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A primary appendiceal Burkitt lymphoma mimicking appendiceal abscess

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ABSTRACT

Interval appendectomy (IA) following nonoperative management with broad-spectrum antibiotics for complicated appendicitis with peri-appendiceal abscess is a one of the beneficial treatment options in children. However, during intervals, the protocol of follow-up blood examination and imaging studies including US/CT/MRI and the treatment policy for the recurrent cases, are not established. Herein, we presented a case which was diagnosed as primary appendiceal Burkitt lymphoma during the treatment policy of IA in a child, and discuss about unusual pitfall of IA policy.

1. Case report

Interval appendectomy (IA) following nonoperative management with broad-spectrum antibiotics for complicated appendicitis with peri-appendiceal abscess is a one of the beneficial treatment options in a high prevalence of inflammation in the appendix upon interval removal in children, and IA is recommended to perform at least 12 weeks from initial presentation [1].

However, during intervals, the protocol of follow-up blood examination and imaging studies including US/CT/MRI and the treatment policy for the recurrent cases, are not established.

Herein, we presented a case which was diagnosed as primary appendiceal Burkitt lymphoma during the treatment policy of IA in a child, and discuss about unusual pitfall of IA policy.

A 9-year-old girl was referred to previous hospital with a 2-day history of right abdominal pain, blood examination showed CRP and WBC account elevation (8.92mg/dl, 11360/μl, respectively), and abdominal ultrasonography and CT scan revealed a mass-forming appendix whose overall diameter was 27 mm (Fig. 1A), and non-operative treatment with PIPC/TAZ administration had started. Both abdominal pain and CRP elevation were improved, she was discharged on the 11th days and IA was scheduled on 12 weeks later. However, four weeks later from the discharge (Day 36), the abdominal pain and CRP elevation (5.89mg/dl) without WBC count elevation(5891/μl) were recurred, and US demonstrated overall diameter of appendiceal mass enlarged to 33mm (Fig. 1B). And she was readmitted previous hospital and the non-operative management with TAZ/PIPC administration was re-started to continue the IA policy. TAZ/PIPC administration was effective again and she was discharged 11th day and IA was re-scheduled for elective appendectomy on 12 weeks after her second discharge. On the outpatient regular examination at four weeks after her second discharge (Day 85), US revealed the appendiceal mass had grown to maximum diameter 48 mm (Fig. 1C), suggesting a neoplastic lesion although she had been asymptomatic. Blood examination showed CRP elevation (8.83mg/dl) without WBC elevation (3865//μl), and

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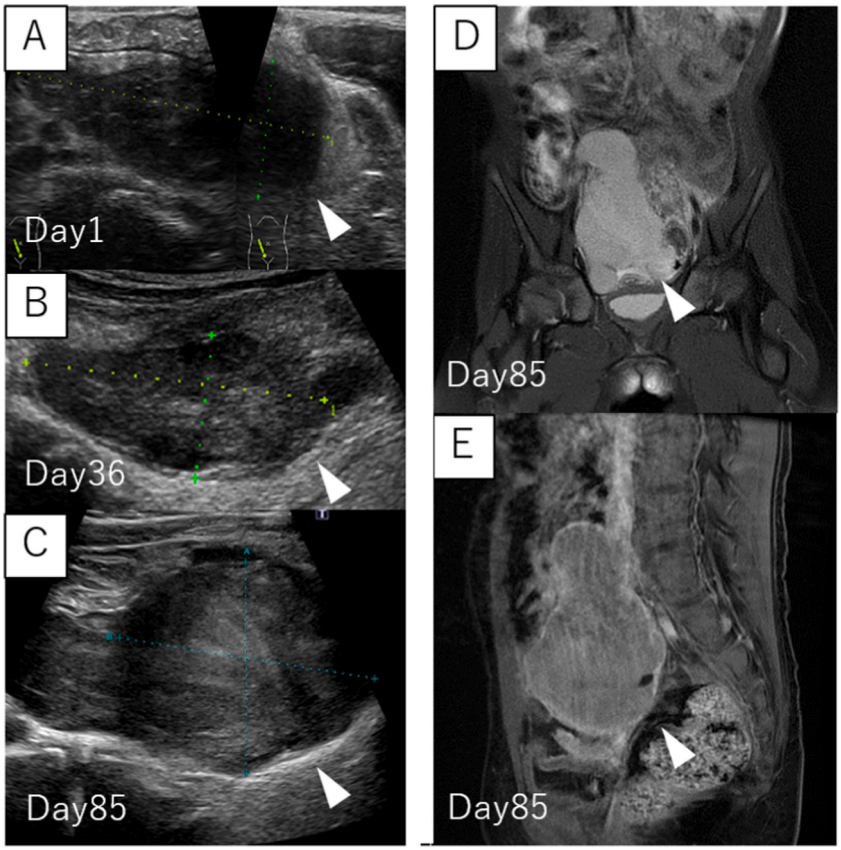

Fig. 1. (A,B,C) Ultrasonography demonstrated an appendiceal mass suggesting peri-appendiceal abscess (white arrow) with overall diameter 27 mm at day1, 33mm at day 36 and 48mm at day 85, respectively.
(D,E) MRI (T2WI) heterogenous mass (white arrow) occupying lower abdomen and pelvic space.

tumor makers including IL-2 receptor (1076U/ml) and NSE (30.9ng/ml) were elevated whereas CEA 0.7ng/ml and CA19-9 2.5U/ml were normal. CT and MRI of the abdomen and pelvis revealed a heterogeneous mass approximately 60x90 mm(Fig. 1D.E). From these findings, a primary appendiceal lymphoma was suspected and she was performed laparotomy. A whitish mass arising from the root of appendix was observed intraoperatively and it was considered a primary appendiceal tumor. The ovaries, fallopian tubes, and uterus were intact, but the tumor was firmly adherent to the sigmoid colon, upper rectum, and retroperitoneum. As the root of the appendix was intact, subtotal appendiceal tumor resection with the resection of the infiltration of the rectal wall were performed.

Histopathological examination showed the wall of the appendix is markedly thickened, the wall was preserved on the root side. Further immunostaining revealed CD3-, $CD20^{+}$, $CD10^{+}$, bcl2-, Ki67-positive cells in 99% of specimens, and the diagnosis of Burkitt lymphoma was made.

Postoperatively, the patient was diagnosed as stage III b and we performed.

6 courses of chemotherapy based on Japanese Pediatric Leukemia/Lymphoma Study Group (JPLSG) B-NHL03 protocol group 3. Two years have passed after the treatment, the patient is asymptomatic with no recurrence.

2. Discussion

In this case report, the patient presented clinical findings compatible with mass-forming appendicitis, which indicated IA policy without arising suspicion of another diagnosis. Since antibiotic treatment was so effective that clinical symptoms and CRP elevation were improved promptly during both two hospitalizations, in addition overall diameter of appendiceal mass had not changed significantly, diagnosis of appendiceal lymphoma was delayed.

Approximately 1% of appendectomies have an incidental finding of an appendiceal neoplasm, and manifestation of Burkitt's lymphoma as appendicitis or peri-appendiceal abscess has been reported [2].

Primary lymphoma of the appendix is characterized by diffuse swelling of the appendix and circumferential thickening of the wall while maintaining its morphology. Although there are no classical imaging features of appendiceal lymphoma, enlargement of appendix beyond 15mm in diameter on CT should be viewed with suspicion and a diameter above 25 mm should be even more concerning [3], in addition, abscess cavity generally shrinks with antibiotic treatment, therefore, our case may have been suspected as appendiceal lymphoma earlier.

We hope that our case highlights the importance of close observation of appendiceal mass with US focusing on the overall diameter, and the importance of suspicion of the neoplasm when the overall diameter of appendiceal mass does not decrease nevertheless other clinical symptoms improved during IA policy.

3. Conclusion

During IA policy, the close observation of appendiceal mass with the suspicion of the neoplasm is important in case the overall diameter of appendiceal mass does not decrease.

Disclosure

The authors declare no conflict of interest.

Author contribution

A.W., Y.O., M.N. and M.Y. conceptualized and designed the study, drafted the initial manuscript, and approved the final manuscript as submitted. N.Y. reviewed the manuscript and approved the final manuscript as submitted. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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The authors report no financial interests, relationships and affiliations relevant to the subject of the manuscript.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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