

## PARATHYROID CYSTS WITH PRIMARY HYPERPARATHYROIDISM: REPORT OF A CASE

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Cysts of the parathyroid glands are uncommon, and, moreover functioning parathyroid cysts that cause primary hyperparathyroidism are rare. Herein is reported a 53-year-old female with primary hyperparathyroidism accompanied by 2 parathyroid cysts, in one of which adenoma was noticed. Forty-two cases of parathyroid cysts were found in the Japanese literature. Twelve of them were in the hyperparathyroid state, but infarction of the adenoma lead to cystic degeneration in most of such cases and so the cyst wall were lined with adenoma cells. In only 2 cases including our case were the cyst walls lined with cuboid cells and the adenoma evident in the wall. The pathogenesis of our case seems to be a common embryonic defect or dilatation of vestigial remnants rather than a degenerative change of the adenoma.

**Key words:** Parathyroid cyst, Primary hyperparathyroidism, Oxyphilic adenoma

Functioning parathyroid cysts are rare. Only 12 cases were found in the Japanese literature. Infarction of the adenoma leads to cystic degeneration in most of such cases. This report describes a case of a functioning parathyroid cyst which may originate from common embryonic defect or dilatation of vestigial remnants.

### CASE REPORT

A 53-year-old female patient was admitted to our clinic on September 24, 1981 because of headache, general malaise and constipation. Her history revealed that she had undergone left ureterolithotomy 4 years ago at our clinic, and right renal stones were also diagnosed at that time. During the first hospitalization, hypercalcemia as well as hypophosphatemia were pointed out, but the patient refused further examination.

She denied any history of peptic ulcer, lethargy, polyuria, polydipsia, pururitis, or bone pain. She had not taken any antihypertensive drugs or excessive vitamin D.

On physical examination, her pulse rate was 80 per minute and regular, blood pressure was 134/84 mmHg. No cervical mass was palpable. Laboratory studies revealed serum calcium of 11.1 mg/dl, chloride 111 mEq/L, phosphate 2.6 mg/dl and PTH 1.32 ng/ml. Tubular reabsorption of phosphate was 76.5 %. Urinalysis showed crystalluria of amorphous phosphate. The hemogram, ECG and chest X-ray were all normal.

IVP showed left contracted kidney and small stone shadows in the right kidney. Skeletal roentgenograms revealed endosteal resorption of the proximal phalanges.

Under the preoperative diagnosis of primary hyperparathyroidism, the cervical region was explored as shown in Fig. 1. A cystic mass with normal parenchymal tissue around it was found at the inferolateral to the lower pole of the right thyroid lobe. The size was  $2 \times 1 \times 1$  cm. The upper parathyroid gland was normal. A  $1.3 \times 0.5 \times 0.5$  cm tear-shaped, thin walled, colorless translucent cyst was detected at the lower pole of the left thyroid gland.

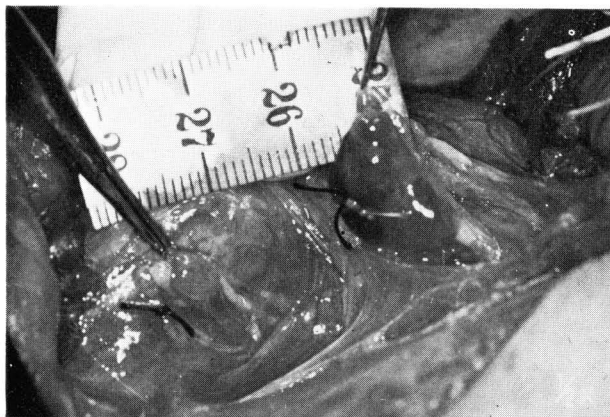


Fig. 1. Intraoperative photograph shows right parathyroid cyst in the right and right upper parathyroid gland which are indicated by the clamps

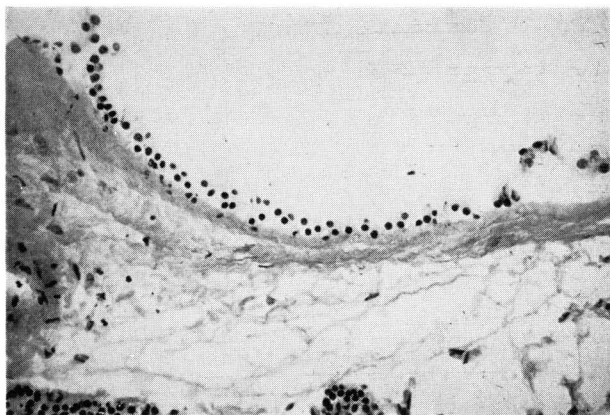


Fig. 3. Photomicrograph of the right parathyroid cyst wall shows cuboid cells lining the internal surface of the cyst ( $\times 400$ )

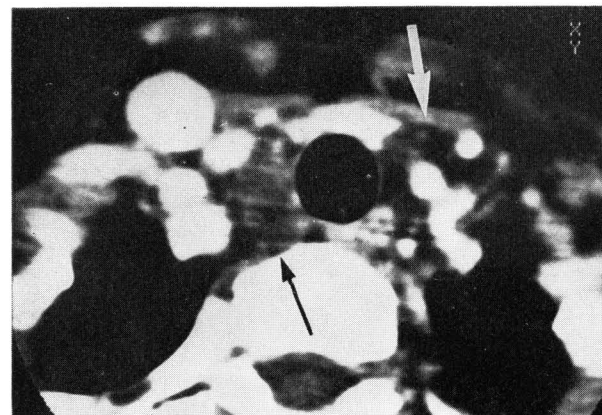


Fig. 2. Neck CT shows two cystic masses which were not accurately identified preoperatively. The black arrow indicates a right parathyroid cyst accompanied by an oxyphilic adenoma, and the white arrow reveals a left parathyroid cyst

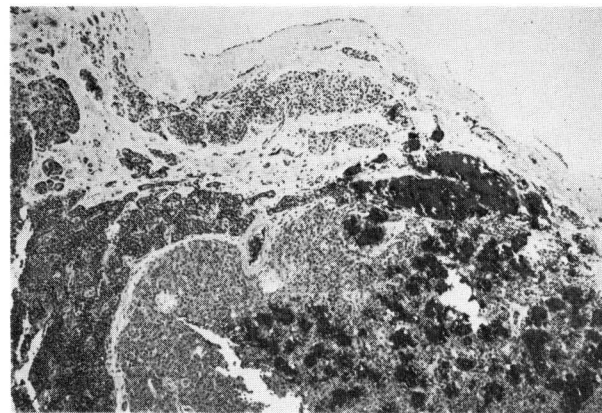


Fig. 4. Photomicrograph of the right parathyroid cyst wall shows an encapsulated oxyphilic adenoma on the lower right surrounded by chief cells ( $\times 400$ )

The upper one seemed to be normal. All but one third of the left upper gland were removed.

In the retrospective study of the neck CT, these two cystic lesions could be identified as shown in Fig. 2.

Pathological examination showed that the right inferior cystic parathyroid wall was lined with cuboid cells (Fig. 3), and the wall of the cyst was primarily composed of encapsulated oxyphilic adenoma (Fig. 4). The cyst wall in the left inferior side was also lined with cuboid cells, but normal parathyroid tissue was noticed in the cyst wall.

Convalescence was uneventful except numbness and tingling of her fingers several days postoperatively; these were well controlled with calcium and  $1\alpha$  (OH)  $D_3$  (alfarol®). Three months after the operation, the calcium level was normal without any medication.

### DISCUSSION

Goris was the first to report the removal of a parathyroid cyst in 1905, although the association of a parathyroid cyst with primary hyperparathyroidism was first reported by Greene et al in 1952<sup>11</sup>. In Japan, Shingu reported the first case in 1956.

Up to now, 42 cases have been accumulated in the Japanese literature. Age, sex, location of the cyst, size and the nature of the cystic fluid are summarized in Table 1. Parathyroid cysts are more common in women than in men. With regard to the location of the cysts, the lower parathyroid is more prone to be affected than the upper. Only 15.3 % of the cysts are located in the upper portion. Large cysts are more common than smaller ones, thus presenting as a cervical mass, or causing symptoms arising from compression to adjacent organs<sup>21</sup>. Sizes range from 1 to 10 cm in diameter. About two thirds of the cysts contain a watery clear fluid. Rarely the parathyroid cyst may combine with primary hyperparathyroidism. For a parathyroid cyst to be considered functional and the cause of hyperparathyroidism, the following criteria should be met<sup>21</sup>:

- 1) Preoperative biochemical and clinical evidence of hyperparathyroidism
- 2) Identification and normality of the remaining parathyroid glands
- 3) Histologic identification of parathyroid tissue within the cyst wall
- 4) Postoperative correction of hyperparathyroidism.

The case reported herein meets all of

Table 2. Reported cases of functioning parathyroid cysts

| No. | Authors        | Year | Age/sex | Clinical presentation                    | location                   | Size                       | Content   | Cyst wall            |
|-----|----------------|------|---------|------------------------------------------|----------------------------|----------------------------|-----------|----------------------|
| 1   | Shingu         | 1956 | 22/M    | bone fracture                            | Rt superior                | 3.0×2.5×2.0                | Colloid   | Adenoma degeneration |
| 2   | Mizutani       | 1962 | 35/F    | Urolithiasis                             | Rt inferior<br>Lt inferior | 0.8×0.7×0.5<br>0.5×0.5×0.4 | Colloid   | Cuboid cell.         |
| 3   | Nagahara       | 1962 | 64/F    | Autopsy                                  | Lt inferior                | 7.0×3.0×3.5                | —         | Adenoma degeneration |
| 4   | Sato           | 1966 | 36/M    | Neck tumor                               | Rt superior                | 5.5×4×4                    | Bloody    | Adenoma degeneration |
| 5   | Sato           | 1974 | 48/F    | —                                        | Lt                         | 4.3×2.6×2.4                | chocolate | Adenoma degeneration |
| 6   | Sato           | 1974 | 47/F    | bone fracture                            | Lt inferior                | 5×4.5×4                    | —         | —                    |
| 7   | Hattori        | 1975 | 40/F    | bone fracture                            | Lt inferior                | 2.0×1.9×1.7                | —         | Adenoma degeneration |
| 8   | Fujimoto       | 1976 | 30/F    | Neck tumor                               | Lt superior                | 5.8×3.5×2.9                | Brownish  | Adenoma              |
| 9   | Fukase         | 1976 | 30/F    | Neck tumor                               | Lt superior                | 1.5×1.5×1.5                | Yellowish | Parathyroid tissue   |
| 10  | Nunoi          | 1978 | 55/F    | Urolithiasis                             | Lt                         | —                          | Bloody    | —                    |
| 11  | Sakamoto       | 1980 | 64/M    | Constipation, thirsty<br>general malaise | —                          | —                          | —         | —                    |
| 12  | Present report | 1983 | 53/F    | Urolithiasis                             | Rt inferior<br>Lt inferior | 2.0×1.0×1.0<br>1.3×0.5×0.5 | Watery    | Cuboid cell.         |

Table 1. The 42 cases of parathyroid cysts reported (1982. 2)

| (1982. 2)    |                                   |    |         |
|--------------|-----------------------------------|----|---------|
| 1) Age :     | 21~64, average 43.8               |    |         |
| 2) Sex :     | Male : Female = 13 : 29 (1 : 2.2) |    |         |
| 3) Location: | Lt inferior                       | 24 | (61.5%) |
|              | Rt inferior                       | 5  | (12.8%) |
|              | Lt superior                       | 2  | (5.1%)  |
|              | Rt superior                       | 4  | (10.2%) |
|              | Bilat. inferior                   | 2  | (5.1%)  |
|              | Mediastinum                       | 2  | (5.1%)  |
| 4) Size :    | diameter $\geq 3^{\text{cm}}$     | 20 | (69.0%) |
|              | diameter $< 3^{\text{cm}}$        | 9  | (31.0%) |
| 5) Content : | Watery clear                      | 15 | (60.0%) |
|              | Colloid                           | 3  | (12.0%) |
|              | Bloody                            | 7  | (28.0%) |

these criteria. As shown in table 2, 12 cases of the parathyroid cysts reported in Japan are considered to be functional<sup>6-15)</sup>.

The major theories concerning the origin of parathyroid cysts are as follows<sup>3, 4)</sup>:

- 1) Gradual accumulation of colloid or simple retention cysts
- 2) Dilatation of vestigial remnants of the third and fourth brachial clefts
- 3) Coalescence of smaller microcysts
- 4) Cystic degeneration of parathyroid adenoma.

The cyst fluid of functional parathyroid cysts can be divided into bloody or straw-colored fluid are considered to be due to the infarction or degeneration of parathyroid adenomas<sup>5)</sup>. High PTH level in the cyst fluid is expected. In most of them, the cyst wall is lined with adenoma cells or hyperplasia cells. Only in case 9, was the cyst wall lined with normal parathyroid cells, but the fluid in the cyst showed a high PTH level. The author attributes it to the the cyst degeneration of the adenoma.

In our case as well as case 2, clear cyst fluid and cyst wall lined with cuboid

cells are recognized. No evidence of degeneration of the adenoma was found. The encapsulated oxyphilic adenoma existed in the right parathyroid cyst wall. Symptoms of hyperparathyroidism are attributed to the oxyphilic adenoma and the origin of both parathyroid cysts located in the inferior portion seems to be a common embryonic defect or dilatation of vestigial remnants rather than a degenerative change of the adenoma<sup>1,2,4, 5)</sup>.

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## 和文抄録

## 副甲状腺機能亢進症を伴った両側副甲状腺嚢腫の1例

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症例は53歳，女性．頭痛，全身倦怠感を主訴として1981年9月24日当科に入院した．入院時諸検査にて副甲状腺機能亢進症と診断され，頸部CTで右上方に位置する腺腫を疑われ手術した．術中所見では，術前“腺腫”と予想されたのは正常の副甲状腺組織であり，甲状腺下極両側に2個の副甲状腺嚢腫が発見された．嚢胞壁は一層の立方上皮細胞に被われ，内容液は無色透明で漿液性であった．また右側嚢腫壁内に実質部分があり，病理組織学的に Oxyphilic adenoma と診断された．

本邦文献上，副甲状腺嚢腫例の報告は現在までに42例あり，そのうち，副甲状腺機能亢進症を伴ったものは12例（28.6％）で，尿路結石，病的骨折を契機にして発見されたものが多い．しかし，これら12例中のほとんどは，腺腫の一部が壊死あるいは出血などにより嚢胞変性したものと考えられる症例で，自験例のように嚢胞壁内が立方上皮に被われ内容液が無色透明の所謂“真性副甲状腺嚢胞”で，しかも嚢胞壁内実質部に腺腫を認める例は，本邦では水谷らの報告につき第2例目である．