTC with intrapelvic extension who has preoperative Magnetic Resonance Imaging (MRI) follow-ups of 12 years, and we demonstrate the annual growth rate and the time-size correlation of a GTC.

**Methods** Case report.

**Clinical presentation** A 37-year-old woman was admitted with left gluteal pain radiating to left foot, left leg numbness, progressed over 12 years. On MRI, starting from the L5–S1 level, a giant Tarlov cyst with an atypical configuration, is observed. The patient had a known sacral Tarlov cyst, first discovered on MRI obtained 12 years before the surgery. She had 6 consecutive MRI follow-ups in 12 years preoperatively. The cysts diameters have been measured and the growth rate was estimated. We showed for the first time that presented GTC grows in in both Sagittal Diagonal (SD) and Sagittal Craniocaudal (SC) diameters over time with overall annual growth rates, 7.671% for RGR_SD and 6.237% for RGR_SC.

**Conclusion** When the time-size correlation is observed, it becomes evident that the GTSs' growing speed increases over the years because of minimal resistance in the intrapelvic cavity. Early surgery may be considered to prevent rapid growth in the intrapelvic cavity and to reduce possible complications of the giant cyst.

**Keywords** Giant Tarlov cyst · Growth rate · Perineural cyst · Pelvic cyst · Presacral mass · Sacral meningocele

## Background and importance

Tarlov cysts (TCs) were first described by Tarlov [1]. Spinal Meningeal cysts (MCs) are classified into three categories; Type I MCs do not have spinal nerve root fibers, while Type II MCs have them. In Type III, MCs are intradural. TCs were named Type II meningeal cysts by Nabors and colleagues in 1988 [2]. The TC can expand due to cerebro spinal fluid pressure, which can cause stretching or compression of the sacral nerve roots. This can ultimately lead to progressive painful radiculopathy, sacral nerve root dysfunction or localized pain [3]. Bladder and sexual dysfunction, as well as symptoms related to the perineal area or the gastrointestinal system, may also be observed [4].

The etiology remains unclear and may be congenital or acquired, secondary to trauma, infection, inflammation, or iatrogenic causes [5].

Giant Tarlov cysts (GTCs) are exceptionally rare, although TCs are seen at a rate of 4.6%, and only 1% of these cases present with symptoms in adults. In GTCs, common symptoms include back pain and radicular findings [6]. They grow and extend into pelvis. There is a fistula path associated with the cysts content and the subarachnoid space and this path continues with the perineural structures that surround the nerve roots [7]. Intrapelvic cystic extensions may exceed 10 cm in diameter, and these patients may experience symptoms related to abdominal complaints such as

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abdominal pain and ureteral compression. When the cyst exceeds the boundaries of the sacral bone, its growth may accelerate.

We did not find any data about the growth rate of a GTC in the English literature. Here, we present a case of GTC with intrapelvic extension who has preoperative Magnetic Resonance Imaging (MRI) follow-ups of twelve years, and we demonstrate the annual growth rate and the time-size correlation of a GTC.

## Clinical presentation

A 37-year-old woman was admitted with left gluteal pain radiating to left foot, left leg numbness, progressed over 12 years. Her left leg numbness and severe pain is constant and aggravated when do up shoelace. On neurologic exam, her left S1 innervated muscles were slightly weak, left Achill reflex was absent contrary to the hyperreflexia on right Achill, her straight leg raising test was negative bilaterally. On MRI, starting from the L5-S1 level, a giant Tarlov cyst with an atypical configuration, filling and expanding the sacrum, especially in the left half, extending to the presacral area on the left with an extraspinal component, thin-walled in multiloculated character, isointense with CSF in all sequences, is observed. The patient had a known sacral TC, first discovered on an MRI obtained 12 years before. She had 6 MRI follow-ups in 12 years. MRIs belong to 12, 9, 6, 3 years, and 6 and 2 months before surgery. It is clear that the cyst has grown in years and reached to an intrapelvic giant mass (Fig. 1).

## Treatment plan and case follow-up

During the operation, only the posterior approach was performed in the prone Trendelenburg position. After opening the dorsolumbar fascia, left S1–S4 laminectomy was performed. The neck of the inferior cyst was opened by dissecting it from the nerve roots, and the defect in the left medial wall of the dura, which had active CSF leakage and filled the inferior cyst, was repaired with muscle grafts. Later, the superior cyst was opened at the S1–S2 level. Anterior dissection of the cyst allowed visualization of the cyst neck associated with the intrapelvic region. Then, the S1 nerve root sheath was opened, and flow was allowed into the cyst around the rootlets. CSF flow was restored with muscle grafts. Pain and numbness complaints completely disappeared in the postoperative period. On neurological examination, left S1 innervated muscle weakness disappeared

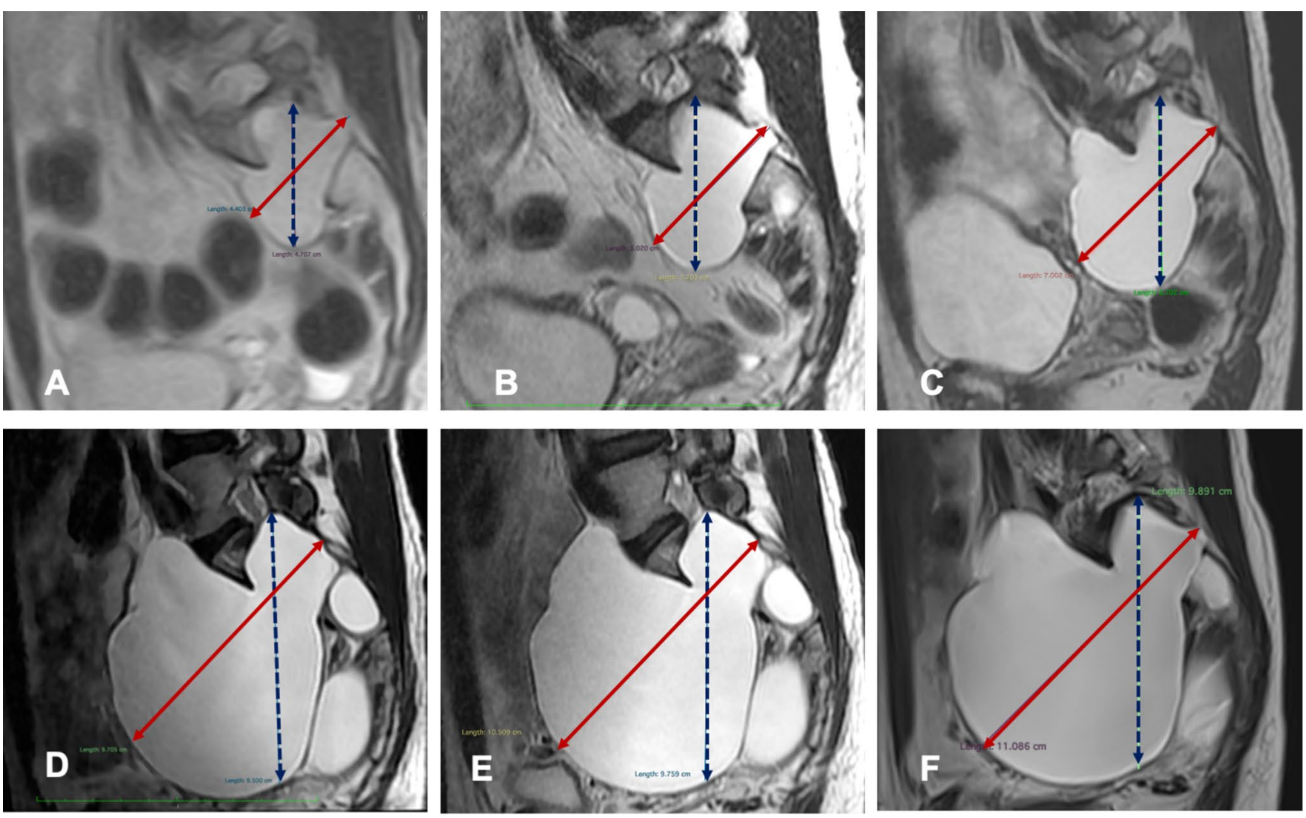


**Fig. 1** Figure shows the growth of a sacral GTC in twelve years with 6 consecutive sagittal T2 WI of lumbosacral MRI. **A**-12 years, **B**-9 years, **C**-6 years, **D**-3 years, **E**-6 months, and **F**-2 months before the surgery. Cyst clearly grows in years and reaches to an intrapelvic giant mass

and no deficit was detected. He could walk long distances without complaint. On MRI, the intrapelvic cyst completely disappeared. It has been followed for 24 months without any complaints.

## Radiological follow ups and growth rate measurements

### Method

We evaluated 12 years of 6 MRI follow-ups. The Sagittal Diagonal (SD) and Sagittal Craniocaudal (SC) maximum lengths at each MRI-date were used to calculate the size of the cyst at the given date. Maximum length of the SD axis was measured at 45° angle and SC axis was measured at 90° angle on sagittal T2WI sections (Fig. 2).

The annual relative growth rate was then calculated separately for SD and SC lengths. The Relative Growth Rate (RGR) of the cyst was calculated using the formula:

$$\mathrm{RGR} = \left(\ln W_2 - \ln W_1\right) / \left(t_2 - t_1\right)$$

where $W_1$ and $W_2$ are the maximum lengths at consecutive MRI dates and $t_2 - t_1$ is the time between consecutive MRIs. This formula is the classical method for calculating plant growth in biology and was adapted to this case [8]. MATLAB was utilized for calculations and creating graphical representations, ensuring in-depth analysis. Microsoft Excel was used for table creation, ensuring clear and concise presentation.

## Result

Table 1 is a summary of the data and provides a detailed breakdown of the annual RGR values for each MRI period, presenting the annual RGRs for both the SC (RGR_SC) and Sagittal Diagonal (RGR_SD) axes (Table 1). All calculations and evaluations demonstrated that the cyst grows in both directions over time and the line representing the diagonal length ascends more rapidly than the line for craniocaudal length (Fig. 3).

The first values for RGR_SC and RGR_SD are 0, due to the absence of preceding data for length comparison. The overall annual growth rates, 7.671% for RGR_SD and 6.237% for RGR_SC, represent the annual growth observed between the first and last MRIs. These annual growth rates aren't the averages of the five RGR values but rather the annual RGR within the irregular time intervals

**Table 1** The table provides a detailed breakdown of the annual Relative Growth Rate (RGR) values for each MRI period. RGR_SC represents annual relative growth rate on the Sagittal Craniocaudal and RGR_SD Sagittal Diagonal represents annual relative growth rate on the diagonal length in millimeters

| Date | Sagittal_ Craniocaudal | RGR_SC | Sagittal_ Diagonal | RGR_SD |
|---|---|---|---|---|
| 2010-02-18 | 47 | 0 | 44 | 0 |
| 2013-05-30 | 52 | 3.083 | 50 | 3.899 |
| 2018-09-13 | 66 | 4.504 | 70 | 6.357 |
| 2021-06-28 | 94 | 12.662 | 97 | 11.684 |
| 2021-12-04 | 97 | 7.212 | 105 | 18.176 |
| 2022-01-26 | 99 | 14.075 | 110 | 32.083 |

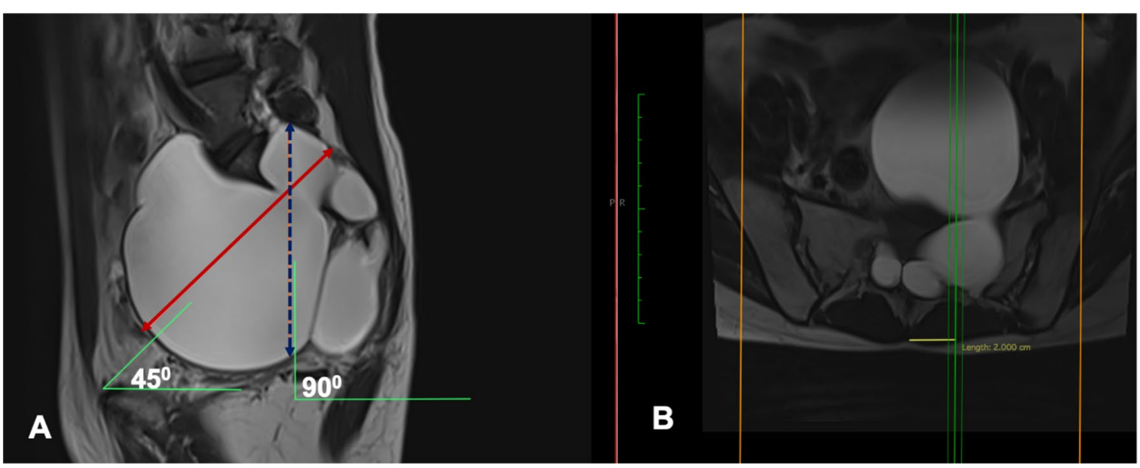


**Fig. 2** **A** shows the measurements of Tarlov cyst on MRI in sagittal plane. Dotted arrow represents vertical (Sagittal Craniocaudal) measurements of cyst diameter in 90° in all MRI's. Straight arrow represents anteroposterior oblique (Sagittal Diagonal) diameters in 45° in all MRI's. Because the cyst located laterally to left side, all measurements were performed 20 mm left to midline as shown in **B**

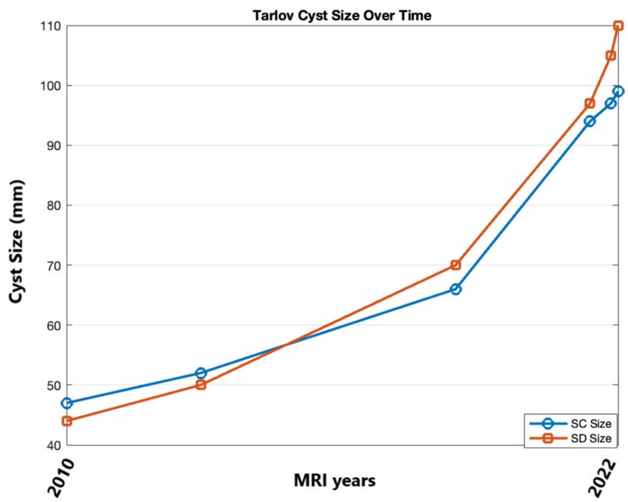


**Fig. 3** The graph illustrates the size of the cyst using sagittal diagonal and sagittal craniocaudal lengths between the first and last MRI dates. The cyst grows in both directions over time, and the line representing the diagonal length ascends more rapidly than the line for craniocaudal length. The size is in millimeters

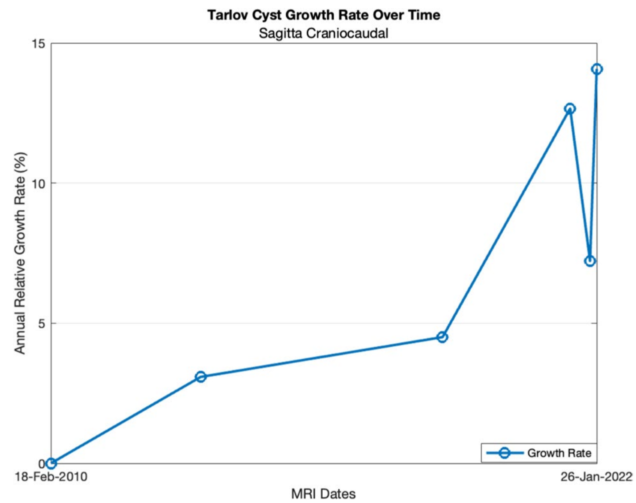


**Fig. 5** Annual Growth Rate Graph shows a parabolic upward inclination. When the time-size correlation is observed, it becomes evident that the cyst growing speed increases over the years in the SD axis. The line of the growth rate is fluctuating yet consistently positive, signifying continuous growth at varying speeds. The x-axis spans MRI dates, while the y-axis portrays the annual relative growth rate, offering a visual narrative of the cyst's behavior over time

in 12 years. The cyst grows 2,1 times in the craniocaudal and 2,5 in the diagonal axis over 12 years.

Figures 2 and 3 demonstrates that the annual growth rate of the cyst (Figs. 4 and 5). The x-axis spans MRI dates, while the y-axis portrays the annual relative growth rate, offering a visualnarrative of the cysts behavior over time. In the Sagittal Diagonal axis in Fig. 2, the line shows a parabolic upward inclination. When the time-size correlation is observed, it becomes evident that the cyst growing speed increases over the years in the SD axis. In the SC axis in Fig. 3, the line the growth rate is fluctuating yet consistently positive, signifying continuous growth at varying speeds.

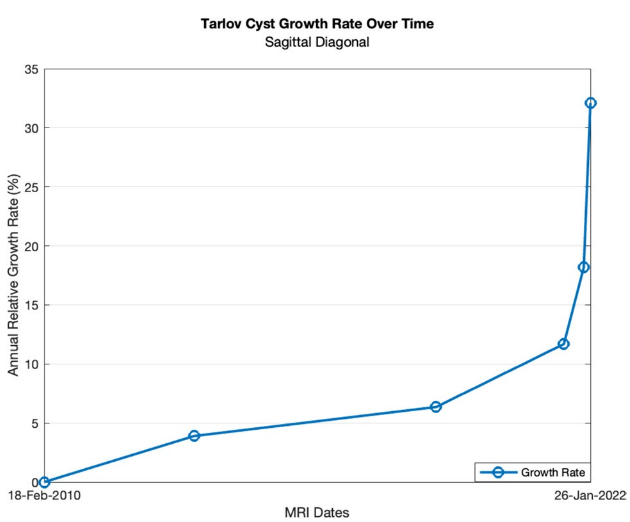


**Fig. 4** Annual Growth Rate graph shows a parabolic upward inclination. When the time-size correlation is observed, it becomes evident that the cyst growing speed increases over the years in the SD axis. The x-axis spans MRI dates, while the y-axis portrays the annual relative growth rate, offering a visual narrative of the cyst's behavior over time

## Discussion

The study spanned 12 years and utilized MRI data to evaluate the growth of a giant Tarlov cyst. Measurements were taken along the SD and SC axes at different angles. The volume calculations were not made due to its irregular shape and insufficient data. The annual RGR for both axes were calculated using a classical growth formula adapted for this case as 7.671% for RGR_SD and 6.237% for RGR_SC. The results showed that the cyst grew in both directions, with the SD axis exhibiting a faster growth rate. In the SC axis, the line the growth rate is fluctuating yet consistently positive, signifying continuous growth at varying speeds. This could be due to the bone shape and anatomy of the patient as the cyst growing speed decreases when it reaches the pelvic floor and is limited to bone and pelvic floor boundaries. Although these calculations were made in only one case, this provides us a reliable clue about the growth of a GTCs. These calculations need to be confirmed and refined with later research with a case series.

TCs tend to grow even though they are in a narrow volume within the bony structures of the sacrum, which limits their growth. Yang et al. demonstrated that growth of lumbosacral perineural TCs and their natural history [9, 10]. This is the only study shows the growth rate of TCs in the English literature. In their 28 patients, 5 subjects demonstrated growth. They showed that none of the cysts decreased in size between successive MRIs.

When the TCs overcomes the high resistance of the sacrum bone and reaches the low-pressure cavity in the pelvis, the growth rate increases rapidly and can reach giant sizes. All the GTCs can grow fast because they are reported to have a free flow of CSF between the cyst body and subarachnoid space [5]. The question we cannot answer now that is under what circumstances an intrasacral cyst may extend into pelvic cavity. Three theories may be proposed to explain intrapelvic extension; first, there is a congenital bone and soft tissue defect in the anterior sacral outlet where the cyst can find a natural route to pelvic cavity as in giant sacral anterior meningocele [11, 12]. Second, TCs have been associated with connective tissue disorders [13] but our patient did not have a diagnosis of connective tissue disorder. The third, the pressure within the cyst is high enough to enlarge the foramen in the anterior sacrum and the cyst can create a route to pelvic cavity. This theory is supported by publications of Urquiaga et al. and Puffer et al. [14, 15] in which they reported cases of TC causing significant progressive bone erosion in the sacrum, leading to an insufficiency fracture and instability, which required lumbosacropelvic stabilization.

GTCs usually cause back pain, radiating pain due to nerve root irritation, numbness, abdominal pain, increasing abdominal girth, and stress urinary incontinence secondary to bladder compression from the cyst. Although Ishii et al. reported that changes in posture did not alter the patient's symptoms, Yang et al. [5, 9, 10] proposed that complaints increase in situations where pelvic pressure increases and decrease when in a lying position. In our case symptoms were aggravated with increased pelvic pressure. We also believe that in intrasacral TCs the symptoms are due to compression of the sacral nerves, whereas in GTCs nerve stretching can also cause these symptoms.

One of the most important questions is when to operate on a symptom-free GTC patient. And is it important to size of the cyst when we make decision for operation? TCs usually grow over years, largely limited to bone and most are asymptomatic [9, 10]. Yang et al. [10] demonstrated that growth of lumbosacral perineural (Tarlov) cysts and their natural history. In their study, there was no association detected between worsening of symptoms and cyst growth. There is no consensus on the indication, timing and size of TC in the surgical treatment of TCs. Voyadzis et al. [16] recommended surgery for large sacral TCs (> 1.5 cm) with the presence of associated radicular symptoms. Burke et al. [17] recommended surgery for delayed contrast "filling of the cysts was identifed on computed tomographic myelography. Nulen et al. [18] recommended surgery for younger age, a single TC, and shorter duration of symptoms. The surgical decision does not directly correlate with the size of the cyst, but symptoms.

GTCs should differ from intrasacral TCs in surgical decision-making. In the present study, we showed for the first time that, when the cyst size gets bigger, its intrapelvic annual growing rate is getting faster. GTCs encounter minimal resistance in the intrapelvic distance comparing to sacral bone and therefore their growth increases rapidly in years. Therefore, surgical decision for GTCs can be made according to the size of the cyst. If cyst has wide intrapelvic extension, the operation should be considered, even if the patient is asymptomatic. Early surgery may reduce peroperative complications and postoperative headache. Acute CSF loss during GTC excision can reach up to 1000 ml. and sudden onset, progressive, severe headache can be seen [19]. Akahori et al. [20] reported spontaneous rupture of a huge presacral TC and resultant neurologic improvement in a case of symptomatic presacral TCs. Although this patient resulted in recovery, one should try to prevent spontaneous rupture.

## Conclusion

GTCs extending into the pelvis are quite rare compared to common TCs located into bony of sacrum. Additionally, their symptoms, diagnosis, treatment, follow-up, and management processes are different from intrasacral TCs. We showed for the first time that presented GTC grows in both directions over time with overall annual growth rates, 7.671% for RGR_SD and 6.237% for RGR_SC. Eventually, the cyst grows 2,1 times in the craniocaudal and 2,5 times in the diagonal axis over 12 years. When the time-size correlation is observed, it becomes evident that the cyst growing speed increases over the years because of minimal resistance in the intrapelvic cavity. Early surgery may be considered to prevent rapid growth in the intrapelvic cavity and to reduce possible complications of the giant cyst.

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## Declarations

**Conflict of interest** No funding was received to assist with the preparation of this manuscript.

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