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Management of Pediatric Intussusception: Review of the Treatment Algorithm (Retrospective Study)

Pediatrik İnvajinasyon Yönetimi: Tedavi Algoritmasının Gözden Geçirilmesi (Retrospektif Çalışma)

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ABSTRACT Objective: In large series, the clinical features, management steps, and treatment results of intussusceptions have been studied. **Material and Methods:** Retrospective study including intussusception patients in a single center between 2008-2021. **Results:** There were 406 patients: 353 (86.9%) patients with ileocolic, 30 (7.4%) patients with small bowel, and 23 (5.7%) patients colocolic intussusception. In small bowel intussusceptions, 60% (18/30) of patients were treated surgically and in 40% (12/30) spontaneous reduction happened. Pathological lead points (PLPs) were found in 23.3% (7/30) of patients. In colocolic intussusception, the nonoperative reduction was used in 22/23 patients with a 90.9% success rate. The rate of PLPs was 4.3% (1/23). In ileocolic intussusception, spontaneous reduction happened in 18% (64/353) of cases. Nonoperative reduction was used as initial treatment in 272/353 patients with 79% (215/272) success rate and surgical management was used in 22.2% (78/353) of patients. The recurrence rate was 17.2% (61/353) with 85 attacks. Nonoperative reduction was used in 65/85 of attacks with 93% success rate. PLPs rate in different age groups was: 3.8% (0-2 years), 8% (2-5 years) and 17.4% (>5 years) (p value<0.001). In the patients who are >5 years old the frequency of Burkitt lymphoma was remarkable 10.8%. **Conclusion:** Although the first-line approach in small bowel intussusceptions is surgical exploration, the frequency of spontaneous reduction is high in cases with short segments (<4 cm). Nonoperative reduction should be the standard initial treatment for all age groups in ileocolic and colocolic intussusceptions. Management should be done carefully in patients who are >5 years old with ileocolic intussusceptions because of the high incidence of PLPs and Burkitt's lymphoma.

Keywords: Intussusception; acute abdomen; Burkitt lymphoma

ÖZET Amaç: Geniş hasta serisinde, pediatrik invajinasyonun klinik özellikleri, tedavi basamakları ve tedavi sonuçları incelenmiştir. **Gereç ve Yöntemler:** 2008-2021 yılları arasında tek merkezde tüm invajinasyon hastalarının dosyaları retrospektif olarak incelenmiştir. **Bulgular:** Dört yüz altı hasta çalışmaya dâhil edildi: 353 (%86,9) ileokolik, 30 (%7,4) ince bağırsak ve 23 (%5,7) kolokolik invajinasyon hastası mevcuttu. İnce bağırsak invajinasyonlarında hastaların %60'ı (18/30) cerrahi olarak tedavi edildi, %40'ında (12/30) spontan redüksiyon gerçekleşti. Hastaların %23,3'ünde (7/30) patolojik lead point (PLP) bulundu. Kolokolik invajinasyonda 22/23 hastada %90,9 başarı oranıyla nonoperatif redüksiyon uygulandı. PLP oranı %4,3 (1/23) idi. İleokolik tipte olguların %18'inde (64/353) spontan redüksiyon gerçekleşti, 272/353 hastada %79 (215/272) başarı oranıyla nonoperatif redüksiyon uygulandı, %22,2 (78/353) hastada cerrahi tedavi uygulandı. Nüks oranı %17,2 (61/353) (85 atak) idi. Atakların 65/85'inde %93 başarı oranıyla nonoperatif redüksiyon uygulandı. Farklı yaş gruplarında PLP oranı: %3,8 (0-2 yaş), %8 (2-5 yaş) ve %17,4 (>5 yaş) (p değeri <0,001) idi. Ülkemize özel son yaş grubunda (>5 yaş) Burkitt lenfoma sıklığı %10,8 dikkat çekiciydi. **Sonuç:** İnce bağırsak invajinasyonlarında temel yaklaşım cerrahi eksplorasyon olmakla beraber kısa segmentli (<4 cm) vakalarda spontan redüksiyon sıklığı yüksektir. Nonoperatif redüksiyon, ileokolik ve kolokolik invajinasyonlarda tüm yaş grupları için standart tedavi yaklaşımı olmalıdır. Beş yaş ve üzeri ileokolik invajinasyon hastalarında PLP ve Burkitt lenfoma insidansının yüksek olması nedeniyle tedavi dikkatli yapılmalıdır.

Anahtar Kelimeler: İntussusepsiyon; akut karın; Burkitt lenfoma

Intussusception is the most frequent cause of bowel obstruction in infants and toddlers, with an incidence of 15 to 300/100,000 children per year.[1] It was first described in 1674 by Paul Barbette of Amsterdam, defined by Treves in 1899, and operated on successfully in 1873 by John Hutchinson.[2] Intussusception may occur in any portion of the bowel, and according to its localization, it can be classified into

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small bowel intussusceptions (jejunojejunal or ileoileal), ileocolic intussusceptions and colocolic intussusceptions. Intussusception can occur at any age. Most affected patients are well-nourished, healthy infants, and approximately two-thirds are boys. The highest incidence occurs in infants between ages 4 and 9 months.[3]

Reduction of intussusception was traditionally performed using barium or other liquid contrast agents (hydrostatic enema), but can also be performed using air or carbon dioxide (pneumatic enema).[4]

In this study, in a large series of patients, the clinical features, results, frequency of recurrence, frequency of pathologic lead points (PLPs) in different age groups, and whether a change in management steps or behavioral differences is required according to intussusception localization and patient age have been studied.

MATERIAL AND METHODS

This study was carried out in pediatric surgery and radiology departments in concordance with international ethical standards and the World Health Organization's Helsinki Declaration. The study was approved by the Ege University Medical Research Ethical Committee (date: June 2, 2021; no: 21-5.1T/42) and informed consent was obtained from all the subjects.

MANAGEMENT PROTOCOL

According to our current protocol, patients presenting with classical clinical findings of intussusception are first evaluated by detailed ultrasonography (USG) and abdominal radiograph (AXR). A careful and detailed examination is performed for the possible PLPs. If findings compatible with intussusception are detected, according to its localization: If it is in the small intestine or if there is evidence of intestinal perforation on abdominal X-ray, preparation is made for surgical exploration. Before the operation, the patients are re-examined. Ultrasound is repeated in patients with clinically regressed symptoms. Surgery is canceled in spontaneously reduced patients.

In patients with ongoing symptoms surgical exploration and manual reduction of the invaginated segment is performed. If there is intestinal perforation, primary repair or resection-anastomosis is performed. In this study, we excluded the patients with transient small intestine intussusception, which is frequently seen in normal small intestine, physiologically.

In other types of intussusceptions, the initial management is nonoperative with hydrostatic (saline) reduction and it is applied under USG guidance in all age groups. After the reduction, the patient is kept under clinical follow-up for 12-24 hours and is discharged if the clinical complaints regress and the patient achieves spontaneous gas and stool discharge. If a history of longer than 36 hours is reported during the clinical presentation, the management is approached cautiously due to the risk of intestinal ischemia. If the patient's general condition is unstable, if laboratory and physical examination findings are supporting clinical signs of peritonitis, or if evidence of intestinal perforation is observed on an abdominal X-ray, the patient is prepared for surgical exploration. In cases of recurrent intussusception, in patients with non-small bowel origin and without signs of peritonitis, the first option is hydrostatic reduction under USG guidance, and post-reduction follow-up is longer for additional laboratory and radiological examinations due to the high probability of PLPs and recurrence.

STUDY DESIGN

This study retrospectively evaluated the data of 406 patients presenting with intussusception admitted to our clinic from June 1, 2008 to May 31, 2021. The clinical characteristics of the patients (age, gender, presentation complaint, complaint period, the history of new viral infection, the month of presentation, the type and site of intussusception, and the length of invaginated segment), diagnostic methods, management procedures, outcomes, analysis of the incidence of PLPs in different age groups, analysis of recurrence after initial treatment and management of recurrent patients were included in this study.

The patients were divided into 3 groups according to the type of intussusception (small bowel intus-

susceptions, large bowel intussusceptions, and ileocolic intussusceptions). And every group was divided into 3 sub-group according to the patient's age (0-24 months, 25-60 months, and>60 months).

STATISTICAL ANALYSIS

For the importance control of groups, a cross table was created on the categorical data and a chi-square analysis was performed, with a p value <0.05 considered significant. For numerical variables, a t-test will be used for those with normal distribution, and Mann-Whitney test analysis will be used for others.

RESULTS

DEMOGRAPHIC DATA

During the study period, with clinical features and ultrasonographic examination, intussusception was prediagnosed in 406 patients. There were 267 males and 139 females with a male-to-female ratio of 1.9: 1. The average age of diagnosis was 32±29.15 months (1-180 months). In half of the patients, the first intussusception attack occurred under 2 years of age (51%). The peak age of occurrence was between the age of 6 and 12 months. Whereas it decreased dramatically after 5 years of age (14%).

Most of the (89.8%) patients presented with abdominal pain. Nausea and vomiting were seen in 50.3% of the patients. The strawberry jelly blood in stool was seen in only 20%. Some of the patients presented with the inability to defecate 27.6% and on the other hand some of them presented with diarrhea.

In this study, we found that intussusception attacks may occur at any time of the year with minimal increase in the summer (Figure 1).

FIRST GROUP: SMALL BOWEL INTUSSUSCEPTIONS

In this study, we evaluated the data of 30 patients presenting with small bowel intussusception (28 patients with ileoileal and 2 patients with jejunojejunal intussusception). There were 21 (70%) males and 9 (30%) females.

The distribution of patients by age was different from other intussusception types. Most of the patients 73.4% were older than 2 years old.

Eighteen patients (18/30) underwent abdominal exploration. The surgical manual reduction was applied in 40% (12/30) of patients, and bowel resection-anastomosis was required in 20% (6/30) of patients.

The spontaneous reduction occurred in 40% (12/30) of patients. These patients were all symptomatic, all patients were admitted with abdominal pain, 6 patients with vomiting, and 2 with bloody stools.

The rate of PLPs was high in this type 23.3% (7/30). The most frequently seen PLP was Meckel's diverticulum (2/30) and hamartomatous polyps (2/30). We have also reported cases of gastric heterotopia, Henoch-Shöenlein purpura, and polyarteritis nodosa.

We found that the rate of PLPs was so low (1/12) in the spontaneous reduction group when compared with patients who required operation (6/18). We have also noticed that the length of invagination was less

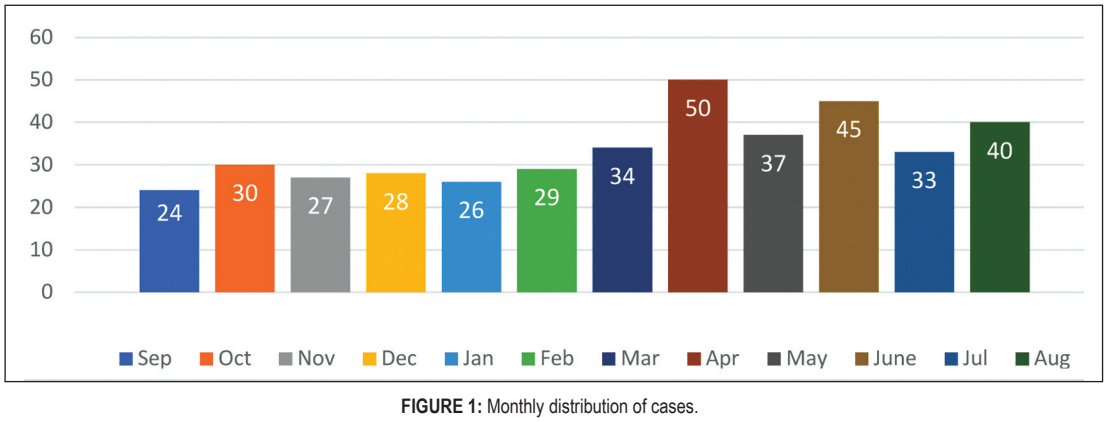

FIGURE 1: Monthly distribution of cases.

than 4 cm (ranging from 2.5 cm to 8 cm) in the majority of the spontaneous reduction group (10/12).

The rate of recurrence in small bowel intussusceptions was 13.3% (4/30). In 2 patients bowel resection-anastomosis was required, in 1 patient spontaneous reduction occurred and in the last patient the recurrence occurred with a different type of intussusception (ileocolic intussusception) and this patient was treated with hydrostatic reduction. PLP -which was gastric heterotopia- was detected in only one of these 4 patients.

LARGE BOWEL INTUSSUSCEPTIONS

There were 23 patients with colocolic intussusception. 61% (14/23) of them were male and 39% (9/23) were female. Most of them (60%) were under 2 years old.

In most of the patients (22/23) the initial management was nonoperative management with hydrostatic reduction and it was successful in 90.9% (20/22) of them.

The surgical management was applied in 3/22 patients. One patient had symptoms of peritonitis therefore abdominal exploration was performed initially. Two patients who had failed hydrostatic reduction underwent laparoscopic reduction.

The rate of recurrence was 21.3% (5/23). Four of them were in the hydrostatic reduction group and 1 was in the surgical reduction group.

The initial management after recurrence was nonoperative in 4 patients. It was successful in three and unsuccessful in one patient who was treated surgically.

The rate of PLPs was 4.3% (1/23); It was detected in one patient after recurrence. The PLP was a hamartomatous polyp in this patient.

ILEOCOLIC INTUSSUSCEPTIONS

In this study, we evaluated the data of 353 patients presenting with this type of intussusception. There were 232 (66%) males and 121 (34%) females. 52% (184/353) of patients were under 2 years old, 35% (123/353) were between 2-5 years old and 13% (46/353) were older than 5 years old.

In this group, the initial management was nonoperative management with hydrostatic reduction and it was performed in 77% (272/353) of patients with a 79% (215/272) success rate. In the unsuccessful nonoperative management group (57/272) we found that PLPs were detected in 13 patients. In 23 patients accompanying lymphadenopathy (LAP) was detected. The length of the invaginated segment in 23 patients was longer than 6 cm. In 6 patients the appendix was located in the invaginated segment, and in 13 patients complaint duration was longer than 3 days.

The surgical reduction was performed as initial treatment in only 5% (17/353) of patients. Otherwise, 16.1% (57/353) of patients required surgical reduction due to failed hydrostatic reduction. Despite the successful hydrostatic reduction, 4 (1.1%) patients needed abdominal exploration because of PLPs.

In total, surgical reduction was required in 22.2% (78/353) of patients with ileocolic intussusception.

The spontaneous reduction occurred in 18% (64/353) of cases. The invaginated segment length was <4 cm in 41/64 patients, >4 cm in 7/64 patients and it was unmeasured in 16/64 patients. The recurrence rate in this group was 11% (7/64). No PLP was detected in any of the patients. Five patients were treated successfully with hydrostatic reduction, in 1 patient laparoscopic reduction was applied, and spontaneous reduction occurred in 1 patient.

The general rate of PLPs in ileocolic intussusception was 7% (25/353). It was 5.6% (20/353) in the patients presented with the first attack and 8.2% (5/61) in recurrent patients (p value <0.001) (Table 1). In different age groups, it was: 3.8% (7/184) (in patients of 0-2 years old), 8% (10/123) (in patients of 2-5 years old), and 17.4% (8/146) (in patients >5 years old patients) (p value <0.001). In this group, the frequency of Burkitt lymphoma was remarkable, in the first attack it was 1.4% (5/353), and in the recurrent patients, it was 4.9% (3/61) (p value <0.001). In different age groups, it was 0% (0-2 years old patients), 2.4% (3/123) (2-5 years old patients), and 10.8% (5/46) (>5 years old patients) (Table 2).

The recurrence rate was 17.2% (61/353) with 85 attacks. 12.9% (11/85) of the attacks were sponta-

TABLE 1: PLPs rate in the first attack of intussusception vs recurrent intussusception.

PLPs detected in the first attack	Meckel's diverticulum	2.8% (10/353)
	Burkitt's lymphoma	**1.4%** (5/353)
	Hamartomatous polyp	0.8% (3/353)
	Celiac disease	0.3% (1/353)
	Henoch-Shöenlein purpura	0.3% (1/353)
	Total	**5.6%** (20/353)
PLPs detected in recurrent attacks	Meckel's diverticulum	3.3% (2/61)
	Burkitt's lymphoma	**4.9%** (3/61)
	Total	**8.2% (5/61)**

PLPs: Pathologic lead points.

TABLE 2: The distribution of PLPs rate by the age of patients.

Patients with ages 0-24 months old	Meckel's diverticulum	2.2% (4/184)
	Hamartomatous polyp	1.1% (2/184)
	Celiac disease	0.5% (1/184)
	Total	**3.8% (7/184)**
Patients with ages 25-60 months old	Meckel's diverticulum	5.6% (7/123)
	Burkitt lymphoma	**2.4%** (3/123)
	Total	**8% (10/123)**
Patients with the age of >60 months old	Burkitt lymphoma	**10.8%** (5/46)
	Meckel's diverticulum	2.2% (1/46)
	Hamartomatous polyp	2.2% (1/46)
	Henoch-Shöenlein purpura	2.2% (1/46)
	Total	**17.4% (8/46)**

PLPs: Pathologic lead points.

neously reduced, nonoperative reduction was applied in 76.5% (65/85) of attacks with 93% (61/65) success rate while other cases were treated surgically.

A significant portion of the recurrences (39.3% in the ileocolic type, 20% in the colocolic type, and 25% in the small bowel intussusceptions were detected) in the first 24 hours after reduction.

The average length of hospital stay after nonoperative reduction was 1.8 days while it was 3.34 days after surgical management.

In total, the hydrostatic reduction was applied safely with no complication in different age groups 363 times.

DISCUSSION

In 1876, Hirschsprung made the first attempt at hydrostatic reduction of intussusception using water.[5] Kim and his colleagues described the first successful sonographic-guided hydrostatic reduction in 1982.[6]

Hydrostatic reduction with barium under fluoroscopic guidance was historically used. More recently, children's hospitals have transitioned to air or water-soluble isotonic contrast because of the potential hazard of barium peritonitis in patients with intestinal perforation.[7]

Despite the high success rate of hydrostatic reduction, the application of this method is still controversial in some age groups due to the increased risk of PLPs.

SMALL BOWEL INTUSSUSCEPTIONS

Kornecki et al. recommended that the management of all these patients can depend on the clinical findings.[8] When a short-segment intussusception is found, which is thought to be in the small bowel, with no recognizable lead point, particularly in an asymptomatic patient, conservative observation is suggested.

According to Kim study, typical USG findings of the transient small bowel intussusception included small size without wall swelling, short segment (1.8±0.5 cm), preserved wall motion, and absence of the lead point. Conservative management with USG monitoring rather than an immediate operation is recommended for those patients.[9]

In the Güney et al. study, no spontaneously reduced intussusceptum was greater than 2.3 cm in length, while all intussusceptums that required surgery were at least 4 cm.[10]

In another study, spontaneous reduction frequently developed in cases of intussusception with invaginated segment less than 3 cm.[11]

Currently, even though the first-line approach is still surgical exploration, we noticed a high frequency of spontaneous reduction in short-segment intussusceptions (<4 cm) in our study. We recommend that more prospective studies can be done to determine if only clinical follow-up is enough in patients who are hemodynamically stable and don't have symptoms of peritonitis.

ILEOCOLIC AND COLOCOLIC INTUSSUSCEPTIONS

Success rates of hydrostatic reduction reported in the literature vary between 51.5% to 96.5%.[12,13]

In our study, the success rate of hydrostatic reduction in colocolic intussusception was similar to reported rates, but our success rate in ileocolic intussusception was relatively lower than the literature.[13-15]

The failure of hydrostatic reduction in some patients in the current study was considered to be multifactorial. The presence of PLPs, LAP, appendiceal intussusception, >6 cm invaginated segment length, and >3 days complaint duration were thought to be risk factors for failing the hydrostatic reduction.

In the Xiaolong et al. study an age under 1 year old, duration of symptoms more than or equal to 48th, rectal bleeding, constipation, palpable abdominal mass, and location of mass (left over right side) were risk factors for failure of hydrostatic reduction of intussusception.[16] The duration of symptoms as a risk factor was found to be compatible with our study but other factors were not compatible.

THE RECURRENCE OF INTUSSUSCEPTION

In different literature, the recurrence rate varies between 8% to 20%.[15,17]

In our study, the recurrence rate was 13.3% in small bowel intussusceptions, 21.3% in colocolic intussusceptions, and 17.2% in ileocolic intussusceptions.

The hydrostatic reduction was applied successfully in approximately 93% of recurrent attacks of ileocolic intussusceptions without any complications regardless of age.

In the different types of intussusceptions, a significant portion of the recurrences was detected in the first 24 hours after reduction (40% in ileocolic type, 20% in colocolic type, and 25% small bowel type) so we recommend that these patients must be followed up at least 24 hours after reduction. And if the patient will be discharged early from the hospital, it can be followed up with phone calls.[18]

The probability of recurrence was 22.9% after 2nd episode, 42% after 3rd episode, and 100% after the 4th episode of intussusception in our study similar to the Hsu et al. study.[19] So, the surgical intervention should be considered in the 4th episode of intussusception.

THE PATHOLOGICAL LEADS POINTS OF INTUSSUSCEPTION

The general rate of PLPs in colocolic intussusception was 4.3% (1/23). In this type, no PLPs were detected after the first attack. They were detected after recurrence with a rate of 20% (1/5), so the necessity of surgery after the first reduction should be kept in mind.

In the ileocolic intussusceptions, the frequency of Burkitt lymphoma was higher after recurrence and in patients >5 years old. Therefore, in this group management should be done carefully. We recommend close follow-up and detailed investigation with USG, computed tomography, or magnetic resonance imaging regardless of the successful hydrostatic reduction in these patients.

Below in Table 3, a comparison of different studies about intussusception is presented (since only ileocolic intussusception was investigated in this literature, only the ileocolic part of our study was included in the comparison).

LIMITATION

A limitation of the present study was the retrospective nature of the data. All procedures were conducted at a single institution, and the number of patients especially in small intestine intussusception was limited, allowing for descriptive, rather than comparative, analyses. USG was not performed by a single radiologist and mostly, the diameter of the intussuscepted segment was not specified.

CONCLUSION

Even though the first-line approach in small bowel intussusceptions is surgical exploration, the frequency of spontaneous reduction is high in cases with short segments (<4 cm). Nonoperative reduction

TABLE 3: Summary of the literature review.

Study	Number of patients	PLPs rate in patients <5 y	PLPs rate in patients >5 y	Malignancy rate in patients <5 y	Malignancy rate in patients >5 y	Nonoperative treatment success rate	Recurrence rate	PLPs rate after recurrence
Current study	353	5.5%	17.4%	1%	10.8%	79%	17.3%	8.2%
Hsu et al.[19]	686	General PLPs rate: 2.2%		-	-	83%	-	-
Banapour et al.[12]	153	4.9%	40%	-	-	51.5%	-	-
Rubinstein et al.[20]	124	General PLPs rate: 0.8%		-	-	75%	-	-
Savoie et al.[21]	7.412	-	-	<3 y: 5%	>3 y: 6%	%82	-	-
Niramis et al.[17]	1.340	-	-	-	-	-	8%	9.3%
Fisher et al.[14]	666	3%	29%	-	-	91.6%	15%	-
Cho et al.[13]	491	General PLPs rate: 1%		-	-	96.5%	13.8%	-
Chen et al.[15]	5.778	1%	8.1%	General malignancy rate: 0.15%		93.5%	20%	2.2%

PLPs: Pathologic lead points.

should be the standard initial treatment for all age groups in ileocolic and colocolic intussusceptions. Management should be done carefully in patients who are >5 years old with ileocolic intussusceptions because of the high incidence of PLPs and Burkitt's lymphoma.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Ahmet Çelik;* ***Design:*** *Ahmet Çelik, Samir Hasan;* ***Control/Supervision:*** *Ahmet Çelik;* ***Data Collection and/or Processing:*** *Samir Hasan, Ülgen Çeltik;* ***Analysis and/or Interpretation:*** *Ahmet Çelik, Geylani Özkök, Orkan Ergün, Emre Divarcı;* ***Literature Review:*** *Ahmet Çelik, Samir Hasan;* ***Writing the Article:*** *Samir Hasan, Ülgen Çeltik;* ***Critical Review:*** *Ahmet Çelik, Geylani Özkök, Orkan Ergün, Emre Divarcı.*

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