Asymptomatic diffuse idiopathic skeletal hyperostosis as a potential risk for severe dysphagia following partial laryngopharyngectomy

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ABSTRACT
Diffuse idiopathic skeletal hyperostosis (DISH) is a common disease in which ossification lesions occur in the bones including the vertebrae. Dysphagia may occur in advanced cases, but there are few cases that require treatment. A 68-year-old man was diagnosed with hypopharyngeal cancer of the left pyriform sinus and asymptomatic DISH on the anterior cervical vertebrae. Due to prior history of radiation, partial laryngopharyngectomy was performed. After surgery, severe dysphagia and aspiration pneumonia occurred, and the patient needed to undergo total laryngectomy. It was determined that dysphagia was due to multiple factors, including insufficient laryngeal elevation and esophageal compression by osteophytes of DISH. Asymptomatic DISH can cause severe dysphagia after partial laryngopharyngectomy. We suggest that evaluation of the swallowing function and surgical options, including laryngeal suspension and cricopharyngeal myotomy should be considered when performing partial laryngopharyngectomy in patients with DISH, even if they demonstrated no difficulties in swallowing before treatment.

Introduction
Diffuse idiopathic skeletal hyperostosis (DISH) is a common disease in which ossification lesions occur in the vertebrae, pelvis, and limb joints [1]. Majority of DISH cases are asymptomatic. Dysphagia may occur in advanced cases with anterior cervical vertebrae DISH, but there are few cases that require treatment [2]. Partial laryngopharyngectomy (PLP) is suitable for early stage hypopharyngeal cancer, especially for formerly radiated patients [3], but caution is needed to prevent postoperative dysphagia and/or aspiration pneumonia [4–7]. We report here T1 pyriform sinus cancer patient with asymptomatic DISH. He eventually needed to undergo the total laryngectomy following PLP because of severe dysphagia and aspiration pneumonia due to DISH.

Case report
A 68-year-old man was admitted to our department because of a hypopharyngeal tumor found via gastroscopy. He had the following history for cancer treatments: total gastrectomy for esophageal cancer invasion into the stomach, chemo-radiotherapy for esophageal cancer, and resection of oropharyngeal upper wall cancer. His height was 158 cm, and his weight was 52.3 kg. Hypopharyngoscopy (Figure 1(a)) revealed that the tumor was localized to the left pyriform sinus, and biopsy revealed squamous cell carcinoma. Computed tomography (Figure 1(b)) revealed the left pyriform sinus tumor (circle) without nodal metastasis, and the patient was diagnosed with hypopharyngeal cancer (pyriform sinus, T1N0). Computed tomography also revealed that osteophytes had developed on anterior cervical vertebrae (white arrow-heads) and that the esophagus was shifted to the left (arrow). However, he had no difficulties with oral intake. PLP was selected for the hypopharyngeal cancer, due to the history of irradiation of the neck. Pharyngeal reconstruction with a forearm flap was performed with left selective neck dissection (levels II, III, and IV). The tumor was excised together with upper lateral thyroid cartilage and a 2-cm mucosal safety margin. The resection area was confined to the pyriform sinus and adjacent posterior wall of the hypopharynx without extension to the apex of the pyriform sinus, the base of the tongue, or the larynx. The cervical esophagus was processed into a fish mouth, and the pyriform sinus was reconstructed with...
the forearm flap. The hypoglossal nerve, superior laryngeal nerve, and ansa cervicalis to the strap muscles were all preserved. Paratracheal dissection was omitted so that the inferior laryngeal nerve could be preserved. Pathological examination was negative for the surgical margin.

Swallowing rehabilitation was initiated immediately after surgery, but saliva aspiration was profuse, and the patient developed pneumonia. An intestinal fistula was created on the 64th postoperative day for the purpose of supplemental feeding, and the patient was discharged from the hospital with complete tube feeding on the 125th postoperative day. However, aspiration pneumonia returned. Neither pharyngeal stenosis nor tumor recurrence was found during hypopharyngoscopy under general anesthesia at 6 months after PLP. Videofluorography (Figure 2) revealed that laryngeal elevation was insufficient due to cervical vertebral osteophytes (white arrowheads). Just a small amount of contrast medium passed only through the left pyriform sinus to the esophagus (black arrowheads), and aspiration was profuse.

Figure 1. Pretreatment findings: (a) hypopharyngoscopy (b) contrast-enhanced soft tissue window images of computed tomography. (a) The tumor was localized to the left pyriform sinus (black arrowheads). (b) The tumor was seen in the left pyriform sinus as a contrast-enhanced lesion (circle). Osteophytes were developed on anterior cervical vertebrae (white arrowheads) and the esophagus was shifted to left (arrow). PC: postcricoid area, AP: apex of pyriform sinus.

Figure 2. Videofluorography examination after PLP. The laryngeal elevation was inhibited by cervical vertebral osteophytes (white arrowheads). A limited amount of contrast medium passed only through the left pyriform sinus to the esophagus (black arrowheads), and aspiration was profuse (arrow).
Computed tomography (Figure 3(a–c)) showed osteophytes of thoracic and lumbar vertebrae in addition to cervical vertebrae (arrowheads). The osteophytes of the fourth to the seventh cervical vertebrae (Figure 3(b), arrowheads) protruded above the cricoid cartilage lamina (arrow) and were thought to inhibit laryngeal elevation. Osteophytes were deemed due to DISH because they were observed on four continuous cervical vertebrae and intervertebral discs were nearly normal in height (Figure 3(a–c)). Based on these findings, it was concluded that dysphagia was caused by insufficient laryngeal elevation and esophageal compression by osteophytes caused by DISH. Aspiration prevention surgery was required due to repeated pneumonia and weight loss from 52.3 kg to 44.5 kg. The patient was eager to improve his swallowing function and accepted the loss of his voice.

Total laryngectomy and pharyngeal reconstruction with a pectoralis major muscle cutaneous flap were performed 7 months after PLP (Figure 4). The forearm flap and residual pharyngeal mucosa were preserved. Osteophytes of the cervical vertebrae were palpated as hard ridges during surgery. Pathological examination revealed no tumor recurrence. Pharyngography at the 12th postoperative day showed
that contrast medium could easily pass from the phar-
ynx to the esophagus (data not shown). The patient
was orally ingestible and did not require tube feeding.
He was discharged on the 34th postoperative day. At
two years after total laryngectomy, there was no
recurrence of dysphagia, pneumonia, or hypopharynge-
eal cancer.

Discussion
DISH is a disease in which remarkable ossification
lesions occur in the vertebrae, pelvis, and limb joints
[1]. DISH is diagnosed based on the following find-
ings: large, flowing osteophytes of four or more verte-
brae; intervertebral discs with normal or mild
decreases in height; and no presence of disorders of
intervertebral joints or sacroiliac joints [8]. Prevalence
of DISH is relatively high among elderly people, with
reported frequencies of 3.8% and 2.6% in men and
women, respectively, over the age of 40. Majority of
DISH cases are asymptomatic. Dysphagia may occur
in advanced cases due to compression of the pharynx
and the esophagus by the osteophytes, secondary
inflammation, and a decrease in the range of cervical
vertebral motion, but there are few cases that require
treatment [2].

PLP is a suitable treatment for early-stage hypo-
pharyngeal cancer that is amenable to larynx-preserv-
ing surgery [3]. There is a general consensus on the
indications for PLP to prevent complications and to
provide safe resection of the tumor. These are as fol-
 lows: (1) The primary tumor must originate from
either the pyriform sinus or the pharyngeal wall of
the hypopharynx. (2) The tumor must not extend to
either the apex of the pyriform sinus or the base of
tongue. (3) The bilateral vocal cords must be mobile.
(4) The tumor may extend to the supraglottic larynx,
but should not involve the transglottic larynx [7,9].
To prevent severe dysphagia and aspiration pneu-
monia, preservation of the bilateral superior and inferior
laryngeal nerve is necessary to retain laryngeal func-
tion [10]. We followed for all these criteria, and all
nerves related to swallowing function as above were
preserved. However, severe dysphagia and aspiration
pneumonia occurred after PLP. We suppose that it
was mostly due to insufficient laryngeal elevation and
esophageal compression by DISH, while prior surgery
and chemo-radiotherapy for head and neck region
might also influence on this complication. Kuo et al.
reported that two of 39 cases (5%) required additional
total laryngectomy due to aspiration after PLP [4]. Joo
et al. reported that seven (16%) cases required tube
feeding and five (12%) cases required tracheostomy
after PLP, among a total of 43 cases [5]. Based on
these reports, we suggest that dysphagia might not be
avoidable in all cases after PLP and the asymptomatic
DISH could be one of risks for post-operative severe
dysphagia and aspiration in PLP. In such a case, sim-
ultaneous performing of dysphagia surgeries including
laryngeal suspension and cricopharyngeal myotomy
should be considered [11,12].

In conclusion, asymptomatic DISH, which is a
common disorder in elderly patients, can cause severe
dysphagia and aspiration after PLP. Laryngeal suspen-
sion and cricopharyngeal myotomy should be consid-
ered for patients with DISH when conducting PLP,
even though they demonstrated no difficulties in swal-
lowing before treatment.

Disclosure statement
No potential conflict of interest was reported by the authors.

References
[1] Resnick D, Shaul SR, Robins JM. Diffuse idiopathic
skeletal hyperostosis (DISH): Forestier’s disease with
extraspinal manifestations. Radiology. 1975;115:
513–524.
[2] Akhtar S, O’Flynn PE, Kelly A, et al. The manage-
ment of dysphasia in skeletal hyperostosis.
J Laryngol Otol. 2000;114:154–157.
[3] National Comprehensive Cancer Network. NCCN
Clinical Practice Guidelines in Oncology in Head
and Neck Cancers version 1. 2018. [cited 2018 Apr
1]. Available from: https://www.nccn.org/professio-
nals/physician_gls/pdf/head-and-neck.pdf
[4] Kuo YL, Chang CF, Chang SY, et al. Partial laryngo-
pharyngectomy in the treatment of squamous cell
carcinoma of hypopharynx: analysis of the oncologic
results and laryngeal preservation rate. Acta
Otolaryngol. 2012;132:1342–1346.
[5] Joo YH, Cho KJ, Park JO, et al. Role of larynx-pres-
serving partial hypopharyngectomy with and without
postoperative radiotherapy for squamous cell carcino-
ma of the hypopharynx. Oral Oncol. 2012;48:
168–172.
[6] Plouin-Gaudon I, Lengé B, Desuter G, et al. Conser-
vation laryngeal surgery for selected pyriform
sinus cancer. Eur J Surg Oncol. 2004;30:1123–1130.
[7] Ogura JH, Marks JE, Freeman RB. Results of conser-
vation surgery for cancers of the supraglottis and
pyriform sinus. Laryngoscope. 1980;90:591–600.
[8] Resnick D, Niwajama G. Radiographic and pathol-
ogic features of spinal involvement in diffuse idiop-
athic skeletal hyperostosis (DISH). Radiology.
1976;119:568.
[9] Shah JP, Patel SG, Singh B. Partial laryngo-pharyn-
gectomy. In: Shah JP, editor. Head and neck surgery
and oncology. 4th ed. Philadelphia (PA): Elsevier;
2012. p. 333–335.
[10] Bailey BJ, Calhoun KH, Healy GB, et al. Partial and total pharyngectomy. In: Bailey BJ, editor. Atlas of head and neck surgery – otolaryngology. 2nd ed. Philadelphia (PA): Lippincott & Williams Wilkins; 2001. p. 266–267.

[11] Goode RL. Laryngeal suspension in head and neck surgery. Laryngoscope. 1976;86:349–355.

[12] Tiwari R, Karim AB, Greven AJ, et al. Total glossectomy with laryngeal preservation. Arch Otolaryngol Head Neck Surg. 1993;119:945–949.